frontiers | Research Topics

Pathway of care and gaps in services for children and adults with autism spectrum disorder

Edited by
Maria Luisa Scattoni, Andy Shih and Diana Schendel

Published in
Frontiers in Psychiatry

ISSN 1664-8714
ISBN 978-2-8325-2266-0
DOI 10.3389/978-2-8325-2266-0

About Frontiers

Frontiers is more than just an open access publisher of scholarly articles: it is a pioneering approach to the world of academia, radically improving the way scholarly research is managed. The grand vision of Frontiers is a world where all people have an equal opportunity to seek, share and generate knowledge. Frontiers provides immediate and permanent online open access to all its publications, but this alone is not enough to realize our grand goals.

Frontiers journal series

The Frontiers journal series is a multi-tier and interdisciplinary set of open-access, online journals, promising a paradigm shift from the current review, selection and dissemination processes in academic publishing. All Frontiers journals are driven by researchers for researchers; therefore, they constitute a service to the scholarly community. At the same time, the *Frontiers journal series* operates on a revolutionary invention, the tiered publishing system, initially addressing specific communities of scholars, and gradually climbing up to broader public understanding, thus serving the interests of the lay society, too.

Dedication to quality

Each Frontiers article is a landmark of the highest quality, thanks to genuinely collaborative interactions between authors and review editors, who include some of the world's best academicians. Research must be certified by peers before entering a stream of knowledge that may eventually reach the public - and shape society; therefore, Frontiers only applies the most rigorous and unbiased reviews. Frontiers revolutionizes research publishing by freely delivering the most outstanding research, evaluated with no bias from both the academic and social point of view. By applying the most advanced information technologies, Frontiers is catapulting scholarly publishing into a new generation.

What are Frontiers Research Topics?

Frontiers Research Topics are very popular trademarks of the *Frontiers journals series*: they are collections of at least ten articles, all centered on a particular subject. With their unique mix of varied contributions from Original Research to Review Articles, Frontiers Research Topics unify the most influential researchers, the latest key findings and historical advances in a hot research area.

Find out more on how to host your own Frontiers Research Topic or contribute to one as an author by contacting the Frontiers editorial office: frontiersin.org/about/contact

Pathway of care and gaps in services for children and adults with autism spectrum disorder

Topic editors
Maria Luisa Scattoni — National Institute of Health (ISS), Italy
Andy Shih — Autism Speaks, United States
Diana Schendel — Drexel University, United States

Citation
Scattoni, M. L., Shih, A., Schendel, D., eds. (2023). *Pathway of care and gaps in services for children and adults with autism spectrum disorder*. Lausanne: Frontiers Media SA. doi: 10.3389/978-2-8325-2266-0

Table of contents

frontiers | Frontiers in Psychiatry

TYPE Editorial
PUBLISHED 13 April 2023
DOI 10.3389/fpsyt.2023.1193166

Editorial: Pathway of care and gaps in services for children and adults with autism spectrum disorder

Maria Luisa Scattoni[1]*, Andy Shih[2] and Diana Schendel[3,4,5]

[1]Research Coordination and Support Service, Istituto Superiore di Sanità, Rome, Italy, [2]Autism Speaks, Inc., New York, NY, United States, [3]Department of Public Health, Aarhus University, Aarhus, Denmark, [4]Lundbeck Foundation Initiative for Integrative Psychiatric Research, iPSYCH, Aarhus, Denmark, [5]Department of Economics and Business, National Centre for Register-Based Research, Aarhus University, Aarhus, Denmark

KEYWORDS

care pathways, services, care coordination, autism spectrum disorder, children, adults

Check for updates

OPEN ACCESS

EDITED AND REVIEWED BY
Antonio M. Persico,
University of Modena and Reggio Emilia, Italy

*CORRESPONDENCE
Maria Luisa Scattoni
✉ marialuisa.scattoni@iss.it

SPECIALTY SECTION
This article was submitted to
Autism,
a section of the journal
Frontiers in Psychiatry

RECEIVED 24 March 2023
ACCEPTED 28 March 2023
PUBLISHED 13 April 2023

CITATION
Scattoni ML, Shih A and Schendel D (2023) Editorial: Pathway of care and gaps in services for children and adults with autism spectrum disorder. *Front. Psychiatry* 14:1193166.
doi: 10.3389/fpsyt.2023.1193166

Editorial on the Research Topic
Pathway of care and gaps in services for children and adults with autism spectrum disorder

This Research Topic entitled "*Pathway of care and gaps in services for children and adults with autism spectrum disorder*" aimed to provide evidence, data, or strategies for the definition and implementation of appropriate care pathways and services addressing the complex and heterogeneous nature of autism and providing high-quality, patient-centered medical care for autistic people. The pathway of care for autistic individuals is complex and should involve multiple systems, including healthcare, education, social services, and community support. The pathway begins with the diagnosis of autism, which can occur at any age but is typically diagnosed in early childhood. After the diagnosis, autistic individuals generally require ongoing support and care from multiple healthcare providers, including pediatricians, psychiatrists, psychologists, and other specialists. Their care should be personalized and, also, adapted to the cultural context. Access to early intervention programs is critical for improving outcomes for autistic children. As autistic individuals transition into adulthood, they may require additional support and services to help them achieve independence and integrate into the community. This Research Topic examines the journey of autistic individuals through the healthcare system and the available services and support. It also focuses on identifying gaps in the existing services and interventions for autistic individuals and how to address them to improve the quality of life for autistic people and their families. The specific barriers to accessing services in Africa, China, and Europe have been studied and analyzed by several authors on this Research Topic, which consists of 11 original articles, one systematic review, one review, one policy and practice review, and one mini-review, all published in 2022.

1. Low middle-income countries and minorities: diagnostic and intervention services

The definition, organization, and implementation of multidisciplinary care and effective coordination between different health/social care providers and services dedicated to children, adolescents, and autistic adults represent a real challenge, especially for low and middle-income countries. Pillay et al. examined providers' perspectives and offered solutions to meet the educational needs of autistic children by analyzing the data from semi-structured interviews in South Africa. The authors suggested learning from international best practices, developing long-term and integrated policies, including users and carers in the decisions, planning specific training programs for educators, supporting children, and enhancing the early diagnostic process (Pillay et al.). Liu et al. explored the intervention status and burden of autistic children in Guizhou, a region in the south of China, through a cross-sectional survey completed by 231 families with autistic children. The participants referred to a lack of intervention resources and the high psychological and economic burden of autism as the major challenge (Liu et al.). Borokova et al. performed a systematic investigation of the needs, access to services, and priorities of families of children with developmental disorders in Bulgaria using an online family survey; in the survey, 195 parents of autistic children and 73 parents of children with neurodevelopmental disorders (NDD) reported a lack of resources in services for diagnosis, treatment, and assistance. Autistic children needed different pathways compared to children with NDD and parents focused on the necessity of raising awareness of autism and protecting their children's rights (Borokova et al.). In their scoping review, Beauchamps et al. identified three levels of barriers to accessing services for minority-language speakers' families with children with NDD: (a) systems level, e.g., lack of high-quality treatment and training for healthcare professionals, difficulties accessing interpreters, and lack of available information in minority languages; (b) provider level, e.g., personal characteristics of healthcare practitioners or personal erroneous beliefs regarding language development; and (c) family experiences level, e.g., feelings of distrust in providers or feelings of stigma. Possible solutions proposed by the authors were: the development and uptake of policies and guidelines, practitioner's training, referral pathways for specialized services, access to appropriate tools and resources, and partnership with carers. Antony et al. showed, in their policy practice review, that the policy practices should support the conceptualization of autism in indigenous populations and their interaction with the Canadian health system and justice system. In addition, appropriate care strategies for this population should facilitate the coordination between health and social services and provide culturally appropriate multidisciplinary care (Antony et al.). Montenegro et al. conducted a study in Argentina stating that attention to the needs of autistic children has increased compared to a few years ago. For example, from 2015 to 2020, carers reported that the age of diagnosis of autistic children had decreased. However, some family members needed to leave their job to take care of their children. They also perceived that their rights should be protected and advocated for (Montenegro et al.). Zhang et al. described the use of a screening tool that combined the Modified Checklist for Autism in Toddlers, Revised with Follow-up (M-CHAT-R/F) and the Binomial Observation Test (BOT) in a resource-limited and highly populated Chinese context to improve early diagnosis in the routine 18- and 24-month of age visits. The study, conducted on a sample of 11,190 toddlers, showed that the diagnostic rate of autism through community screening was 0.32% (95% CI: 0.23–0.45%); the mean age at diagnosis for the children was 23.1 ± 4.55 months which was 20 months earlier than the autistic children not screened in the community screening program (Zhang et al.).

2. Autistic youths and adults: mapping of services and users' experiences

In 2017, the ASD in the European Union (ASDEU) conducted a survey for autistic adults, carers, and professionals exploring the services and practices needs of autistic adults (Micai et al.). The study reported the top choices by autistic adults, carers, and professionals for services best suiting their current needs as residential services, e.g., "help in own home" and "fulltime residential facility"; employment services: "job mentors" and "sheltered employment"; education services: "support in regular education setting"; financial services: financial support in lieu of employment or supplementing employment earnings for carers of highly independent adults and professionals; social services: "behavior training" and "life skills training." The knowledge of good local service models that work well for autistic adults was generally low across all service areas (Micai et al.). Knowing where to find a specialized and dedicated service for autism is often one of the main barriers for adults and their carers. McCormick et al. described the "4-H," a national inclusive program for life skills and transition for autistic youths. The authors conducted focus groups involving 20 educators that expressed the need to enhance training opportunities and resources.

3. The World Health Organization caregivers skills training: effectiveness, feasibility, and telemedicine

The World Health Organization Caregivers Skills Training (WHO CST) is an evidence-based treatment for families with autistic children. In this Research Topic, some studies focused on the effectiveness of the training and its application *via* telemedicine. Telemedicine improves access to services. Pacione et al. conducted a brief review of the application of the WHO CST for autistic children *via* telemedicine. Results showed a positive effect of the treatment in terms of feasibility, acceptability, and effectiveness. Montiel-Nava et al. tested the WHO CST in a rural community in Missouri, US, through an online group format for families with autistic children. The WHO CST was feasible and enhanced access to treatment and parental sense of competence. In addition, children showed fewer atypical behaviors and improvement in communication skills after the training. Finally, Lau et al. conducted a randomized comparison of the control waiting list and WHO CST (*via* eLearning asynchronous, videoconferencing with online coaching, and in-person hybrid modalities) in 34 Chinese dyads of parents and children with autism or other NDD. They showed that the WHO CST synchronous interaction, both online and in person, was effective in terms of children's clinical improvement of communicative skills (Lau et al.). An effectiveness and feasibility study of the WHO CST delivered and adapted in Taiwan revealed that this approach is positive in terms of the increase in knowledge and confidence of parents, and the reduction of the severity of autistic symptoms in children, with a maintenance effect, after 3 months (Seng et al.). Finally, Glumbic et al. showed the significant improvement in speech, language, and communication of the WHO CST program on children with developmental disabilities and evaluated the process of implementation, cultural appropriateness, and parental opinions on the program in Serbia.

4. Training of non-specialist personnel: promising resources and challenges

A scoping review by Naithani, Goldie, et al. on intervention approaches for families with autistic children in low and middle-income countries showed preliminary fidelity, acceptability, and effectiveness of delivering intervention elements and techniques within parent-mediated programs by non-specialist personnel (i.e., bachelor-level graduates with no prior exposure to child development or autism) delivered at home and in clinical settings, with a supervision team (Naithani, Goldie, et al.). However, in another study by the same authors, the training of non-specialist practitioners reported mixed results (Naithani, Sangwan, et al.). A task-sharing approach to support the delivery of parent-mediated treatment for autism in South Asia (COMPASS) tested in a group of non-specialist Indian workers by video training did not appear to be an effective method to ensure the motivation of participants (Naithani, Sangwan, et al.).

5. Conclusions

In summary, despite the availability of many services and interventions for autistic individuals, there are still significant gaps in care. These gaps include disparities in access to diagnosis and care, inadequate training for healthcare providers, and limited funding for research and services. One significant gap in care for autistic individuals is the lack of access to early diagnosis and intervention services, particularly in underserved communities. Many families face long waitlists and limited availability of specialized services, leading to delays in accessing critical care. Another significant gap in care for autistic individuals is the lack of training for healthcare providers in autism diagnosis and management. Many healthcare providers report feeling unprepared to provide care for autistic individuals and lack knowledge of evidence-based interventions. Identifying and addressing gaps in care is essential for improving outcomes for autistic individuals and reducing disparities in access to care. Further research is needed to understand the best practices for delivering care and improving outcomes for autistic individuals.

Author contributions

MS wrote the first draft of the manuscript. All authors contributed to the conception of the Editorial. All authors contributed to the manuscript revision, read, and approved the submitted version.

Acknowledgments

We would like to acknowledge Letizia Gila, Martina Micai, Angela Caruso, and Francesca Fulceri for their great support in the management of the Research Topic.

Conflict of interest

AS was employed by Autism Speaks, Inc.

The remaining authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

frontiers | Frontiers in Psychiatry

ORIGINAL RESEARCH
published: 26 April 2022
doi: 10.3389/fpsyt.2022.879625

Screening for Autism Spectrum Disorder in Toddlers During the 18- and 24-Month Well-Child Visits

Ying Zhang[1†], Zhaoe Zhou[2†], Qiong Xu[1], Huiping Li[1], Yujing Lv[1], Guowei Zhu[2], Ping Dong[1], Dongyun Li[1], Yi Wang[1], Xinrui Tang[1] and Xiu Xu[1]*

[1] Department of Child Healthcare, Children's Hospital of Fudan University, Shanghai, China, [2] Department of Child Healthcare, Xuhui Maternal and Child Healthcare Hospital, Shanghai, China

Objective: Early screening contributes to the early detection of children with autism spectrum disorder (ASD). We conducted a longitudinal ASD screening study in a large community setting. The study was designed to investigate the diagnostic rate of ASD screening and determine the effectiveness of ASD screening model in a community-based sample.

Methods: We enrolled children who attended 18- and 24-month well-child care visits in Shanghai Xuhui District. Modified Checklist for Autism in Toddlers, Revised with Follow-up (M-CHAT-R/F) and Binomial Observation Test (BOT) were selected as screening instruments. Screen-positive children were referred to a tertiary diagnostic center for comprehensive ASD diagnostic evaluation. Screen-negative children received well-child checkups and follow-up every 3–6 months until age three and were referred if they were suspected of having ASD.

Results: A total of 11,190 toddlers were screened, and 36 screen-positive toddlers were diagnosed with ASD. The mean age at diagnosis for these children was 23.1 ± 4.55 months, diagnosed 20 months earlier than ASD children not screened. The diagnostic rate of ASD was 0.32% (95% CI: 0.23–0.45%) in this community-based sample. In addition, 12 screen-negative children were diagnosed with ASD during subsequent well-child visit and follow-up. The average diagnostic rate of ASD rose to 0.43% (95% CI: 0.32–0.57%) when toddlers were followed up to 3 years old. The positive predictive values (PPVs) of M-CHAT-R/F, M-CHAT-R high risk, and BOT for ASD were 0.31, 0.43, and 0.38 respectively.

Conclusion: Our findings provide reliable data for estimating the rate of ASD detection and identifying the validity of community-based screening model. M-CHAT-R/F combined with BOT can be an effective tool for early detection of ASD. This community-based screening model is worth replicating.

Keywords: autism spectrum disorder, early screening, M-CHAT-R/F, BOT, community-based

OPEN ACCESS

Edited by:
Andy Shih,
Autism Speaks, United States

Reviewed by:
Marc Woodbury-Smith,
Newcastle University, United Kingdom
Francesca Fulceri,
National Institute of Health (ISS), Italy

****Correspondence:***
Xiu Xu
xuxiu@fudan.edu.cn

†These authors have contributed equally to this work and share first authorship

Specialty section:
This article was submitted to Autism, a section of the journal Frontiers in Psychiatry

Received: *19 February 2022*
Accepted: *31 March 2022*
Published: *26 April 2022*

Citation:
Zhang Y, Zhou Z, Xu Q, Li H, Lv Y, Zhu G, Dong P, Li D, Wang Y, Tang X and Xu X (2022) Screening for Autism Spectrum Disorder in Toddlers During the 18- and 24-Month Well-Child Visits. Front. Psychiatry 13:879625. doi: 10.3389/fpsyt.2022.879625

INTRODUCTION

Autism spectrum disorder (ASD) is a group of heterogeneous neurodevelopmental disorders, which are characterized by deficits in social communication and interaction, and restricted and repetitive patterns of behaviors, interests, or activities (1). The current prevalence is assessed to be about 1.5% in developed countries and 1% in worldwide (2, 3). In China, the prevalence of ASD has been reported ranged from a low of 0.2% to as high as 1% (4, 5). Zhou et al. reported a prevalence of 0.7% among 6- to 12-year-old children in 2019 (5), which is the largest epidemiological study in China to date. ASD tends to be accompanied by a kind of serious neuropsychiatric disorder in adulthood if there is no effective intervention in time, which might become a heavy burden to an individual, a family, or even the whole society (6–8). Studies have shown that early behavioral treatment can largely improve the cognitive and adaptive abilities of children with ASD, and early intensive interventions before age three can improve the prognosis to a large extent (9–11).

Early screening and early diagnosis play a key role in affecting the prognosis of this disease. Signs of ASD can occur very early, even in the first year of life and a diagnosis can be made at as early as 12 months (12). A formal diagnosis may be possibly made only at 18–24 months of age, and the stability of the diagnosis is quite high over time (13, 14). Therefore, early diagnosis of ASD is possible. However, at present, the diagnosis of ASD is made around the age of 4–5 years on average (15–17). There is a significant delay between the onset of ASD symptoms and diagnosis, which means that young children miss the opportunity for intervention during the optimal period of neuroplasticity. The American Academy of Pediatrics (AAP) recommends that children be screened for ASD at the 18- and 24-month checkups (18). There are many studies of early screening for ASD in developed countries (19–24). In a large early screening study, Robin et al. reported a diagnostic rate of 0.67% in toddlers (19). In another screening study of low-risk young children, the diagnostic rate of ASD was 0.65% (20). The initiative of early ASD screening starts much later in China than that in the developed countries. The first large early screening study for ASD in China was initiated by our team in 2013 and lasted for 4 years. In that study, the early diagnostic rate of ASD was 0.21% (25). With the large population in China, the early screening will detect many children with ASD. Early screening would significantly shorten the average time from onset to diagnosis and intervention of these children, thus increasing the possibility of improving their prognosis and relieving the families' burdens.

For widespread use and well-implementation, screening tools should be brief, easy to complete and effective. The Modified Checklist for Autism in Toddlers, Revised with Follow-up (M-CHAT-R/F) is a two-stage screening tool for ASD that has been reported to have adequate sensitivity and specificity (19, 20). The M-CHAT-R/F has been translated into more than 40 languages and requires little time and cost, making it one of the most widely used ASD screening tools. However, few studies have evaluated whether this screening tool performs adequately in Chinese Han toddlers. Similar to other screening questionnaires, the screening results are closely related to the quality of parental completion of a symptom checklist (19, 26). If parents underreport their children's symptoms, there will be a possibility of misdiagnosis. Brief observation of physicians who administer screening may increase the likelihood of early identification (25). We speculate that M-CHAT-R/F combined with brief observation of physicians would be an effective screening tool.

This study aimed to investigate the diagnostic rate of ASD screening and identify the validity of screening model combining M-CHAT-R/F with brief observation of physicians in the current Chinese three-level healthcare system.

METHODS

Participants

This study was implemented at 13 community healthcare centers in Xuhui District, Xuhui Maternal and Child Healthcare Hospital (XMCHH), and Children's Hospital of Fudan University (CHFU). Xuhui District, a central district of Shanghai with about 5,000–6,000 births per year, has a well-established three-level child healthcare system. Toddlers aged 18 to 24 months who were at well-child visit in Xuhui district from January 2018 to December 2019 were enrolled. Parents of all participants provided informed consent. The study was approved by the Ethical Committee of the Children's Hospital of Fudan University.

Screening Instruments and Procedure

When children aged 18–24 months attended routine well-child visits at community healthcare center, they underwent early screening using the M-CHAT-R/F and Binomial Observation Test (BOT).

M-CHAT-R/F is a two-stage screening questionnaire consisting of 20 questions on a scale of 0–20 (www.mchatscreen.com). The M-CHAT-R refers to the initial screening, while the M-CHAT-R/F refers to the second-stage screening process with follow-up. Positive screening for the M-CHAT-R includes 3 or more high-risk responses (total score: 3–7, moderate risk; total score: 8–20, high risk). If children receive a score of "high risk" (total score ≥ 8) on M-CHAT-R, they would bypass the follow-up and are considered positive on M-CHAT-R/F. If with a score of "moderate risk" (total score: 3–7), a screening process for follow-up is required. Positive screening for follow-up (M-CHAT-R/F) includes 2 or more risky reactions (**Figure 1**). In this study, parents first completed 20 questions in M-CHAT-R according to their children's daily performance. If children received a score of "moderate risk", the follow-up interview was completed by the primary care physicians (PCPs) at the 13 community healthcare centers.

Besides M-CHAT-R/F, PCPs also administered a two-step observational test which was called Binomial Observation Test (BOT). The first step is "Response to name" and the second step is "Follow commands." In the first step, PCP called the child's name twice in a clear voice at a normal volume. If the child failed to look toward the PCP, the second step was performed. In the "Follow commands" step, the child was required to follow two simple instructions such as waving goodbye, or blowing a kiss. If

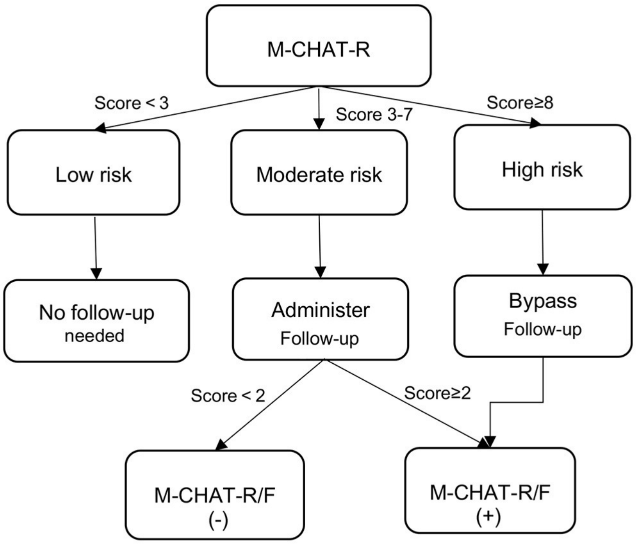


FIGURE 1 | Recommended algorithm based on 2-stage M-CHAT-R/F screening.

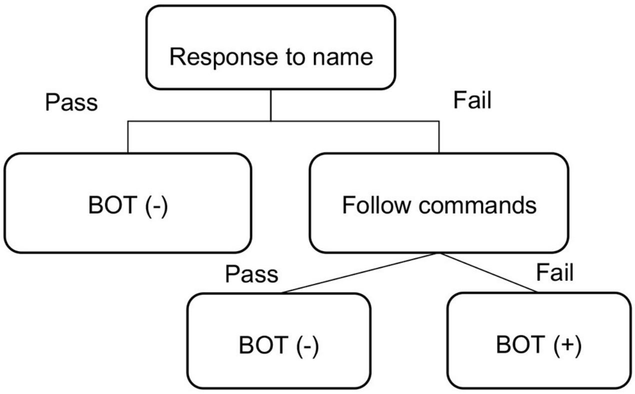


FIGURE 2 | Procedure of the binomial observational test (BOT).

the child could not follow either command, she/he failed the test. It meant that the child was screened positive on BOT (**Figure 2**).

Although the whole screening process was completed by PCPs at the 13 community healthcare centers, the pediatricians at XMCHH were responsible for supervision and quality management of PCPs' screening work.

Referral and Diagnosis

Toddlers who screened positive on M-CHAT-R/F and/or BOT were deemed positive and were referred to the tertiary diagnostic center: CHFU, for comprehensive ASD diagnostic evaluation and counseling on next step recommendations. The timing of these referrals was a hinge, as it could directly impact the age of diagnosis as well as the receipt of appropriate interventions. Therefore, a special green channel had been set up to help get referrals as soon as possible.

The diagnosis of ASD was made by developmental pediatricians at CHFU based on the ASD criteria in the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5), and further confirmed by the Autism Diagnostic Observation Schedule, second edition (ADOS-2). When parents refused to complete evaluations, a telephone follow-up was completed by pediatricians at CHFU and XMCHH.

To maximize the detection of missed cases, children who screened negative would take well-child checkups and follow-up at community healthcare centers every 3–6 months until they reached 3 years of age. Children were also referred to the CHFU if they were suspected of having ASD by PCPs during follow-up well-child visit.

Statistical Analyses

R statistical software was used to perform data analysis. Measurements such as ages were presented as mean ± SD (X ± S), and numerical data such as number of patients were presented as numbers and percentages. The *t*-test was applied for detecting differences in measurement data between groups, and the Chi-Square test was used to analyze differences in numerical data. If $P < 0.05$, it was considered statistically significant for all tests. Also, 95% Confidence interval (CI) were determined on the basis of the approximate normal distribution method.

RESULTS

M-CHAT-R/F Screening and Diagnostic Outcome

From January 2018 to December 2019, there were 11,190 toddlers with 18- and 24-month well-child visit were screened in Xuhui District. Of the total 11,190 toddlers, 474 (4.2%) were positive on M-CHAT-R or BOT and 126 (1.1%) were positive on M-CHAT-R/F or BOT. Ultimately, 36 children were diagnosed as ASD. The diagnostic rate of ASD through community screening was 0.32% (95% CI: 0.23–0.45%).

M-CHAT-R

Among the 474 children with positive screening results, 459 children had positive M-CHAT-R screening results (402 toddlers screened positive on M-CHAT-R only and 57 toddlers screened both positive on M-CHAT-R and BOT), with a screen-positive rate of 4.1% (459/11,190). Among the 459 children, 33 children were finally diagnosed with ASD, and the positive predictive value (PPV) of M-CHAT-R for ASD was 0.07 (33/459). Forty-six children scored in the high-risk range on M-CHAT-R and 20 of them were diagnosed with ASD. The PPV of M-CHAT-R high risk for ASD was 0.43 (20/46).

M-CHAT-R/F

The follow-up interviews were administered by trained PCPs from community healthcare centers. Among the 459 toddlers who were positive on M-CHAT-R, 22.9% (105) of them were screened positive on M-CHAT-R/F. Of the 105 children, 33

TABLE 1 | PPVs of M-CHAT-R, M-CHAT-R/F, and BOT.

	Total number	Diagnosed with ASD	PPV
M-CHAT-R (+)	459	33	0.07
M-CHAT-R high risk	46	20	0.43
BOT (+)	72	27	0.38
BOT & M-CHAT-R (+)	57	24	0.42
M-CHAT-R/F (+)	105	33	0.31

children were diagnosed with ASD. The PPV of M-CHAT-R/F was 0.31 (33/105), which was significantly higher than M-CHAT-R ($\chi^2 = 46.271$, $P < 0.001$). With a comparison between PPV of M-CHAT-R high-risk and that of M-CHAT-R/F for ASD, it is found that the former is higher with no statistically significant difference ($\chi^2 = 1.5441$, $P = 0.214$).

BOT Screening and Diagnostic Outcome

A total of 72 toddlers were screened positive on BOT (15 toddlers were screened positive on BOT only and 57 toddlers were screened both positive on M-CHAT-R and BOT). Of the 72 toddlers, 27 children were finally diagnosed as ASD, and the PPV of BOT for ASD was 0.38 (27/72), which was significantly higher than M-CHAT-R ($\chi^2 = 54.065$, $P < 0.001$) and similar to M-CHAT-R/F ($\chi^2 = 0.45782$, $P = 0.4986$). There were 57 toddlers screened both positive on M-CHAT-R and BOT. Of these 57 children, 24 were finally diagnosed with ASD. The PPV of M-CHAT-R & BOT was 0.42 (24/57). See **Table 1** for PPVs of M-CHAT-R, M-CHAT-R high risk, M-CHAT-R/F, and BOT.

In addition, there were 15 children who were positive on BOT but negative on M-CHAT-R. Of these 15 children, 3 were diagnosed with ASD. It meant that there was a 20% chance of being diagnosed with ASD in these children. They accounted for 8.3% (3/36) of all screen-positive ASD patients. We can see the flowchart and screening results in **Figure 3**.

Comparison to the Non-screened ASD Group in Shanghai

Thirty-six screen-positive children were diagnosed with ASD on the community-based early ASD screening model. Among them, 30 were male, and 6 were female (sex ratio = 5:1). The mean age at screening and diagnosis were 21.1 ± 2.71 months and 23.1 ± 4.55 months, respectively. The average interval time from initial screening to diagnosis was 2 months.

We compared the diagnostic age of community screened children with those who located in Shanghai but didn't receive community ASD screening. During the same period, 473 ASD children located in Shanghai were first diagnosed without ASD screening, including 399 males and 74 females. The ratio of males to females in non-screened group was 5.4:1, which was similar to the community screened ASD group. Overall, 60.3% of children with ASD did not have a comprehensive evaluation until after age 3 years old. The average diagnostic age of these patients was 43.2 ± 17.91 months. The diagnostic age of community screened group was significantly younger than the non-screened ASD group in Shanghai ($P < 0.001$). On average, screen-positive children were diagnosed 20 months earlier than children not screened (see **Table 2**).

False Negative Cases

A total of 12 children with negative screening results were diagnosed with ASD at the age of 30 ± 4.1 months, diagnosed 13 months earlier than children not screened on average. These 12 children all had negative screening results at 18 months of age and were referred to CHFU because of the PCPs' concern about ASD at the age of 27 ± 3.4 months. The interval time from initial negative screening to diagnosis was 12 ± 4.2 months.

Total Diagnostic Rate of ASD

Thirty-six screen-positive children were diagnosed with ASD, and 12 screen-negative children were diagnosed with ASD during follow-up well-child visit at community healthcare centers. Totally, 48 children were diagnosed with ASD.

In addition, 30 children who screened positive on M-CHAT-R/F or BOT refused to take an evaluation at CHFU. Among the 30 children, 16 children were positive on M-CHAT-R/F only, 8 children were positive on BOT only, and 6 children were M-CHAT-R/F and BOT both positive. The follow-up telephone interviews were made with their parents. The primary reason for refusal of evaluation was that the parents did not believe the child had an ASD-related problem and reported as social normal (28/30, 93.3%). And the secondary reason was that the parents moved house and could not be contacted (2/30, 6.7%). The 30 missing data were imputed as non-ASD, and an adjusted diagnostic rate was calculated. The 95% CI for the diagnostic rate was determined based on the approximate normal distribution method. When children aged 18–24 months were followed up to 3 years of age at community healthcare center, the average diagnostic rate of ASD was 0.43% (48/11,190, 95% CI: 0.32–0.57%).

DISCUSSION

Shanghai, one of the largest cities in China, has a well-established three-level child healthcare system that provides basic healthcare services and monitoring for children aged 0–3 years. PCPs at the community healthcare center provide screening, referral (level 1); pediatricians at the district maternal and child health centers provide monitoring, further referral (level 2); and pediatricians at the specialized children's hospitals provide diagnosis, consultation, and treatment (level 3). The three-level child healthcare system plays the significant role of pediatricians at each level. In particular, the PCPs at the community healthcare center, as the front line of defense, are essential. The three-level connection and cooperation can achieve early screening, early diagnosis and early intervention for ASD.

The AAP recommended screening for ASD at 18 and 24 months in 2007 (18). In 2016, the US Preventive Services Task Force (USPSTF) published a controversial report concluding that there was insufficient evidence to assess the balance of benefits and harms of early ASD screening (27). In response, the

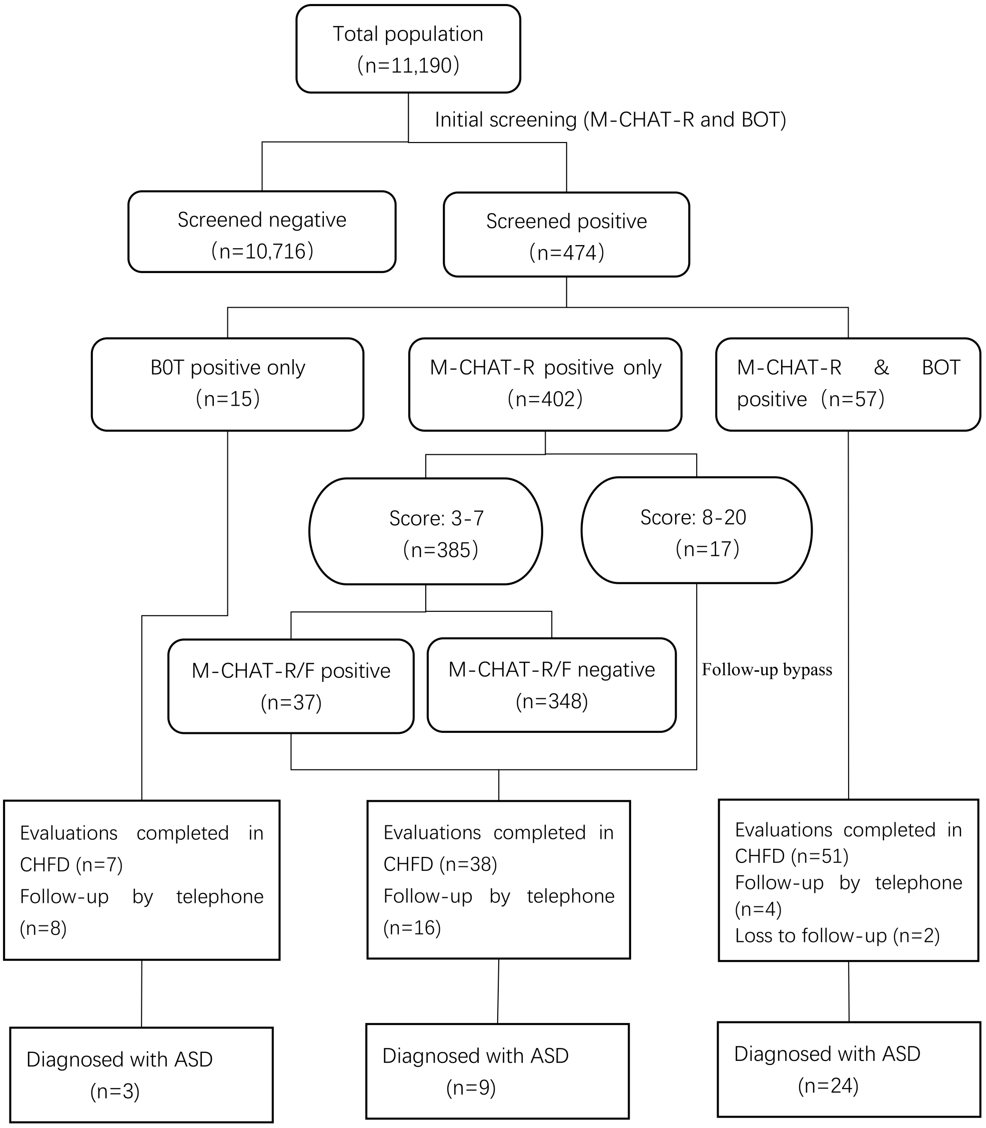


FIGURE 3 | Flowchart displaying the screening and diagnosis results for all toddlers studied.

TABLE 2 | Comparison of sex distribution and age at diagnosis between community screening ASD group and non-screening ASD group.

	Total number	Age at diagnosis (x ± s)	Male/female
Community screening	36	23.1 ± 4.55	5:1
Non-screening group	473	43.2 ± 17.91	5.4:1
χ 2/t		−17.507	$2.43*10^{-30}$
P-value		<0.0001	0.9999

AAP promptly issued a statement on their website, remaining committed to their recommendation for universal screening of 18- and 24-month-old children for ASD. Some other professional and advocacy organizations such as the American Academy of Child Neurology, the American Academy of Child and Adolescent Psychiatry and the American Academy of Pediatrics' Bright Futures also recommend early universal screening (28). There is ample evidence that strongly supports the universal ASD screening in children aged 18–24 months (29, 30). In 2017, the Chinese expert consensus on early screening for ASD, issued by the Chinese Medical Association (CMA), recommended that pediatricians at all levels of hospitals should provide regular early ASD screening for infants and toddlers at 9, 18, and 24 months of age, on the basis of China's three-level child healthcare system (31). The corresponding author of the present paper is also one of the main contributors to this expert consensus.

The current study was based on the three-level child healthcare system. However, the whole screening process was completed in community (level 1). Children who screened positive at community healthcare center (level 1) were directly referred to the tertiary diagnostic center (level 3) for diagnostic evaluation. What pediatricians at the district maternal and child healthcare hospital (level 2) should do is playing a connecting role in supervision and quality management of the community healthcare center. Such a referral model that lessens intermediate referral can reduce the loss of follow-up visit and avoid the potential time delay.

In our previous study, only 64.8% (283/437) of positive cases from primary screening (level 1) completed the face-to-face second screening (level 2) (25). Through an analysis of causes of loss of follow-up visit during the intermediate referral from the primary to the secondary, we found that many families skipped the secondary hospital due to the inconvenient transportation or the urgent demands for medical treatment. Hence, we canceled the intermediate referral from the community healthcare center to the district maternal and child healthcare hospital in this longitudinal study.

In this study, a total of 11,190 toddlers were screened and 36 screen-positive children were diagnosed with ASD. Screen-positive children were diagnosed 20 months earlier than children not screened, which means they could significantly improve the long-term outcomes. The diagnostic rate of ASD through community screening was 0.32% (95% CI: 0.23–0.45%), which was higher than the 0.21% we reported in 2018 (25). There were several explanations that may account for the increased early detection rate. First, the next follow-up screening was completed immediately after the initial M-CHAT-R screening, which reduced the loss of follow-up visits. Second, the experience of PCPs increased. PCPs who started ASD screening back in 2013 had gained some experience. They were more agile than before to detect toddlers with ASD. Third, public awareness of ASD had increased. In recent years, the extensive scientific knowledge propagation of ASD by the government and media had caught more attention among the public. Thus, parents were more likely to detect abnormal behaviors of their children and were willing to send them to the tertiary hospital for diagnosis. However, this rate was still a little lower than studies conducted in some developed countries (19–22). This may be due to different study designs, the ethnic and geographical differences.

To minimize missed false-negative cases, children who screened negative at 18–24 months received routine well-child checkups and follow-up every 3–6 months at the community healthcare centers until they reached 36 months of age. If they were suspected of having ASD by PCPs, they would be referred to the CHFU for evaluation and diagnosis. Benefiting from the healthcare and referral networks, a total of 12 children with negative screening results at 18 months of age were identified as having ASD. They were diagnosed with ASD at the mean age of 30 months, 13 months earlier than the children who did not go through community screening and systematic management. We recommend that children who pass ASD screening at 18 months still need developmental surveillance until at least 3 years old. The follow-up well-child visit at community healthcare center and the referral network are necessary.

The diagnostic rate of ASD was 0.43% (95% CI: 0.32–0.57%) When children were followed up to 3 years of age at the community healthcare center. This rate was slightly lower than the prevalence of ASD in children aged 6–12 years reported by Zhou et al., which was 0.7% (5). In any case, it cannot be expected that all cases of ASD will be found in toddlers. In fact, children with high-functioning autism are usually diagnosed at preschool or even school age, when they enter a group setting with high demands on social communication. We investigated the sex ratio in screen-positive ASD and non-screened ASD groups, which was very similar in both groups, around 5:1 (male/female). Interesting, the sex ratio in false-negative cases was also 5:1 (male/female).

The PPV of M-CHAT-R/F was 0.31, which was significantly higher than M-CHAT-R. The follow-up interview can improve PPV and conserve evaluative resources. Therefore, it is necessary to administer the follow-up interviews for children with medium risk on M-CHAT-R screening. The PPV of M-CHAT-R/F for ASD reported in previous studies was between 0.4 and 0.5 (17, 18), which was slightly higher than 0.31 in this study. However, even without follow-up interviews, the PPV of M-CHAT-R high risk for ASD was 0.43, which was still higher than M-CHAT-R/F and showed good diagnostic significance. Therefore, we recommend that children who receive score of "high risk" on M-CHAT-R can skip follow-up interviews and should receive immediate specialist evaluation.

There were 15 children who were positive on BOT but negative in the questionnaire. Of these 15 children, 3 were diagnosed with ASD, accounted for 8.3% of all screen-positive ASD toddlers. The PPV of BOT was 0.38, which was slightly higher than M-CHAT-R/F (0.31). BOT is two-step behavioral test and very easy to practice. The inclusion of BOT in the screening process can reduce missed diagnoses and affect the prognosis of a small number of toddlers with ASD. Moreover, it is helpful to improve the early identification skills of the PCPs by training and test through the simple process of observation, thus fully exerting themself to the front line of defense.

The major limitation of this study is that it was carried out in Shanghai Xuhui District, bringing about certain regional limitations. Currently, similar research is urgently needed in other areas. We have planned multiple studies in other jurisdictions to get more robust data fur further analysis. These studies are still ongoing and we can look forward to the release of the results in the future.

CONCLUSIONS

Based on the above results, we can conclude that an efficient large-scale ASD screening in a large community-based population need the support from a well-established child healthcare system, primary care physicians with basic knowledge of ASD screening, and a standardized screening tool. In China, child healthcare system is well-established in most of the cities. We can rely on the three-level child healthcare system, reliable screening tools and surveillance strategies to conduct ASD screening in community-based populations. The screening model combining M-CHAT-R/F with BOT is worth replicating. With a large population in China, a considerable number of ASD cases can be detected and their families will benefit from early ASD screening.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Ethical Committee of the Children's Hospital of Fudan University. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

All authors listed have made a substantial, direct, and intellectual contribution to the work and approved it for publication.

FUNDING

This study was supported in part by the Key Subject Construction Project of Shanghai Municipal Health Commission (shslczdzk02903), the National Natural Science Foundation of China (NSFC, 61733011), and Industry-University-Research High Tech Transformation Incubation Project (FDEKCXY08).

ACKNOWLEDGMENTS

We sincerely thank all the enrolled children and their parents for their kind participation and support, as well as the primary care physicians in Shanghai Xuhui District who performed ASD screening.

REFERENCES

1. American Psychiatric Association. *Diagnostic and Statistical Manual of Mental Disorders*. 5th ed. Washington, DC: American Psychiatric Association (2013).
2. Lyall K, Croen L, Daniels J, Fallin MD, Ladd-Acosta C, Lee BK, et al. The changing epidemiology of autism spectrum disorders. *Annu Rev Public Health.* (2017) 38:81–102. doi: 10.1146/annurev-publhealth-031816-044318
3. Lai MC, Lombardo MV, Baron-Cohen S. Autism. *Lancet.* (2014) 383:896–910. doi: 10.1016/S0140-6736(13)61539-1
4. Sun X, Allison C, Wei L, Matthews FE, Auyeung B, Wu YY, et al. Autism prevalence in China is comparable to western prevalence. *Mol Autism.* (2019) 10:7. doi: 10.1186/s13229-018-0246-0
5. Zhou H, Xu X, Yan W, Zou X, Wu L, Luo X, et al. Prevalence of autism spectrum disorder in china: a nationwide multi-center population-based study among children aged 6 to 12 years. *Neurosci Bull.* (2020) 36:961–71. doi: 10.1007/s12264-020-00530-6
6. Ten Hoopen LW, de Nijs PFA, Duvekot J, Greaves-Lord K, Hillegers MHJ, Brouwer WBF. Children with an autism spectrum disorder and their caregivers: capturing health-related and care-related quality of life. *J Autism Dev Disord.* (2020) 50:263–77. doi: 10.1007/s10803-019-04249-w
7. Lavelle TA, Weinstein MC, Newhouse JP. Economic burden of childhood autism spectrum disorders. *Pediatrics.* (2014) 133:e520–9. doi: 10.1542/peds.2013-0763
8. Bejarano-Martín Á, Canal-Bedia R, Magán-Maganto M, Fernández-Álvarez C, Martín-Cilleros MV, Sánchez-Gómez MC, et al. Early detection, diagnosis and intervention services for young children with autism spectrum disorder in the European Union (ASDEU): family and professional perspectives. *J Autism Dev Disord.* (2014) 50:3380–94. doi: 10.1007/s10803-019-04253-0
9. Dawson G, Jones EJ, Merkle K, Venema K, Lowy R, Faja S, et al. Early behavioral intervention is associated with normalized brain activity in young children with autism. *J Am Acad Child Adolesc Psychiatry.* (2012) 51:1150–9. doi: 10.1016/j.jaac.2012.08.018
10. Dawson G, Rogers S, Munson J, Smith M, Winter J, Greenson J, et al. Randomized, controlled trial of an intervention for toddlers with autism: the early start denver model. *Pediatrics.* (2010) 125:e17–23. doi: 10.1542/peds.2009-0958
11. Shi B, Wu W, Dai M, Zeng J, Luo J, Cai L, et al. Cognitive, language, and behavioral outcomes in children with autism spectrum disorders exposed to early comprehensive treatment models: a meta-analysis and meta-regression. *Front Psychiatry.* (2021) 12:691148. doi: 10.3389/fpsyt.2021.691148
12. Pierce K, Gazestani VH, Bacon E, Barnes CC, Cha D, Nalabolu S, et al. Evaluation of the diagnostic stability of the early autism spectrum disorder phenotype in the general population starting at 12 months. *JAMA Pediatr.* (2019) 173:578–87. doi: 10.1001/jamapediatrics.2019.0624
13. Guthrie W, Swineford LB, Nottke C, Wetherby AM. Early diagnosis of autism spectrum disorder: stability and change in clinical diagnosis and symptom presentation. *J Child Psychol Psychiatry.* (2013) 54:582–90. doi: 10.1111/jcpp.12008
14. Loubersac J, Michelon C, Ferrando L, Picot MC, Baghdadli A. Predictors of an earlier diagnosis of autism spectrum disorder in children and adolescents: a systematic review (1987–2017). *Eur Child Adolesc Psychiatry.* (2021). doi: 10.1007/s00787-021-01792-9. [Epub ahead of print].
15. Zwaigenbaum L, Penner M. Autism spectrum disorder: advances in diagnosis and evaluation. *BMJ.* (2018) 361:k1674. doi: 10.1136/bmj.k1674
16. Baio J, Wiggins L, Christensen DL, Maenner MJ, Daniels J, Warren Z, et al. Prevalence of autism spectrum disorder among children aged 8 years -autism and developmental disabilities monitoring network, 11 sites, United States, 2014. *MMWR Surveill Summ.* (2018) 67:1–23. doi: 10.15585/mmwr.ss6706a1
17. Constantino JN, Abbacchi AM, Saulnier C, Klaiman C, Mandell DS, Zhang Y, et al. Timing of the diagnosis of autism in African American children. *Pediatrics*. (2020)146:e20193629. doi: 10.1542/peds.2019-3629
18. Johnson CP, Myers SM. American Academy of Pediatrics Council on Children With Disabilities. Identification and evaluation of children with autism spectrum disorders. *Pediatrics.* (2007) 120:1183–215. doi: 10.1542/peds.2007-2361
19. Robins DL, Casagrande K, Barton M, Chen CM, Dumont-Mathieu T, Fein D. Validation of the modified checklist for Autism in toddlers, revised with follow-up (M-CHAT-R/F). *Pediatrics.* (2014) 133:37–45. doi: 10.1542/peds.2013-1813
20. Bradbury K, Robins DL, Barton M, Ibañez LV, Stone WL, Warren ZE, et al. Screening for autism spectrum disorder in high-risk younger siblings. *J Dev Behav Pediatr.* (2020) 41:596–604. doi: 10.1097/DBP.0000000000000827
21. Baduel S, Guillon Q, Afzali MH, Foudon N, Kruck J, Rogé B. The French version of the Modified-Checklist for Autism in Toddlers (M-CHAT): a validation study on a French sample of 24 month-old children. *J Autism Dev Disord.* (2017) 47:297–304. doi: 10.1007/s10803-016-2950-y
22. Kamio Y, Inada N, Koyama T, Inokuchi E, Tsuchiya K, Kuroda M. Effectiveness of using the modified checklist for autism in toddlers in two-stage screening of autism spectrum disorder at the 18-month health check-up in Japan. *J Autism Dev Disord.* (2014) 44:194–203. doi: 10.1007/s10803-013-1864-1
23. Carbone PS, Campbell K, Wilkes J, Stoddard GJ, Huynh K, Young PC, et al. Primary care autism screening and later autism diagnosis. *Pediatrics.* (2020) 146:e20192314. doi: 10.1542/peds.2019-2314
24. Monteiro SA, Dempsey J, Berry LN, Voigt RG, Goin-Kochel RP. Screening and referral practices for autism spectrum disorder in primary pediatric care. *Pediatrics.* (2019) 144:e20183326. doi: 10.1542/peds.2018-3326
25. Li C, Zhu G, Feng J, Xu Q, Zhou Z, Zhou B, et al. Improving the early screening procedure for autism spectrum disorder in young children: experience from a community-based model in Shanghai. *Autism Res.* (2018) 11:1206–17. doi: 10.1002/aur.1984

26. Marlow M, Servili C, Tomlinson MA. A review of screening tools for the identification of autism spectrum disorders and developmental delay in infants and young children: Recommendations for use in low- and middle-income countries: a review of screening tools for autism and developmental delay. *Autism Res.* (2019) 12:176–99. doi: 10.1002/aur.2033
27. Siu AL, US Preventive Services Task Force (USPSTF), Bibbins-Domingo K, Grossman DC, Baumann LC, Davidson KW, et al. Screening for autism spectrum disorder in young children US preventive services task force recommendation statement. *JAMA.* (2016) 315:691–6. doi: 10.1001/jama.2016.0018
28. Fein D, Baby Sibs Research Consortium. Commentary on USPSTF final statement on universal screening for autism. *J Dev Behav Pediatr.* (2016) 37:573–8. doi: 10.1097/DBP.0000000000000345
29. Zwaigenbaum L, Bauman ML, Choueiri R, Kasari C, Carter A, Granpeesheh D, et al. Early screening of autism spectrum disorder: recommendations for practice and research. *Pediatrics.* (2015) 136(Suppl. 1):S41–59. doi: 10.1542/peds.2014-3667D
30. Dawson G. Why it's important to continue universal autism screening while research fully examines its impact. *JAMA Pediatr.* (2016) 170:527–8. doi: 10.1001/jamapediatrics.2016.0163
31. Xu X, Zhou XB Li TY. Expert consensus on early screening and early intervention of children with autism spectrum disorders. *Zhonghua Er Ke Za Zhi.* (2017) 55:890–7. doi: 10.3760/cma.j.issn.0578-1310.2017.12.004

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers | Frontiers in Psychiatry

ORIGINAL RESEARCH
published: 31 May 2022
doi: 10.3389/fpsyt.2022.913600

"Setting Them Up for Success": Including Youth on the Autism Spectrum in 4-H

Carolyn E. B. McCormick[1]*, **Veronika Peskova**[1], **Abby Morgan**[2], **Emily Carlson**[1] and **Rose A. Mason**[3]

[1] Department of Human Development and Family Studies, College of Health and Human Sciences, Purdue University, West Lafayette, IN, United States, [2] Purdue Extension, Montgomery County, Crawfordsville, IN, United States, [3] Department of Educational Studies, College of Education, Purdue University, West Lafayette, IN, United States

Youth on the autism spectrum often face challenges accessing services in rural communities compared to those who live in higher resource areas. There is a particular need for services that support skills that will help youth transition to adulthood and future employment. 4-H is a national youth development program that is well-positioned to address the needs of youth on the autism spectrum; however, minimal empirical evidence exists about the implementation and effectiveness of inclusive practices in 4-H programs. The goal of this study was to better understand barriers to enrollment and to identify gaps in support for youth on the autism spectrum participating in 4-H. Twenty Extension Educators in Indiana participated in two focus groups. Thematic analysis of focus group transcripts identified barriers to enrollment including awareness of 4-H as an inclusive program and difficulties navigating 4-H culture. Our analysis identified themes related to new training content and delivery including a resource portfolio, communication, individualized accommodations, and working within the existing leadership training structure. Findings support the benefits of 4-H as a program that can promote life skills and personal development for youth on the autism spectrum but also highlight a significant need for additional training opportunities and resources to increase uptake and improve the implementation of inclusive practices.

Keywords: autism, extracurricular activities, inclusion, Cooperative Extension, community-based programs

OPEN ACCESS

Edited by:
Andy Shih,
Autism Speaks, United States

Reviewed by:
Gloria K. Lee,
Michigan State University,
United States
Jonna Bobzien,
Old Dominion University, United States

****Correspondence:***
Carolyn E. B. McCormick
mccorm37@purdue.edu

Specialty section:
This article was submitted to
Autism,
a section of the journal
Frontiers in Psychiatry

Received: *05 April 2022*
Accepted: *04 May 2022*
Published: *31 May 2022*

Citation:
McCormick CEB, Peskova V, Morgan A, Carlson E and Mason RA (2022) "Setting Them Up for Success": Including Youth on the Autism Spectrum in 4-H.
Front. Psychiatry 13:913600.
doi: 10.3389/fpsyt.2022.913600

INTRODUCTION

Youth with autism spectrum disorder (on the autism spectrum) face unique challenges with social communication, including difficulty with initiating and maintaining conversations, understanding social nuances, and developing relationships with peers (1). Youth on the autism spectrum also tend to perseverate or have a limited repertoire of interests, making it more difficult to identify peers with similar interests (2). In adulthood, these differences contribute to higher rates of under and unemployment (3), which impacts the quality of life for adults on the autism spectrum. Stakeholders have highlighted the need for more supports and services with a specific focus on developing life skills that would promote success across the entire lifespan (4, 5). Accessing services that support the development and education of youth on the autism spectrum is often difficult due to cost and availability (6, 7). This is particularly salient for youth who live in rural areas, where issues with the availability of resources are even more pronounced for children on the autism spectrum (8, 9).

One avenue for overcoming resource obstacles, is through the implementation of interventions in natural contexts, capitalizing on well-established social structures and groups (10), such as those provided by community-based programs (11). Community-based programs typically have established peer groups and may also have multiple interest groups which increase the likelihood that involved youth, with and without disabilities, can identify a peer group with common interests. For youth on the autism spectrum, participation provides valuable opportunities to practice communication and social interaction skills, as well as the potential to develop friendships (2). Involvement in organized extracurricular activities has also been associated with less depression and loneliness (12). Young adults on the autism spectrum with a record of high academic achievement have also reported participation in extra-curricular activities as a positive venue to develop both their interests and social skills (13).

One such community program is 4-H https://4-h.org/. 4-H, which stands for Head, Heart, Hands, & Health, is delivered through Cooperative Extension, a community-university partnership that is comprised of more than 100 public universities across the nation. 4-H is the largest youth development program in the United States and serves over 6 million youth with the support of adult volunteers and 4-H professionals. This national program provides local communities with research-based programming that centers around science, healthy living, civic engagement, and agriculture. Programs offered through 4-H focus on using hands-on experiences that empower young people with the practical and life skills they need to be leaders now and in the future. Practical skills and content knowledge can be gained across a wide range of topics such as robotics, animal husbandry, sewing, and photography. Life skills such as leadership, resiliency, and communication, are learned and enhanced through various delivery methods such as camps, community clubs, and in-school and afterschool programs. 4-H programs also offer opportunities for peers to connect and form new friendships. Most importantly for youth on the autism spectrum, 4-H has supporting diversity, equity, and inclusion within its program goals. This means that children with all disabilities, including autism, should have access to all 4-H programming.

Despite the benefits of participation in extracurricular activities including an avenue to develop and explore interests, create new friendships, and learn everyday skills (14), youth with disabilities participate at lower rates than their typically developing peers (15). Additional challenges when it comes to enrollment and participation in extracurricular activities negatively impact the rate of participation for youth with disabilities (15, 16). There are a variety of reasons that may contribute to why many youth with disabilities refrain from engaging in extracurricular activities, including barriers related to awareness, skills, opportunities, and support from teachers, parents, or guardians (17). Countless organizations, including 4-H, have no specific training program for volunteers and employees on how to serve youth with disabilities (18). Investment in inclusive practices in out of school environments increases participation from youth with disabilities (19). Within 4-H, there have been attempts to implement additional programs or resources to specifically address the inclusion of youth with disabilities, however, these have been limited in scope and/or longevity (20–22). Together this indicates a need to increase recruitment and retention efforts for youth on the autism spectrum.

Despite positive attitudes toward 4-H (23) and the national availability of the program, it is unclear why this potentially valuable resource is currently underutilized. As a first step in understanding ways to promote 4-H for youth on the autism spectrum, the goal of this project was to assess the current experiences and practices of Extension Educators to address the following research questions: (1) What are the barriers to enrollment in 4-H for youth on the autism spectrum; (2) What types of training resources are available to Extension Educators within 4-H to create inclusive programming; (3) What supports and strategies do Extension Educators already use to support youth on the autism spectrum within their programs; (4) What challenges do Extension Educators and youth on the autism spectrum face within 4-H; (5) What types of new strategies and resources would help to improve inclusion?

METHODS

We report our methods according to the COREQ criteria (24).

Participants

The participants for two focus groups were recruited through purposeful sampling via electronic flyers sent through email to the full list of Extension Educators in Indiana and through social media posts. Extension Educators collaborate with the university, organizations, businesses, and schools to provide evidence-based educational programs to their communities. Extension Educators who oversee 4-H Youth Development programs provide leadership to club leaders, counselors, and others who support the implementation of programming throughout the year. They also lead and participate in the implementation of programs. In Indiana, there is one 4-H Youth Development Extension Educator in each of the 92 counties. All 20 participants (5 male, 15 female) were current Extension Educators in Indiana who worked with 4-H programs. Eight (five in focus group one and two in focus group two) of the 20 participants reported a close personal connection to someone on the autism spectrum (2- immediate family, 4-extended family, 2-friends).

Focus Group Procedures

All research was conducted with approval from the Purdue University Institutional Review Board. The two individual focus groups (10 participants per group) took place at the Fall Youth Staff Conference for Extension Educators in September of 2019. Focus group questions were developed by a team consisting of two faculty members, four Extension Educators, and one graduate student. The semi-structured focus groups consisted of ten participants each, the moderator (CM) and the assistant moderator (VP). Moderators were both female with expertise in autism and intervention services for children on the autism spectrum, but no direct experience with 4-H. The moderator introduced the purpose of the study and participants received folders with consent forms to read and sign. The participants also filled out a brief survey. During the focus groups, participants were asked open-ended questions [see **Table 1**] and had space to share and discuss their opinions and experiences. In addition to questions in the guide, moderators used probes to clarify (e.g., "can you describe a little bit more..;" "What do you mean by that?"), to expand perspectives [e.g., "any on this side of the table?" "So I haven't heard from (Name), is there anything that you've…"], and to amplify responses (e.g., "You have touched a little bit on this before, but I would love to hear a little bit more about the types of supports and strategies you implemented to help students on the spectrum?"; "So do you have ideas about what kinds of formats would be more acceptable?"). Sessions were each 1 hour with 10 minutes for introductions and consent and ∼50 minutes for questions (51.51 and 49.47). All participants contributed to the discussion. After the focus group, the moderator and assistant moderator held a debriefing session. Each of the focus groups was audio and video recorded. Recordings were later transcribed using Nvivo software.

Analytic Plan

Our team used techniques from grounded theory (25, 26) and thematic analysis (27) in our analysis process. The initial phase was an iterative process where each team member engaged in open coding of the transcripts independently to develop a coding framework/codebook based on our research questions and the transcripts that resulted in both structural and data-driven themes. The team at this phase included two faculty members, two extension educators, one graduate student, and two undergraduate students. During this phase, team members met to discuss the identified codes and together, finalized a codebook. To build the codebook, individual codes were also initially grouped into larger categories or themes based on our research questions and those that emerged from the data (27). For each code, example quotes from within the data that best illustrated that code were also selected. This process allowed us to include themes we anticipated based on our research objectives and focus group guide but also allowed for openness to capture relevant themes that emerged during the coding process. In the second phase, individual team members (3 undergraduate students, 1 graduate student) independently coded each transcript within Nvivo. A larger team (all coders and two faculty members) then met to review the transcript coding line by line and discuss any coding discrepancies. Discrepancies were resolved via consensus. After consensus coding, a constant comparative approach was used to identify subthemes (26). In the final stage, axial coding was performed to link overlapping categories, reduce the final number of categories, and develop our final themes (25).

RESULTS

Five main themes emerged that directly addressed our research questions: (1) enrollment process and barriers; (2) Extension Educator challenges; (3) youth challenges; (4) active strategies; and (5) new supports and resources. Two data-driven themes were also identified that would potentially impact the development of new resources: (1) benefits; and (2) Extension Educator perspectives. Each theme and associated subtheme are described below with representative quotes. Themes are outlined in **Table 2** and additional quotes are also reported in **Supplementary Table 1**.

Enrollment Process

During discussions of the enrollment process for families, participants described the types of programs and frequency with which they encountered youth on the autism spectrum within 4-H. They also described facilitators and barriers to enrollment.

Autistic Participation

All participating educators reported youth on the autism spectrum participating in their programs. The frequency varied from one youth member to several youth members over the years. As noted by one of the participants "We have had kids that have been involved in virtually every project." Commonly reported programs were 4-H Camp, 4-H Academy, and a wide range of 4-H clubs (e.g., robotics, alpacas, arts and crafts).

Getting Involved

Extension Educators described several aspects of the enrollment process that facilitated getting youth on the autism spectrum enrolled in 4-H programs. Many Extension Educators mentioned that the youth on the autism spectrum within their programs came, "always from a family tradition." This prior familiarity with the program made parents more likely to encourage their children to enroll as well. Several Extension Educators emphasized the importance of communication with new families when they described conversations that were necessary to make families feel comfortable enrolling. Educators and club leaders with personal experiences related to children with disabilities were also a draw for families to join. "I said you know you're, you're coming to a group of people that not only know how to do this but live it every day." For the youth, Extension Educators mentioned that finding or creating clubs that aligned with their interests was a big draw for getting new 4-H members enrolled.

Awareness

The first major barrier related to enrollment was general awareness related to 4-H. Extension Educators reported talking to parents who had never heard of 4-H or were not aware that it was an inclusive program where children with disabilities "aren't treated differently." In addition, knowing what's available and how to find the best fit could be a challenge. "Because I mean we have like 13 clubs you can pick from. So, how do know unless you're in dog obedience or you're in horse and pony, how do know what would be best for my kid to join?" Extension Educators also reported minimal referrals from schools or other service providers, indicating a lack of community awareness of 4-H as an option for youth on the autism spectrum.

4-H Culture

Participants shared that being new to the 4-H environment and reading through documents, manuals, and rules books that families receive might be "like navigating a whole other

TABLE 1 | Research questions and corresponding focus group questions.

Research question	Focus group question
1.What are the barriers to enrollment in 4-H for youth on the spectrum	a) How did the youth with autism first get involved with 4-H b) What barriers, if any, do you see for students with autism who want to participate in 4-H but aren't already involved?
2. What types of training resources are available to Extension Educators within 4-H to create inclusive programming	a) What training did you receive on inclusion for students with autism?
3.What supports and strategies do Extension Educators actively use to support youth on the spectrum within their programs?	a) What types of supports or strategies have you implemented to support students with autism?
4.What challenges do Extension Educators and youth face related to inclusion?	a) As an Extension educator, what are the challenges you face when engaging students with autism? b) What do you see as the biggest challenges for students with autism who participate in 4-H?
5.What types of new strategies and resources would help to improve inclusion?	a) In what areas, if any, do you feel you need more support or resources?

language." They also shared that all information is in written form, and therefore, the amount of reading required to learn about 4-H may be discouraging to families. The same difficulty might apply to the recruitment materials as it might be a challenge to communicate the benefits of 4-H programs when people do not want to read through the materials.

Benefits

Although there were no specific probes related to benefits for youth participating in 4-H programs, positive aspects of the programs were a frequent theme within the discussion related to inclusion. This theme highlighted the potential impact of 4-H programs if they were able to reach more youth and improve their inclusive practices. Across both groups, participants shared that youth achieved the general goals of 4-H program related to practical and life skill development.

Practical Skill Development

As expected for all participants of 4-H programs, Extension Educators described how youth on the autism spectrum develop valuable practical skills through 4-H participation.

> … he just developed a really close relationship with animals through the 4-H program and um you know could communicate with them in a way that was um amazing to watch and that was kinda his thing and he learned everything you know needed to know about those animals and um like you said was just able to really grow in his ability to communicate and um be in front of people.

Educators reported a wide range of skills including leadership, demonstration, independence, and communication, as well as skills specific to program topics (e.g., animal care).

Life Skill Development

Beyond the skills taught within the 4-H curriculum, Extension Educators conveyed that participation of youth on the autism spectrum often resulted in great personal development. "She had told me she always wanted to be a teacher, but she never had the confidence and when 4-H had the confidence in her to lead the robotics club, it gave her confidence uh confidence she had never experienced." Some participants observed that youth on the autism spectrum developed their confidence to such a level that they took on the leadership roles (e.g., 4-H representative, liaison, and counselor).

Youth Challenges

Moderators asked participants to share any challenges they observed youth on the autism spectrum face while participating in programs. Extension Educators' challenges related to specific contexts (fair process, peers) and also general issues related to accessing appropriate accommodations.

Accommodations

Youth on the autism spectrum come to 4-H needing additional supports that are specific and varied. The participants discussed examples of youth facing difficulties when those needs were not met.

> We make sure the counselors know what the schedule is and what's happening all day, but we don't always make sure that those campers know what's gonna happen throughout the day. And I know [name], that was his anxiety with camp was like not knowing what was gonna happen.

Additional examples included youth with minimal verbal abilities who struggled with communication and the daunting amount of paperwork required in some programs. These examples highlighted the need for more flexibility and knowledge on the behalf of 4-H.

Fair Process

The most challenging context for youth on the autism spectrum was overwhelmingly the 4-H fairs. Youth often struggled to adhere to strict rules during judging and deviations from 4-H traditions were often met with resistance by fair organizers. Together this resulted in exclusion from activities or misjudgment of that youth's efforts and abilities. "When it–at the judging, ask my child a question the clock's turning and yet he has a problem with communication processing time. And the judge doesn't think he knows it, so he moves on to something else."

TABLE 2 | Results: Themes and subthemes.

Theme	Subtheme
Enrollment process and barriers	Autistic participation
	Getting involved
	Awareness
	4-H culture
Benefits	Skill development
	Personal development
Educator perspectives	Personal attitudes
	Extension leadership
	Personal experience
Educator challenges	Personalized supports
	Parent communication
	Working with others
	Training gaps
Youth challenges	Accommodations
	Fair process
	Peer issues
Active strategies	Individualizing
	Mentor mentoring
	Peer mentoring
	Outreach
New supports and resources	Increase enrollment
	New Strategies
	Resource portfolio
	Connections with parents
	Flexibility
	Mentoring
	New Trainings
	New Content
	Train-the-trainer
	Stakeholder Involvement

The venue presents additional challenges, particularly for youth with sensory sensitivities.

> It's not mandatory that you come to the judging but of course it's encouraged. And I know there are some who just don't come. Mom comes drops the project off and leaves. And so that 4-'Her's missing out on the opportunity that everyone else is getting because it's, it's just too overloading for them.

Overall the combination of the sensory intensity of the venue and difficulties interacting with unfamiliar adults make the fair process particularly difficult for youth on the autism spectrum.

Peer Issues

Participants described difficulties between youth on the spectrum and their neurotypical peers.

> And so you know, it's hard to take 300 kids at camp and educate them. We try to do a good job with our counselors and our junior staff but sometimes kids don't understand why this person's a little different and can be, can be cruel.

Educators identified that often these challenges arise from a lack of awareness and acceptance on the part of the neurotypical youth that requires additional education to address.

Extension Educator Perspectives

Throughout the focus groups, although not directly asked, participants revealed their perspectives, attitudes, and experiences that influenced their implementation of inclusive practices within their programs.

Personal attitudes

There was strong agreement across both focus groups that inclusion is an important aspect of 4-H. "…we, I think as educators, love the program we are in so much that we want to go to the nth degree to be inclusive and make it inclusive for every kid." Many educators indicated the importance of not identifying children to be "different," so as not to single out children with disabilities.

> And I think that's one reason why 4-H is appealing to parents who are familiar with the program, because they aren't treated differently. They get the same public recognition [Yes] that all of the other 4-Hers get. [Mhmm] Without them physically standing out [Out as] or being different [being - yes!], yes.

Moreover, the participants shared their openness and desire to receive more training and resources on inclusion and including youth on the autism spectrum in their programs together with resources on how to support club leaders/volunteers, counselors, and judges, and lead effective communication with families.

Extension Leadership

All of the participants expressed receiving the overall message from 4-H leadership to be inclusive and support inclusive practices.

> I mean we talk about inclusion all the time, but we don't specifically say include the people with autism or include this group it's just kinda a general everybody is welcome everybody should be included.

This was echoed frequently in emphasis that all youth are not to be "identified as different" within 4-H. However, Educators conveyed a desire for more support from Extension leadership in the form of increased practical training on inclusion.

Personal Experience

The participants reported the use of their own experiences interacting with youth on the autism spectrum as a resource to help guide other educators, club leaders, and volunteers when having youth on the autism spectrum in their programs. "So, umm being able to, to understand what their fears are. Having those conversations with some of my co-workers, you know, through the years that I've dealt with that gives us the resources to, you know, answer those questions." The majority of these experiences originated from (a) being a parent of a child with a disability or being in a close relationship with them, (b) having previous experience of working with a child with a disability in their programs, and (c) having previous experience of working with a child with a disability out of 4-H (special ed. teachers, teachers). Participants reflected on key aspects that they learned from their own previous experiences and interactions when working with families with youth on the autism spectrum.

Extension Educator Challenges

Researchers prompted participants to describe any specific challenges they faced in providing inclusive programming for youth on the spectrum. They shared several areas where they struggled with youth and with others important for the support of 4-H programs.

Personalized Supports

Participants recognized the importance of meeting the individual needs of the youth in their programs but sometimes struggled to create personalized supports for youth on the h autism spectrum.

> I think there's so much diversity as far as the-the range of what to expect [Hmm] that it really is difficult. You really have to get to know that child and talk to the parents in order to find out what, what are the needs that I need to address. You know, that's my biggest challenge…

This was echoed by another participant.

> …finding a way to get her to come to those things and get out of the house or stay longer, don't just come for show your goat and go home, but stay and engage and you know different things we're doing throughout fair week or camps or anything. That might be, that's a challenge for me…

The difficulty of developing and implementing individualized supports or accommodations to maximize youth participation spanned across all 4-H contexts.

Parent Communication

A large factor contributing to being able to provide appropriate supports across all contexts was communicating with parents of youth on the autism spectrum. Some frustration with communication came from not knowing how to facilitate difficult conversations.

> And, and if we had like-we need to-I think, have more training on. You know. I don't know what how to handle a parent who doesn't want-who doesn't want to tell the educator to single a person out.

Breakdown of communication between parents and Extension Educators often intensifies the challenge of providing supports.

Working With Others

Extension Educators provide programs with a team that may have members with different perspectives, attitudes, and skills. These differences sometimes impact their ability to provide inclusive programs for their youth. One particular context that came up frequently was working with judges during 4-H fair activities. The participants shared that judges often lack knowledge about the challenges that youth on the autism spectrum might encounter during the fair process, leading to resistance to providing accommodations.

> When it–at the judging, ask my child a question the clock's turning and yet he has a problem with communication processing time. And the judge doesn't think he knows it, so he moves on to something else. [Yeah. Yeaaah] That-this really upsets me, that they don't understand processing time. The judges need to learn some of those simple facts. [Mmhmm. Yeah. I agree.].

The Extension Educators expressed that they often struggle to communicate expectations and accommodations appropriate for the individual child due to conflicts between expectations of the program, maintenance of youth confidentiality, and respect for youth independence.

Training Gaps

Although the culture of 4-H promotes inclusion, Educators mentioned difficulties actually implementing inclusive practices due to gaps in their skills. This also impacted their ability to support the volunteers who have direct contact with youth "…like with my volunteers now, they want to know what, they want to learn how to work with these kids. And that's something that I can't give them because I haven't been educated on it." Additionally, Extension Educators shared their experience of having negative feedback from club leaders/volunteers and parents on providing accommodations for youth on the autism spectrum. "I think there needs to be some training on how do we. How do you get that message across? [Yeah] That - Yes, adaptations are okay."

Training

Researchers asked participants to describe the training or education they have received on inclusion of youth on the autism spectrum. Participants described the training they received within Extension and beyond as well as the training opportunities they offered their volunteers.

Extension Training

Across both focus groups, all participants agreed that they received minimal official training on inclusion practices and strategies for children with disabilities through Extension. The only available training opportunities mentioned by the participants were webinars and presentations available through 4-H conferences that provide an overview of inclusion. The participants shared: "They weren't autism-specific. So sometimes you can find like those little nuggets, but they're usually like a 45-min webinar and it maybe skims the surface." Further "That's what I was gonna say it was kinda like [kinda touch the surface] yeah, brief, but it wasn't even really a training it was like be respectful of all human beings, be inclusive." Some described creating their own additional training opportunities with support from Extension.

> …we had our area retreat where all the 4-H educators got together and um. I was on the planning committee and we made it a priority to have someone come in and talk to us about working with youth on the autism spectrum.

Non-extension Training

Many of the participants in the focus groups reported receiving training outside of Extension through (a) Individual initiatives to educate themselves on how to work with children with disabilities. "I've also attended sessions at the Indiana Youth Institute conference. Again, not Extension sponsored but - umm, we can attend it"; (b) previous education received as teachers or in classes during their degrees in special education; (c) their initiatives to educate volunteers, counselors, and themselves "…we've had guest speakers on working with kids with disabilities including autism, so I guess we've learning along with them in that type of training that we've provided that's bee like 30–45 min…"; (d) sharing experiences and knowledge with other educators and transferring the knowledge to counselors and volunteers. "I've really learned a lot from my fellow Educators." As these types of learning opportunities were not offered by Extension leadership, they required motivation and initiative from the Educators to pursue.

Volunteer Training

Extension Educators provide training for other people who also work to directly support 4-H programs. The training they reported providing included: (a) sharing personal experiences and expertise and (b) formal and informal training sessions from Educators or service providers.

> So, I have brought in [community service provider] to do a training with my volunteers on inclusion. And they've done, I've had them come one time and it was, I think I had like thirty-five volunteers at it. So, it was highly attended, and it's something needed.

Overall, these formal sessions were typically short, but they were offered regularly and sometimes became a learning opportunity for Educators as well.

Active Strategies

Extension Educators were actively working with youth on the spectrum within their 4-H programs. They describe several types of strategies they already used to promote inclusion.

Individualizing

Extension Educators described creating individualized supports that focus on the youth's specific needs vs. their label." …so I think sometimes we didn't look at it as autistic. We just looked at what do we need to do to make this work." Reported individual supports included accommodations like extra time, creating written documents in place of oral presentations, and presenting projects early, before the arrival of the audience. Financial support to cover fees was also provided by 4-H and community members.

Mentor Mentoring

Further, Extension Educators discussed how they prepare club leaders, volunteers, counselors, and judges for working with and providing accommodations for youth on the autism spectrum. "… and so it helps for me to say: 'hey, here's what's coming, here's how we can help them out." Their goal is to "set them up for success" to secure positive experiences. For example, "like behind the scenes like at a before a show or before a workshop or something just so everybody's clear that nobody's being rude or disrespectful or you know not ignoring you."

Peer Mentoring

As shared by the majority of the participants, the use of peer-mentoring support strategies is very common in the 4-H environment, particularly in the camp setting. They shared "I had 4-H kids that that would bond with him and help him and do it you know, and he, he did better with them than mom standing around." Participant responses highlighted the potential for peer mentoring when implemented with training and supports from Educators.

Outreach

To increase enrollment, Extension Educators shared several outreach strategies they use to help share the message of 4-H as a great opportunity for youth on the autism spectrum. These individual efforts included sharing through after-school programs, special education services, social media groups, and, the most common one, word of mouth. "You're never gonna go wrong with word of mouth [mhm], so um, I mean that's our best-is somebody telling how the 4-H program has influenced them and their children and been a positive impact."

New Supports, Resources

Participants were asked to share their input on what types of new resources, strategies, and supports could improve their ability to provide inclusive programming for youth on the spectrum.

Increase Enrollment

Extension Educators shared strategies they believed would help to increase enrollment of youth on the autism spectrum. The participants agreed on the need for a change in recruitment language to advertise what 4-H has to offer to everyone including families with children with special needs.

> …he's like 80% of the parents are just gonna toss it cause they'll be like it's not really for my kid. Getting words on paper that will actually resonate with parents and youth on the autism spectrum or whatever population you're trying to reach that would make them say "nope 4-H is really for my kid and I do want to be here."

They further suggested using examples and promoting how youth on the autism spectrum can enjoy 4-H by utilizing parents who have lived the situations, they shared:

> It seems to be person to person, so we're trying to develop a network of parents talking to other parents about why-why do they have their children in 4-H. Spread that word with your circle of other parents and, and help them see where their child would fit in.

They also suggested creating other forms of recruitment and advertisement such as videos and brochures.

New Strategies

Across both focus groups, there were several suggestions for new strategies or expanded strategies that could help improve inclusion for youth on the autism spectrum.

Resource Portfolio

Educators shared that they would like to have a "portfolio" of accessible and appropriate accommodations, supports, and resources. Ideally, these resources would include materials for Extension Educators and also include materials they could share with volunteers and parents. "And if just more resources to give [volunteers] and to help them um work with the kids that cause like you said, [name], they have more direct contact than we do." This portfolio could also include direct access to experts with both professional and personal experience.

> . . . a m-mom or a dad or a guardian of some sort you know I can reach out to that I can ask those questions to and I guess have that honest conversation of this is what I'm thinking about doing. Is this inclusive or not inclusive in your mind?

Some suggestions that would fit within a new resource portfolio related to creating sensory-friendly environments.

> Where in our buildings are there space for them to go where they can just have their moment and refocus and come back? We also need to think about-just those learning environments. The lighting that we have [mhm], and things you know, you know those are things we need to think about and those are things we need to be trained on and then be able to take back to our volunteers. And you know, just make them aware and do their best to accommodate those possible.

Ideally, this would be a resource that Educators could access as needed and continue to build.

Connections With Parents

Multiple participants mentioned the need to connect more with parents through spending time in open conversation to assess youth needs. In addition to conversations with parents, participants suggested being more "proactive than reactive" by clearly stating in all early communications with parents that 4-H is inclusive and being open to communication about individual youth needs. One Extension Educator also emphasized the importance of communicating with each parent because of the range of unique experiences and needs.

> I think that most of the parents would be more than willing to share their story and to share um their needs, but what you're going to get from one parent is their specific experience. So it's really important to um gather more.

Increasing communication with parents could help prepare youth for new situations by providing them and their families with detailed information about what to expect.

Flexibility

In the context of talking about 4-H-related paperwork requirements, one Educator suggested a need to change program expectations. "But just like as a program we, that are tradition or that we always do. Just making sure that we know like it's OK if it's not done the way we think it's going to be done." This would be a shift toward more flexibility in the traditional 4-H requirements.

Mentoring

Participants identified peer mentors as an underutilized resource. As opposed to an adult telling them "what do and how to do it," youth-to-youth support has the potential to increase engagement. Another participant stated, "We have to have volunteers who understand [Mmhmm] it who can relay the message to the kids to help them understand." Participants acknowledged that to better implement mentoring this would take support throughout the tiered leadership structure.

New Trainings

Extension Educators provided several recommendations for new trainings related to inclusion.

New Content

Participants expressed a need for more training focused on comprehensive education that addresses the benefits of inclusion for everyone involved in 4-H. Specific suggestions included tangible information about strategies and effective accommodations. Participants also discussed the need for training, with practice opportunities, focused on strategies for communication with parents, youth, and others associated with 4-H (e.g., volunteers and judges). As one participant shared, "I think scenarios of role-playing would be helpful cause in any I don't care what we're doing, but the more practice we have at it the better." So not only the amount of training but also the range of content and the delivery of that content needs to be addressed with new resources.

Train the Trainer

Extension Educators are responsible for supporting and training club leaders and other volunteers with 4-H, thus, several participants highlighted the need for training materials they could implement and share throughout their existing leadership structure. Materials that eventually reach club leaders, camp counselors, and other volunteers are essential because as one participant said, "The first face [youth] will likely see is their club leader and not ours."

Stakeholder Involvement

Several participants expressed the importance of "lived" experience for people developing and delivering trainings. As one participant reflected,

> Just in this room. We've all talked about kids that have been in our program maybe that parents are a good presenter, maybe that grandparent of a - of that child is also for 4-H [Mhmm] could be good presenters. But I think we really need presenters that understand what we do that understand the program and understand the good days and the challenging days. Because I think I think, that's where you walk away from that training feeling that you've got something that - that you can use on a day to day basis.

The inclusion of stakeholders who have experiences working and living with people with disabilities as well as knowledge of 4-H would enhance the impact and effectiveness of new trainings for Extension Educators and the families they serve.

DISCUSSION

The goals of this study were to identify barriers to enrollment, to understand the current challenges of Extension Educators and youth on the spectrum, and to inform the development of resources to improve access and inclusion for youth on the spectrum in 4-H. To accomplish these goals, two focus groups were conducted with Extension Educators who support 4-H programs throughout Indiana. Several areas of need, as well as the potential for new resources, emerged. Although the mission of 4-H as an inclusive program is promoted and there is great willingness from Extension Educators, there is a lack of effective training to prepare those supporting 4-H to deliver fully inclusive programing, which aligns with previous reports (28–31). Additional challenges, active strategies, and potential for new resources were identified.

Two major challenges related to enrollment were awareness and 4-H culture. Despite active strategies on the behalf of Educators that included connecting with new families through school services, social media, and word of mouth, Educators conveyed a lack of knowledge in the general community that 4-H is an inclusive program and that there is a wide range of learning opportunities available for youth. Participants also highlighted the daunting task of navigating the processes and procedures involved with 4-H, especially for families that are new to the program. These results indicate a need for messaging across all levels of 4-H to highlight the diversity of programs available and emphasize that all youth, including those with disabilities, are welcome to participate. Many Extension Educators emphasized the effectiveness of personal connections in encouraging new families to try 4-H; however, this strategy is concentrated and time-consuming. Building up the outreach network to maximize the reach of current 4-H families, school staff, and other service providers could help reach more new families. Additionally, 4-H programs may benefit from building in more support and flexibility during the onboarding process. As current numbers of youth with disabilities enrolled within 4-H are unknown, future studies would need to closely track enrollment numbers.

Once enrolled in the program, Educators noted that youth on the autism spectrum face specific challenges. The most common context for challenges noted was 4-H fairs. The sensory environment, social demands, and rigidity of the rules for participation create a difficult situation for youth to navigate, which all relate to common themes of environmental factors that hinder the participation of youth on the autism spectrum (32). This context highlights the need for individualized accommodations, increased flexibility, and better communication between parents, youth, and 4-H personnel.

Some similar needs emerged in the analysis of challenges specific to Extension Educators. In particular, communication with adults involved in 4-H and parents of youth was highlighted as creating difficulties in developing and supporting appropriate inclusive programming. Regular communications between parents and Extension Educators that provide space for all parents of youth participating in 4-H to share information about the needs of their children, could help facilitate better communication without requiring parents to disclose their child's diagnosis (33).

The focus group data provided useful information about additional resources that could help improve the inclusiveness of 4-H programming. Extension Educator views reflected the consensus across many reports related to the inclusion of youth with disabilities, that there is a major need for more training and resources to effectively include youth with disabilities in 4-H programs (31). New materials to meet this need would best fit into the current model of 4-H if they were designed for Extension Educators as the primary consumer, which they would then share with volunteers and other 4-H supporters who work directly with youth. A train-the-trainer model would not only fit with the current 4-H structure but has also demonstrated effectiveness in disseminating evidence-based practices in community settings (34). Additional training resources could increase the number of Educators who provide training for club leaders and volunteers (18). Content and trainings should all be developed with input from people with direct personal experience with both 4-H and autism. Trainings and resources that are more generalized and not focused on the label of autism would align with the desire that youth not be singled out. This type of need-focused vs. specific label-focused content could also benefit children with other types of disabilities (e.g., ADHD), who are also impacted by the lack of training on disability inclusion (29).

Limitations

Findings from this study should be reviewed in light of a few limitations. The focus groups included only Extension Educators. Additional challenges, needs, and ideas for new resources could be identified if the focus groups included additional stakeholder groups. Future work will include 4-H volunteers, parents, and youth perspectives. Additionally, some of the participating Extension Educators had close ties to people on the autism spectrum. This experience may have increased support for inclusive practices and also provided perspectives that are not reflective of the perspectives of those Educators without personal connections to autism. Focus group participants were also exclusively from Indiana. Although the basic structure of 4-H across the Unites states is similar, Extension, and subsequently 4-H, does have some state-level variations which may be noted if this study were to be replicated across states.

Conclusions

The structure and purpose of 4-H naturally lends itself to the inclusion of all youth, including those on the autism spectrum. Thus, 4-H has the potential to provide opportunities for skill-building, social-communication practice, and the development of friendships for youth on the autism spectrum within a natural environment. Maximizing this experience necessitates additional training, increased channels of communication between parents and extension educators, and identification of mechanisms for the provision of individualized supports and accommodations to promote positive experiences. With increased support, 4-H and programs like it could address the service needs of youth on the spectrum, particularly in rural areas. As identified in this analysis, perspectives and involvement of stakeholders with personal connections to autism and 4-H are needed to develop new resources that will address youth needs and maximize the positive impact of 4-H.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Purdue University Institutional Review Board. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

CM is the principal investigator and lead author. CM and VP conducted the focus groups. CM, RM, VP, EC, and AM contributed to the data analysis process. All authors contributed to the writing of the manuscript.

FUNDING

This study was funded by the Agricultural Science and Extension for Economic Development (AgSEED) program at Purdue University's College of Agriculture. CM received support from the National Center for Advancing Translational Sciences (KL2TR002530 and UL1 TROO2529).

ACKNOWLEDGMENTS

We want to thank Extension leadership for their support and all of the Extension Educators who shared their experiences with us. We would also like to acknowledge the following students who assisted with this manuscript: Sungwoo Kang, Mackenzie Neher, and Elise Oquendo.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2022.913600/full#supplementary-material

REFERENCES

1. American Psychiatric Association. *Diagnostic and Statistical Manual of Mental Disorders (DSM-5®).* American Psychiatric Publishing, Inc. (2013). doi: 10.1176/appi.books.9780890425596
2. MacCormack JWH, Matheson IA, Hutchinson NL. *An Exploration of a Community-Based LEGO® Social-Skills Program for Youth with Autism Spectrum Disorder.* eei (2015). p. 25. Available online at: https://ojs.lib.uwo.ca/index.php/eei/article/view/7729 (accessed March 23, 2021).
3. Roux AM, Rast JE, Anderson KA, Shattuck PT. *National Autism Indicators Report: Developmental Disability Services and Outcomes in Adulthood.* PA: Life Course Outcomes Program, AJ Drexel Autism Insitute, Drexel University (2017).
4. Pellicano E, Dinsmore A, Charman T. What should autism research focus upon? community views and priorities from the United Kingdom. *Autism.* (2014) 18:756–70. doi: 10.1177/1362361314529627
5. Roche L, Adams D, Clark M. Research priorities of the autism community: a systematic review of key stakeholder perspectives. *Autism.* (2021) 25:336–48. doi: 10.1177/1362361320967790
6. Pickard KE, Ingersoll BR. Quality versus quantity: the role of socioeconomic status on parent-reported service knowledge, service use, unmet service needs, and barriers to service use. *Autism.* (2016) 20:106–15. doi: 10.1177/1362361315569745
7. Taylor JL, Henninger NA. Frequency and correlates of service access among youth with autism transitioning to adulthood. *J Autism Dev Disord.* (2015) 45:179–91. doi: 10.1007/s10803-014-2203-x
8. Mello MP, Goldman SE. Services for children with autism spectrum disorder: comparing rural and non-rural communities. *Educ Train Autism Dev Disabil.* (2016) 54:355–65. Available online at: https://www.jstor.org/stable/26173863
9. Murphy MA, Ruble LA. A comparative study of rurality and urbanicity on access to and satisfaction with services for children with autism spectrum disorders. *Rural Spec Educ Q.* (2012) 31:3–11. doi: 10.1177/875687051203100302
10. Koegel LK, Vernon TW, Koegel RL, Koegel BL, Paullin AW. Improving social engagement and initiations between children with autism spectrum disorder and their peers in inclusive settings. *J Posit Behav Interv.* (2012) 14:220–7. doi: 10.1177/1098300712437042
11. Hepburn SL. *Strengthening Informal Supports to Promote Behavioral Health of Youth With Intellectual and/or Developmental Disabilities in Rural Communities. In: International Review of Research in Developmental Disabilities.* Elsevier (2017). p. 203–34. Available online at: https://linkinghub.elsevier.com/retrieve/pii/S2211609517300039 (accessed March 24, 2021).
12. Bohnert A, Lieb R, Arola N. More than Leisure: organized activity participation and socio-emotional adjustment among adolescents with autism spectrum disorder. *J Autism Dev Disord.* (2019) 49:2637–52. doi: 10.1007/s10803-016-2783-8
13. Reis SM, Gelbar NW, Madaus JW. Understanding the academic success of academically talented college students with autism spectrum disorders. *J Autism Dev Disord.* (2021) 1–14. doi: 10.1007/s10803-021-05290-4
14. Larson RW. Toward a psychology of positive youth development. *Am Psychol.* (2000) 55:170. doi: 10.1037/0003-066X.55.1.170
15. Ratcliff K, Hong I, Hilton C. Leisure participation patterns for school age youth with autism spectrum disorders: findings from the 2016 national survey of children's health. *J Autism Dev Disord.* (2018) 48:3783–93. doi: 10.1007/s10803-018-3643-5
16. Agran M, Wojcik A, Cain I, Thoma C, Achola E, Austin KM, et al. Participation of students with intellectual and developmental disabilities in extracurricular activities: does inclusion end at 3:00? *Educ Train Autism Dev Disabil.* (2017) 52:3–12. doi: 10.1037/0012-1649.44.3.814
17. Carter EW, Swedeen B, Moss CK, Pesko MJ. "What are you doing after school?": promoting extracurricular involvement for transition-age youth with disabilities. *Interv School Clin.* (2010) 45:275–83. doi: 10.1177/1053451209359077
18. Stair KS, Seevers BS, Moore A. State 4-H leader's perceptions of involvement of youth with special needs in the 4-H program. *JYD.* (2012) 7:47–58. doi: 10.5195/JYD.2012.128

19. Anaby D, Hand C, Bradley L, DiRezze B, Forhan M, DiGiacomo A, et al. The effect of the environment on participation of children and youth with disabilities: a scoping review. *Disabil Rehabil.* (2013) 35:1589–98. doi: 10.3109/09638288.2012.748840
20. Tormoehlen R, Field W. A perfect fit: involving youth with disabilities in 4-H. *J Ext.* (1994) 32:1FEA4.
21. Goble CL, Eyre NS. *Incorporating Special Needs Youth into 4-H. p.* 3.
22. Stumpf-Downing M, Henderson K, Luken K, Bialeshki D. Creating inclusive 4-H environments for people with disabilities. *J Ext*. (2004) 42:4FEA1. Available online at: https://archives.joe.org/joe/2004august/a1.php
23. LaVergne DD. Perceptions of 4-H professionals on proposed solutions towards diversity inclusive 4-h youth programs. *JYD.* (2015) 10:139–51. doi: 10.5195/JYD.2015.426
24. Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *Int J Qual Health Care.* (2007) 19:349–57. doi: 10.1093/intqhc/mzm042
25. Saldana J. *The Coding Manual for Qualitative Researchers*. 3rd ed. Los Angeles, CA: Sage Publications Inc. (2016).
26. Corbin J, Strauss A. *Basics of Qualitative Research: Techniques and Procedures for Developing Grounded Theory*. 4th ed. Los Angeles, CA: Sage Publications, Inc. (2015).
27. Braun V, Clarke V. *Thematic Analysis: A Practical Guide*. Los Angelos, CA: Sage (2022).
28. Mouton L, Bruce J. Current practices for training staff to accommodate youth with special health care needs in the 4-h camp setting. *J Ext*. (2013) 51:7. Available online at: https://tigerprints.clemson.edu/joe/vol51/iss1/33
29. Mpofu C, Ingram PD, Radhakrishna R. Perceptions of 4-H Extension Educators and Volunteer Leaders toward the Inclusion of Youth with Attention Deficit Hyperactive Disorder(s) in 4-H Programs. *JYD*. (2010) 5:16–28. doi: 10.5195/JYD.2010.227
30. Peterson R, Grenwelge C, Benz MR, Zhang D. Serving clientele with disabilities: An assessment of TEXAS FCS agents' needs for implementing inclusive programs. *J Ext.* (2012) 50:7. Available online at: https://tigerprints.clemson.edu/joe/vol50/iss6/7
31. Taylor-Winney J, Xue C, Mcnab E, Krahn G. Inclusion of youths with disabilities in 4-H: a scoping literature review. *J Ext.* (2019) 57:12. Available online at: https://tigerprints.clemson.edu/joe/vol57/iss3/22
32. Krieger B, Piškur B, Schulze C, Jakobs U, Beurskens A, Moser A. Supporting and hindering environments for participation of adolescents diagnosed with autism spectrum disorder: a scoping review. Webster AA, editor. *PLoS ONE.* (2018) 13:e0202071. doi: 10.1371/journal.pone.0202071
33. Steinhardt F, Ullenhag A, Jahnsen R, Dolva AS. Perceived facilitators and barriers for participation in leisure activities in children with disabilities: perspectives of children, parents and professionals. *Scand J Occup Ther*. (2021) 28:121–35. doi: 10.1080/11038128.2019.1703037
34. Shire SY, Kasari C. Train the trainer effectiveness trials of behavioral intervention for individuals with autism: a systematic review. *Am J Intellect Dev Disabil*. (2014) 119:436–51. doi: 10.1352/1944-7558-119.5.436

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers | Frontiers in Psychiatry

ORIGINAL RESEARCH
published: 03 June 2022
doi: 10.3389/fpsyt.2022.913142

Positive Effects of the Caregiver Skill Training Program on Children With Developmental Disabilities: Experiences From Serbia

Nenad Glumbic [1]*, ***Roberto Grujicic*** [2], ***Sanja Stupar*** [2], ***Suncica Petrovic*** [3] **and** ***Milica Pejovic-Milovancevic*** [2,4]

[1] Faculty of Special Education and Rehabilitation, University of Belgrade, Belgrade, Serbia, [2] Department for Children and Adolescents, Institute of Mental Health, Belgrade, Serbia, [3] Serbian Society of Autism, Belgrade, Serbia, [4] Faculty of Medicine, University of Belgrade, Belgrade, Serbia

Edited by:
Andy Shih,
Autism Speaks, United States

Reviewed by:
Branko Aleksic,
Nagoya University, Japan
Ahmed Mohammed Saleh Alduais,
University of Verona, Italy
Milena Nikolić,
University of Tuzla, Bosnia and Herzegovina

****Correspondence:***
Nenad Glumbic
nenadglumbic@gmail.com

Specialty section:
This article was submitted to Autism, a section of the journal Frontiers in Psychiatry

Received: *05 April 2022*
Accepted: *16 May 2022*
Published: *03 June 2022*

Citation:
Glumbic N, Grujicic R, Stupar S, Petrovic S and Pejovic-Milovancevic M (2022) Positive Effects of the Caregiver Skill Training Program on Children With Developmental Disabilities: Experiences From Serbia. Front. Psychiatry 13:913142. doi: 10.3389/fpsyt.2022.913142

Background: Intervention programs for children with developmental disabilities increasingly target caregiver training to implement effective strategies for child development. Research conducted in different countries shows that the Caregiver Skills Training Program (CST) developed by the World Health Organization and Autism Speaks could also be a recommended intervention.

Methods: The pre-pilot phase included seven, and the pilot phase included 29 families of children with developmental disabilities trained to implement the intervention program. The caregivers were asked to complete the Autism Treatment Evaluation Checklist at the beginning and at the end of the program.

Results: In the pre-pilot phase, the Wilcoxon signed-rank test determined a statistically significant improvement in Speech, Language and Communication ($z = -2.99$, $p < 0.05$) and Health/Physical/Behavior ($z = -2.375$, $p < 0.05$) after caregiver participation in the training program. In the pilot phase, the paired t-test also determined a statistically significant improvement in Speech, Language and Communication between the first ($M = 24.52$, $SD = 5.57$) and the second testing ($M = 25.66$, $SD = 6.11$), $t_{(28)} = -2.29$, $p < 0.05$, as well as a significant improvement between the first ($M = 36.62$; $SD = 7.15$) and the second testing ($M = 35.38$; $SD = 5.91$), $t_{(28)} = 2.11$, $p < 0.05$ in Health/Physical/Behavior. Eta squared values (0.16 and 0.14) indicate that the intervention effect was significant. No differences were determined in Sociability and Sensory/Cognitive Awareness between the first and the second testing.

Conclusion: The initial results of the Caregiver Skills Training Program are encouraging. For this program to be recommended as an evidence-based intervention, further research should be conducted on larger samples, controlling possible intervening variables.

Keywords: developmental delays, parent-mediated intervention, support, parenting skills program, parent-child relations, public health, Serbia

INTRODUCTION

Children with developmental delays (DD) need continuous lifelong support, from birth to adulthood. In many countries, especially in low and middle-income countries (LMIC), adequate services are poorly accessible or completely inaccessible to this vulnerable group (1). The children with DD from LMIC usually deprived of early recognition and community-based interventions, but sometimes also inclusive and adequate education, health care, evidence-based interventions, and rehabilitation (2). Moreover, the support of caregivers and the whole family is equally important, especially for families with restricted access to such services (3). Interventions implemented in all resources must be evidence-based and tailored for children with DD. Out of all implemented strategies and methods for children with DD, early recognition and timely, evidence-based, individual-oriented interventions have performed best (2, 4). In order to provide a timely diagnosis and refer children to a certain intervention, all service providers for children and families (e.g., healthcare providers, social workers, teachers and employees in the educational system), need to be educated and trained.

However, sometimes even with appropriate support systems and care providers, there is not a sufficient improvement in child's behavior and development. This can happen in cases where experts are mainly focused on delivering interventions to children, neglecting the role of caregivers in overall treatment (5, 6). All service and care providers should recognize the fact that the caregivers of children with DD spend the most time with their child and can thus carry out most of the treatment and support. In contrast, if there is no cooperation with parents, relatively little can be achieved in terms of improving the child's wellbeing and independency (6). Therefore, the caregivers of children with DD should be provided with continuous information and assistance in understanding the diagnosis, as well as be empowered to participate in the decision-making process for intervention design; parents should be given a chance to acquire the skills needed to help in the process of the child's treatment.

Serbia is a country of ∼7 million people where about 1 million people are aged 0–17 (7). Like most of the LIMC, Serbia has a significant problem in providing appropriate, evidence-based mental health care for children with DD, mostly due to the lack of staff, research and infrastructure (8). Even if the needed support is available in certain areas, families face additional problems, including restricted access, financial issues, time constraints, etc. (3, 9). In rural areas the problem is most visible. Despite the fact that approximately half of the population resides in rural areas, almost all kinds of professional support for children with DD are inadequate (9).

One of the most recognized parent-based education programs to date is Caregivers' Skills Training (CST) developed by Autism Speaks (AS) and the World Health Organization (WHO) (10). The training is intended for parents and caregivers of children with DD. This program offers the opportunity for caregivers to fully participate in their child's treatment, as well as to acquire skills for supporting their child's development (10). The program consists of nine group sessions and three individual home visits. The training is designed to teach the caregiver how to use every day play and home routines as opportunities for learning and development (11). This program was initiated in Serbia as part of the overall initiative to strengthen the Early Childhood Intervention (ECI) system in the country. Care providers and policy-makers in Serbia recognized the importance of a family-focused approach and the vital role of caregiver education in the field of ECI. Recognizing the potential role of CST, the professionals involved in ECI programs welcomed it enthusiastically.

To date, the CST program has been implemented, or is in the process of implementation, in more than 30 countries all over the world, and many of these countries are LIMC (12). The CST has shown to be a promising intervention in LIMC due to the fact that it doesn't require the involvement of specialists or other high profile experts (it can be even delivered by educated parents or volunteers), it doesn't require high material/financial resources or expensive equipment. The studies conducted in these countries indicate that CST could be the intervention which can be implemented, culturally adapted and sustainable for many LIMC (13–15).

The main aim of this research is to evaluate the effects of the CST program on different domains of child's development. In addition, this research aims to evaluate the process of implementation, cultural appropriateness and parental opinions on the CST program in Serbia.

MATERIALS AND METHODS

Participants

The CST program in families of children with developmental disabilities was conducted in a pre-pilot and a pilot phase. The pre-pilot phase included seven and the pilot phase 29 families. There was one child with developmental disabilities in each family. **Tables 1**, **2** show demographic data on the caregivers and children with developmental disabilities from both research phases.

Five boys and two girls, 4–8.5 years of age (M = 5.96; SD = 1.61), participated in the pre-pilot phase, while 20 boys (69%) and nine girls (31%), 2–9 years of age (M = 4.11; SD = 1.92), participated in the pilot phase of the study. Both research phases included a convenience sample of caregivers who requested health service support due to developmental disabilities in children (most frequently autism spectrum disorders - ASD, mixed specific developmental disorder, and communication disorder). At the time of research, three children showed significant developmental delay but had not yet been diagnosed.

Procedure

The program was divided into two consecutive phases: pre-pilot and pilot phases. In the first (pre-pilot) phase, the master trainers were trained and delivered the training to the parents. 'Master trainers' is a term determined by the program which describes experts who are in charge of implementation, supervision and delivery of the program in their country. After their extensive training by the founders of CST, the master trainers are able to deliver the CST to other experts in their country (in later text: facilitators) who will continue their work with the families

TABLE 1 | The socio-demographic data of caregivers.

	Pre-pilot (*n* = 7) *n*	Pilot (*n* = 29) *n* (%)
Main caregiver		
Mother	5	27 (93.1)
Father	2	2 (6.9)
Age M (SD)	40.29 (6.95)	35.76 (5.2)
Education		
Primary	0	1 (3.4)
Secondary	2	13 (44.8)
University	5	15 (51.7)
Marital status		
Single	0	1 (3.4)
Married (partner)	6	26 (89.8)
Divorced	1	1 (3.4)
Widowed	0	1 (3.4)
Place of living		
Rural	1	6 (20.7)
Urban	6	23 (79.3)
Working outside the home		
No	2	11 (37.9)
Part-time	0	4 (13.8)
Full time	5	14 (48.3)
People who are cared for		
Children M (SD)	2.14 (1.17)	1.62 (0.6)
Adults M (SD)	0	0.14 (0.5)

TABLE 2 | The demographic data of children.

	Pre-pilot (*n* = 7) *n*	Pilot (*n* = 29) *n* (%)
Sex		
Male	5	20 (69.0)
Female	2	9 (31.0)
Age M (SD)	5.96 (1.6)	4.11 (1.1)
Diagnosis		
Intellectual disability	0	1 (3.4)
Communication disorder	1	5 (17.2)
Autism spectrum disorders	4	8 (27.5)
Motor disorder	0	2 (6.9)
Cerebral palsy	0	2 (6.9)
Mixed specific developmental disorder	2	7 (24.1)
Developmental delay	0	1 (3.4)
Received support		
Health professional (medication)	2	8 (27.6)
Health professional (no medication)	5	9 (31.0)
Special education	6	16 (55.1)
Speech therapy	6	14 (48.2)
Behavioral therapy	0	5 (17.2)
Information on child's problem	2	0 (0.0)
Information on services	0	4 (13.7)

under supervision. Each country has a certain number of trained master trainers who are usually experts in child mental health and development. In our country, eight master trainers (six women and two men), 28–49 years of age (M = 39.25; SD = 8.515), participated in the training program. All master trainers were experts in mental health or health associate professionals in primary healthcare. Two of them were employed in private healthcare institutions, and six worked in public healthcare institutions. Given the differences in their age, the master trainers were expected to have different work experience ranging from four to 22 years. Also, they spent between two and 22 years working with children. Two master trainers had no previous experience in training parents, while six did.

In the second (pilot) phase, each master trainer trained two facilitators to conduct the program with the families. After their training, the facilitators delivered the program to the families under the supervision of a master trainer. Apart from one facilitator who worked in a public education institution, all others were employed in a public healthcare institution.

In each phase, the program was comprised of nine parental group sessions with three home sessions.

Instruments

All instruments and measures created for CST and used in this research were recommended and provided by the WHO and AS. The measures were translated, back-translated and adapted to Serbian language.

Background Measures

Information on master trainers, caregivers, and facilitators, partially presented in the Participants and Procedures sections, was collected using CST background measures: Demographic and professional background information for facilitators and Information about caregivers: Demographic and service history information.

CST Program Implementation Measures

The Post session Feedback Form—Caregivers and the Home Visit Participant Feedback Form were used to assess the implementation of the CST program. After each session, the caregivers completed the Post session Feedback Form stating their observations and impressions. The Form included 14 questions requiring the caregivers to assess the degree of understanding, the relevance of the information received, the usefulness of key messages and tips, the usefulness of advice in achieving their own program goals, learning activities, duration of the session and its parts, etc.

In addition to expressing their views about the sessions, caregivers were asked to share their experiences after each home visit with master trainers/facilitators by completing the Home Visit Participant Feedback Form. This Form included only five questions, requiring caregivers to assess the duration and benefits of the home visit, and the benefits of the feedback received during the recording. Open-ended questions were used to collect additional information about whether video-recording was acceptable from the caregivers' perspective and their suggestions for improving home visits.

Outcome Measures

The Autism Treatment Evaluation Checklist (ATEC) (16) was used at the beginning and the end of the program to determine the effects of the CST program on children with developmental disabilities. ATEC consists of four subtests. The first subtest, Speech/Language/Communication, includes 14 items. The given answers are 1—not true, 2—somewhat true, or 3—very true, and a higher score indicates better-developed communication skills. The Sociability subtest includes 20 items with three possible answers: 1—not descriptive, 2—somewhat descriptive, and 3—very descriptive. The same answers are provided for the Sensory/Cognitive Awareness subtest that includes 18 items. The difference is that a higher score indicates greater difficulties on the Sociability subtest, while it indicates better-developed skills on the Sensory/Cognitive Awareness subtest. The last subtest, Health/Physical/Behavior, includes 25 items graded on a four-point Likert scale (1—not a problem, 2—minor problem, 3—moderate problem, 4—serious problem).

Statistical Analysis

The statistical analysis was performed in IBM SPSS Statistics version 27. Due to the small sample in the pre-pilot research phase, the difference in the participants' achievements on ATEC between the first and the second testing was determined by the Wilcoxon signed-rank test. The paired samples t-test was used to compare the first and the second test results in the pilot phase. Repeated measures two-way ANOVA was used to determine whether the participants' scores on ATEC in the pilot phase differed depending on whether the program was implemented in person or online. The repeated factor tested the difference between the two measurements, and the factor that was not repeated referred to the difference in the way the training was implemented.

Ethics

This research has been approved by the Ethics Committee of the Institute of Mental Health in Serbia. All participants have signed the Informed Consent Form prior to inclusion in the research.

RESULTS

The main aim our research was to evaluate the effects of the CST on child's development and to evaluate the appropriateness of the program in our country. To achieve this, we collected data from caregivers in two subsequent phases on their attitudes toward the program as well as their views on how the program effected their child's development. In this section we will present the obtained quantitative and some qualitative data.

Parental Feedback

The data obtained using the Post session Feedback Form—Caregivers, after each session, and Home Visit Participant Feedback Form, after each home visit, could only be processed descriptively. All scores were very high. Thus, due to the small variability of the obtained data, the satisfaction with individual sessions and home visits was not observed as a control variable in further analysis of the training outcomes. The degree of understanding, the relevance of the information received, the usefulness of key messages and tips, and the usefulness of advice in achieving program goals were assessed by a five-point Likert scale in the Post session Feedback Form. The average scores in both research phases ranged between 4.29 and 5.00. Participants listed different key messages and tips they considered the most or the least significant in each session, but the general tendency was to choose the first ones in a line. With regard to learning activities, group discussion and demonstration were most often mentioned as the favorite ones. More than 80% of the participants positively evaluated the duration of individual sessions and activities. Over 90% of the caregivers felt very or somewhat willing to apply the advice received during sessions at home. Depending on the session, between 80 and 95% of the participants stated that the messages they received during sessions did not contradict what they or their family members considered good or significant.

In addition to expressing their views on the sessions, the caregivers were asked to share their experiences with master trainers/facilitators after every home visit. Almost all participants stated that the home visit lasted as long as necessary, that it was useful, and that the feedback obtained by video recording was valuable. In both research phases, the caregivers were positive about the fact that video recording was an integral part of the program.

Effects on the Child's Development

The **Tables 3**, **4** show the scores on four domains of ATEC scale measured before and after the program. The four domains are as follows: Speech/Language/Communication, Sociability, Sensory/Cognitive Awareness and Health/Physical/Behavior. The **Table 3** shows the results from the families that participated in the pre-pilot phase ($N = 7$), while the **Table 4** shows the results from the families in the pilot phase ($N = 29$).

The Wilcoxon signed-rank test determined significant improvements in Speech/Language/Communication ($Z = -2{,}232$, $p < 0.05$) and Health/Physical/Behavior ($Z = -2{,}375$, $p < 0.05$), with a large effect size by Cohen's criteria ($r = 0.60$ for Speech/Language/Communication and $r = 0.63$ for Health/Physical/Behavior).

Similar results were obtained in the pilot phase on a somewhat larger sample. Significant improvements were found in Speech/Language/Communication ($t = -2.296$, $p < 0.05$, Eta squared $= 0.16$) and Health/Physical/Behavior ($t = 2.109$, $p < 0.05$, Eta squared $= 0.14$), while there were no significant differences between the first and the second testing on the remaining two subscales. Eta squared values indicate that the intervention effect was significant.

Two-way ANOVA did not indicate the interaction between the two factors (repeated assessment and the way the program was implemented), both in Speech/Language/Communication ($F = 0.900$, $p = 0.351$, Eta squared $= 0.032$), and in Health/Physical/Behavior ($F = 0.854$, $p = 0.364$, Eta squared $= 0.031$). This suggests that the effects of the program did

TABLE 3 | Scores of children with developmental disabilities on ATEC at the beginning and the end of the CST pre-pilot program.

ATEC subscales	Baseline ($n = 7$)		After intervention ($n = 7$)		Z	p
	M	SD	M	SD		
Speech/language/communication	29.14	6.41	31.57	6.00	−2.232	**0.026**
Sociability	30.86	3.18	29.14	5.49	−1.382	0.167
Sensory/cognitive awareness	40.86	4.49	43.43	6.27	−1.572	0.116
Health/physical/behavior	40.29	14.47	36.86	13.80	−2.375	**0.018**

The bolded values indicate the values to be statistically significant ($p < 0.05$).

TABLE 4 | Scores of children with developmental disabilities on ATEC at the beginning and the end of the CST pilot program.

ATEC subscales	Baseline ($n = 29$)		After intervention ($n = 29$)		t	df	p
	M	SD	M	SD			
Speech/language/communication	24.52	5.57	25.66	6.11	−2.296	28	**0.029**
Sociability	30.31	7.19	29.14	6.41	1.177	28	0.249
Sensory/cognitive awareness	41.59	7.66	41.72	7.92	−181	28	0.857
Health/physical/behavior	36.62	7.15	35.58	5.91	2.109	28	**0.044**

The bolded values indicate the values to be statistically significant ($p < 0.05$).

not depend on whether the training was conducted in person or online.

DISCUSSION

This research has been conducted as part of the implementing process of the CST project in Serbia. This process was composed of two consecutive phases including the pre-pilot and pilot phases. In the first part of the pre-pilot phase, eight master trainers (MTs) were trained by the representative of AS. All MTs are experts in the field of mental health or health associates in primary health care. The training was supervised by the representatives of AS, who had many years of experience in the CST program implementation. The training of master trainers (TOMT) lasted for 5 days. It was conducted in person and included explaining procedures, key recommendations and advice, role-playing, modeling expected behavior, and direct work with patients. The other part of the TOMT was designed to deliver the whole CST program to the parents. In other words, the trained MTs delivered the whole program to the families of children with DD. Their activities were recorded, thus allowing the supervision of experts from AS.

The CST training of parents was provided in nine parental group sessions and three home visits. The entire training and research material (facilitator guides, home visit facilitator guides, participants booklets for each session, etc.) was translated into Serbian. Then, a material adaptation strategy was developed in direct meetings with the representatives of AS. The Adaptation Team and Adaptation Advisory Group were formed to work together on linguistic and cultural material adaptation. The Adaptation Advisory Group included experts in working with children with DD and neurodevelopmental disorders and their families.

In the pilot phase, the certified MTs trained the facilitators to implement training. The final goal was that the facilitators could then work directly with families of children with DD under the supervision of the MTs. The facilitators had different professions: nurses, educators, health associate professionals in primary healthcare, community nurses, mental health experts, etc.

Most of the respondents from both phases of the research were mothers. This was not surprising, as in Serbia childcare responsibilities are traditionally the job of mothers. Contrary to expectations, two participants in each research stage were fathers. The study that pre-piloted the CST in Ethiopia had a similar result with a low number of fathers (13). The literature review findings reveal a significant and positive impact of father involvement on the child's development, and especially the development of cognitive skills (17). For this reason, we should also aim to include more fathers in the program.

The majority of families who participated in the study were married parents. We do assume that married parents are more willing to participate in training and education processes as sharing caring responsibilities allows greater time for participation. However, we also need to highlight the importance of single-parent and divorced families that already have a significant childcare burden, and even more so if they are a single parent to a child with DD. This is especially the case with single mothers, who face additional burdens associated with gender inequalities typical in the region (e.g., financial problems, professional struggles, lack of time, lack of social support, etc.) (18). A United Nations Children's Fund (UNICEF) study from 2018 revealed that patriarchal gender norms in Eastern Europe and the Balkan region heavily influenced parenting, especially affecting single mothers (19). In the future, married families should not be the only focus of the program. We need to find a way to provide a chance for single parent families, and single mothers, to participate fully in the CST program.

This research was conducted in the centers that implemented the CST, all of which are located in urban areas (i.e., four biggest cities in Serbia). The resulting sample hence only included participants from urban surroundings. However, 46% of families in Serbia live in rural surroundings with very limited resources and access to health and social care (7). Because families from urban surroundings already have better access to services and treatments for children, more families of children with DD from rural surroundings need to be included in future caregiver trainings.

The outbreak of the COVID-19 pandemic during the training program affected the implementation of group sessions and home visits. During the pandemic, the CST program was adapted in order to help in switching the training to online setting (12). Some MTs and facilitators decided to continue the training online. Five families underwent in-person training in the pre-pilot phase, while two families participated in online training. However, as the virus spread over time, the need for online training increased. Only eight (27.6%) families had in-person training in the pilot phase, while 21 (72.4%) families used a video-conferencing system. Although the pilot phase was initially planned to last for 12 weeks, it was extended to 1 year due to the pandemic. As the CST program was not initially designed to be delivered online, some major adaptations had to be performed along the way. If it is to be fully digitalised, the program needs to be further adapted. Additionally, it is possible that families from rural surroundings will require support in handling fully digitalised modes of training. These adaptations were targeted at the delivery of practical exercises and group discussions which were not readily conductable in an online setting.

Most of the families included in the sample received some kind support, commonly in the form of special education, speech therapy and medical attention. In our previous research, we have shown that over ninety per cent of families of children with DD reported that additional support from all systems is important and necessary (3). Other studies confirmed our findings, showing that there is still a wide range of barriers that prevent families from accessing needed support, even with the implementation of strategies for increasing the accessibility of services in LIMC (9). We have also previously identified some predictors of bad overall satisfaction with the provided support, and one of the most significant predictors was parent frustration with accessing services. More positive feedback was gathered from parents who received assistance in managing their child's needs and who had access to an expert as a source of information on autism (3). The CST provides a chance for parents to acquire enough information both from experts and other parents in order to increase their confidence and gain new skills. Promising results from this study lead us to believe that this program can offer a long-term solution to the problem of restricted access and parental frustration in our country.

The majority of the participants evaluated the program positively, as very successful and important for them; the degree of understanding, the relevance of the information received, the usefulness of key messages and tips, and the usefulness of advice in achieving program goals were evaluated between 4.29 and 5.00. This feedback can be interpreted as an endorsement of the CST program, as very well-organized and informative and useful for parents. Our results indicate that there is now an extremely practical, step by step program in Serbia which allows parents to feel understood, appreciated, and respected. This program allows parents to feel as the most valuable actor in their child's treatment and not only a passive observer in the process. It provides parents with very high professional support, as well as with much needed emotional support. The research from other countries that implemented the CST reports similar results, especially in the LIMC (11, 13, 14).

Group discussions and practical exercises were most often voted as favorite parts of the training. Previous literature reviews show that group-based parenting programs show positive and promising results in improving emotional and behavioral functioning of young children as well as parental confidence, knowledge and wellbeing (20, 21). A new study conducted specifically on CST in Italy also shows promising results in the same context (11). From our experience, group support is crucial, mainly because parents can feel understood and accepted. This is a good starting point for future work with families that highlights the importance of organizing parental support groups of children with DD. However, it is important to be mindful of the duration of sessions; too short could be insufficient, and too long could be inaccessible for working parents. CST training obtained balanced timing and participants positively evaluated the duration of individual sessions and activities.

Even though the groups were small to be analyzed in more detail, the parents believe that CST helped them in improving Speech/Language/Communication and Health/Physical/Behavior of their children. This is not surprising, as the program is specifically designed to improve the above-mentioned domains of functioning; out of nine sessions, two sessions are dedicated to understanding and improvement of the child's communication, and two sessions for understanding and controlling the child's behavior. Research conducted on the CST program in other countries reports similar results (11, 13, 14). According to our previous research, the primary concern of parents in our country was related to the child's interaction and communication (3). This finding can also partly explain high rates of parental satisfaction in our county, as the program aims to target the associated concerns with interaction and communication.

Limitations

This research had several limitations. The first is a limited sample size. This was partly due to fact that this was an implementation (pilot) phase of the program which had a limited number of participants. In addition, due to the COVID pandemic outburst, the research suffered a certain dropout of families before the ending of the program. Namely, some families had technical difficulties, some had health-related issues, and some did not want to continue the program due to the shift of priorities in the time of crisis. All of this also resulted in a limited amount of collected data delivered to the research team.

The adaptation of the program in Serbia took place mainly in urban areas, in clinical settings and delivered mainly by professionals with long years of experience in working with families of children with DD. We are aware that delivering the program in rural settings by non-specialists would possibly have different benefits and recommendations, but also possible additional challenges that were not discovered in this round of research.

CONCLUSION

Caregiver's education is a process of systematic provision of necessary information to caregivers which can provide them with the specific knowledge and skills for encouraging their child's development and competence. In the past decade, some systematic reviews, mostly from high-income countries, suggest that caregivers are able to learn and acquire the skills needed to deliver intervention strategies to their children with DD (22, 23). Education and mastering specific skills can significantly improve the mental health of caregivers, the caregiver-child relationship and the process of adapting to new conditions (5, 24). Reviews show that families can significantly benefit from these types of interventions even if they are short and of low intensity (2, 25, 26).

With the introduction of the CST program in Serbia, local partners build the capacities of relevant institutions and professionals. This type of knowledge and skills will be applicable in working with all families of children with disabilities, so that professionals educated through this program remain a permanent resource for participating institutions. The pilot project is implemented in three cities in Serbia (Belgrade, Novi Sad, Nis), in four different health care institutions (primary and tertiary) dealing with the DD of children and their mental health. Steps have been taken to train facilitators from other cities in Serbia, making the program available to more families. With the support of partners from the respective governmental institutions, the program will become a regular service in institutions dealing with early intervention and developmental delay in children, and to will be cost free for the families.

Findings from the application of CST in Serbia indicate that the program could potentially aid a wider variety of parents and caregivers, and thus that the form of its application (thorugh group discussions, online, etc.) can encompass a greater number of parents, providing them adequate support for working with children and as well as for accepting their child's DD. Our indicative results provide the argument for its wider implementation, but it requires consistent advocacy efforts in order to secure the support of decision-makers. Only in that way the CST in Serbia can be widely implemented and sustained over time.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Ethics Committee of the Institute of Mental Health. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

NG contributed with technical support, data collection, literature research, writing and reviewing of the article, and statistical analysis. SP, RG, and SS contributed with data collection, literature research, result analysis, interpretation, writing, and reviewing process. MP-M contributed in the writing and reviewing process, the interpretation of the results, and expertise. All authors contributed to the article and approved the submitted version.

FUNDING

The implementation of the CST Program and this research was supported by UNICEF Serbia and HELP—Mission to the Republic of Serbia. All authors received funding from these two organizations.

ACKNOWLEDGMENTS

We thank the MTs: Nataša Srećković Milenković, Dr. Roberto Grujičić, Dr. Duško Stupar, Dr. Ana Kesić, Dr. Jasminka Marković, Jelena Maraš, Milena Petković Milenović, Jelena Petković and all facilitators who participated in the program for their valuable and constructive contribution during the planning and development of this research work. We thank our colleagues from Autism Speaks who provided supervision, insights and expertise to master trainers that greatly assisted the research. We would also like to show our gratitude to the caregivers, families of children with developmental disabilities for participating in the program and filling out questionnaires and feedback forms during the course of this research.

REFERENCES

1. Neuman MJ, Okeng'o L. Early childhood policies in low- and middle-income countries. *Early Years.* (2019) 39:223–8. doi: 10.1080/09575146.2019.1636571
2. Smythe T, Zuurmond M, Tann CJ, Gladstone M, Kuper H. Early intervention for children with developmental disabilities in low and middle-income countries – the case for action. *Int Health.* (2021) 13:222–31. doi: 10.1093/inthealth/ihaa044
3. Pejovic-Milovancevic M, Stankovic M, Mitkovic Voncina M, Grujicic R, Rudic N, Herrera A, et al. Perceptions on support, challenges and needs among parents of children with autism: the Serbian experience. *Psychiatr Danub.* (2018) 30:354–64.
4. Petrenko CLM. A review of intervention programs to prevent and treat behavioral problems in young children with developmental disabilities. *J Dev Phys Disabil.* (2013) 25. doi: 10.1007/s10882-013-9336-2 Avilable online at: https://www.ncbi.nlm.nih.gov/pmc/articles/PMC3821779/
5. Axford N, Lehtonen M, Kaoukji D, Tobin K, Berry V. Engaging parents in parenting programs: Lessons from research and practice. *Child Youth Serv Rev.* (2012) 34:2061–71. doi: 10.1016/j.childyouth.2012.06.011
6. Smith KA, Samuels AE. A scoping review of parental roles in rehabilitation interventions for children with developmental delay, disability, or long-term health condition. *Res Dev Disabil.* (2021) 111:103887. doi: 10.1016/j.ridd.2021.103887

7. Statistical Office of Republic of Serbia. *Vital Statistics.* (2019). Available online at: http://devinfo.stat.gov.rs (accessed March 8, 2022).
8. Pejovic-Milovancevic M, Grujicic R, Stupar S, Ninkovic M. Overcoming traps and challenges in child and adolescent psychiatry. *Srp Arh Celok Lek.* (2021) 149:236–41. doi: 10.2298/SARH191216009P
9. United Nations Children's Fund (UNICEF). *Situation Analyisis of Children and Adolescents in Serbia.* (2019). Available online at: https://www.unicef.org/serbia/media/13466/file/SitAn_publication_2019.pdf (accessed March 28, 2022).
10. Salomone E, Pacione L, Shire S, Brown FL, Reichow B, Servili C. Development of the WHO caregiver skills training program for developmental disorders or delays. *Front Psychiatry.* (2019) 10:769. doi: 10.3389/fpsyt.2019.00769
11. Salomone E, Ferrante C, Salandin A, Ferrara F, Torchio E, Foletti G, Ghersi S, Pacione L, Servili C. Acceptability and feasibility of the World Health Organization's Caregiver Skills Training implemented in the Italian National Health System. *Autism.* (2021) 39:223–8. doi: 10.1177/13623613211035228
12. Autism Speaks. *Autism Speaks Releases New CST Caregiver Quick Tips Videos to Support Parents and Caregivers of Children With Autism.* (2020). Available olnine at: https://www.autismspeaks.org/science-news/autism-speaks-releases-new-cst-caregiver-quick-tips-videos-support-parents-and (accessed March 29, 2022).
13. Tekola B, Girma F, Kinfe M, Abdurahman R, Tesfaye M, Yenus Z, et al. Adapting and pre-testing the World Health Organization's Caregiver Skills Training programme for autism and other developmental disorders in a very low-resource setting: Findings from Ethiopia. *Autism.* (2020) 24:51–63. doi: 10.1177/1362361319848532
14. Sengupta K, Shah H, Ghosh S, Sanghvi D, Mahadik S, Dani A, et al. World Health Organisation-Caregiver Skills Training (WHO-CST) Program: feasibility of delivery by non-specialist providers in real-world urban settings in India. *J Autism Dev Disord.* (2021). doi: 10.1007/s10803-021-05367-0. [Epub ahead of print].
15. Hamdani U, Akhtar P, Zill-e-Huma, Nazir H, Minhas F, Sikander S, et al. WHO Parents Skills Training (PST) programme for children with developmental disorders and delays delivered by Family Volunteers in rural Pakistan: study protocol for effectiveness implementation hybrid cluster randomized controlled trial. *Glob Ment Heal.* (2017) 4:e11. doi: 10.1017/gmh.2017.7
16. Rimland B, Edelson M. *Autism Treatment Evaluation Checklist*. San Diego, CA: Autism Research Institute (1999). Available online at: https://www.autismeval.com/ari-atec/report1.html
17. Rollè L, Gullotta G, Trombetta T, Curti L, Gerino E, Brustia P, et al. Father involvement and cognitive development in early and middle childhood: a systematic review. *Front Psychol.* (2019) 10:2405. doi: 10.3389/fpsyg.2019.02405
18. United Nations Children's Fund (UNICEF). *Designing Parenting Programmes for Violence Prevention: A Guidance Note.* (2020). 48 p. Available online at: https://www.unicef.org/media/77866/file/Parenting-Guidance-Note.pdf (accessed March 27, 2022).
19. UNICEF. *Regional Study on Parenting Adolescents and Parenting Support Programmes in Belarus, Bulgaria, Georgia, Moldova, Montenegro and Romania.* (2018). Available online at: https://www.unicef.org/eca/media/8556/file/ECARO-study-parenting-adolescents.pdf (accessed March 28, 2022).
20. O'Donovan KL, Armitage S, Featherstone J, McQuillin L, Longley S, Pollard N. Group-based parent training interventions for parents of children with autism spectrum disorders: a literature review. *Rev J Autism Dev Disord.* (2019) 6:85–95. doi: 10.1007/s40489-018-00155-6
21. Barlow J, Smailagic N, Ferriter M, Bennett C, Jones H. Group-based parent-training programmes for improving emotional and behavioural adjustment in children from birth to three years old. *Cochrane database Syst Rev.* (2010) CD003680. doi: 10.1002/14651858.CD003680.pub2
22. Reichow B, Servili C, Yasamy MT, Barbui C, Saxena S. Non-specialist psychosocial interventions for children and adolescents with intellectual disability or lower-functioning autism spectrum disorders: a systematic review. *PLoS Med.* (2013) 10:e1001572. doi: 10.1371/journal.pmed.1001572
23. Odom SL, Boyd BA, Hall LJ, Hume K. Evaluation of comprehensive treatment models for individuals with autism spectrum disorders. *J Autism Dev Disord.* (2010) 40:425–36. doi: 10.1007/s10803-009-0825-1
24. Oono IP, Honey EJ, McConachie H. Parent-mediated early intervention for young children with autism spectrum disorders (ASD). *Cochrane Database Syst Rev.* (2013) CD009774. doi: 10.1002/14651858.CD009774.pub2
25. Palmer ML, Keown LJ, Sanders MR, Henderson M. Enhancing outcomes of low-intensity parenting groups through sufficient exemplar training: a randomized control trial. *Child Psychiatry Hum Dev.* (2019) 50:384–99. doi: 10.1007/s10578-018-0847-z
26. Ingoldsby EM. Review of interventions to improve family engagement and retention in parent and child mental health programs. *J Child Fam Stud.* (2010) 19:629–45. doi: 10.1007/s10826-009-9350-2

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

ORIGINAL RESEARCH
published: 10 June 2022
doi: 10.3389/fpsyt.2022.919234

Autistic Adult Services Availability, Preferences, and User Experiences: Results From the Autism Spectrum Disorder in the European Union Survey

Martina Micai[1], *Francesca Fulceri*[1], *Tommaso Salvitti*[1], *Giovanna Romano*[2], *Luise Poustka*[3], *Robert Diehm*[4], *Georgi Iskrov*[5,6], *Rumen Stefanov*[5,6], *Quentin Guillon*[7], *Bernadette Rogé*[7], *Anthony Staines*[8], *Mary Rose Sweeney*[8], *Andrew Martin Boilson*[8], *Thora Leósdóttir*[9], *Evald Saemundsen*[9], *Irma Moilanen*[10,11], *Hanna Ebeling*[10,11], *Anneli Yliherva*[10,12], *Mika Gissler*[13,14,15], *Tarja Parviainen*[16], *Pekka Tani*[17], *Rafal Kawa*[18], *Eva Pisula*[18], *Astrid Vicente*[19], *Célia Rasga*[19], *Magdalena Budişteanu*[20], *Ian Dale*[21], *Carol Povey*[21], *Noelia Flores*[22], *Cristina Jenaro*[22], *Maria Luisa Monroy*[23], *Patricia García Primo*[24], *Tony Charman*[25], *Susanne Cramer*[26], *Christine Kloster Warberg*[26], *Ricardo Canal-Bedia*[22], *Manuel Posada*[24], *Diana Schendel*[26,27,28]† and *Maria Luisa Scattoni*[1]*†

[1] Istituto Superiore di Sanità, Research Coordination and Support Service, Rome, Italy, [2] Directorate General of Health Prevention, Ministry of Health, Rome, Italy, [3] University Medical Center Göttingen, Department of Child and Adolescent Psychiatry and Psychotherapy, Göttingen, Germany, [4] Department of Child and Adolescent Psychiatry and Psychotherapy, Medical University of Vienna, Vienna, Austria, [5] Institute for Rare Diseases, Plovdiv, Bulgaria, [6] Department of Social Medicine and Public Health, Faculty of Public Health, Medical University of Plovdiv, Plovdiv, Bulgaria, [7] CERPPS, Université Toulouse Jean Jaurès, Toulouse, France, [8] School of Nursing, Psychotherapy and Community Health, Dublin City University, Dublin, Ireland, [9] The State Diagnostic and Counselling Centre, Kópavogur, Iceland, [10] Clinic of Child Psychiatry, University Hospital of Oulu, Oulu, Finland, [11] Oulu University Hospital, Medical Faculty, Oulu, Finland, [12] Logopedic Child Language Research Center, University of Oulu, Oulu, Finland, [13] Department of Knowledge Brokers, Finnish Institute for Health and Welfare, Helsinki, Finland, [14] Research Centre for Child Psychiatry, University of Turku, Turku, Finland, [15] Department of Molecular Medicine and Surgery and Region Stockholm, Academic Primary Health Care Centre, Karolinska Institute, Stockholm, Sweden, [16] Finnish Association for Autism and Asperger's Syndrome, Helsinki, Finland, [17] Department of Psychiatry, University of Helsinki, Helsinki, Finland, [18] Faculty of Psychology, University of Warsaw, Warsaw, Poland, [19] Center for Biodiversity, Functional and Integrative Genomics, Instituto Nacional de Saúde Doutor Ricardo Jorge, Lisbon, Portugal, [20] "Victor Babeş" National Institute for Research and Development in Pathology and Biomedical Sciences, Bucharest, Romania, [21] National Autistic Society, The Centre for Autism, London, United Kingdom, [22] Departamento de Personalidad, Evaluación y Tratamiento Psicológicos Salamanca, Instituto Universitario de Integración en la Comunidad, University of Salamanca, Salamanca, Spain, [23] Departamento de Psicología Evolutiva y de la Educación, Instituto Universitario de Integración en la Comunidad, University of Salamanca, Salamanca, Spain, [24] Instituto de Salud Carlos III, Institute of Rare Diseases Research, Madrid, Spain, [25] King's College London, Institute of Psychiatry, London, United Kingdom, [26] Department of Public Health, Aarhus University, Aarhus, Denmark, [27] Lundbeck Foundation Initiative for Integrative Psychiatric Research, Aarhus, Denmark, [28] Department of Economics and Business, National Centre for Register-Based Research, Aarhus University, Aarhus, Denmark

Edited by:
Antonio Narzisi,
Stella Maris Foundation (IRCCS), Italy

Reviewed by:
Anna Ceraso,
University of Brescia, Italy
April Hargreaves,
National College of Ireland, Ireland

***Correspondence:**
Maria Luisa Scattoni
marialuisa.scattoni@iss.it

† These authors have contributed equally to this work

Specialty section:
This article was submitted to Autism, a section of the journal Frontiers in Psychiatry

Received: *13 April 2022*
Accepted: *23 May 2022*
Published: *10 June 2022*

Citation:
Micai M, Fulceri F, Salvitti T, Romano G, Poustka L, Diehm R, Iskrov G, Stefanov R, Guillon Q, Rogé B, Staines A, Sweeney MR, Boilson AM, Leósdóttir T, Saemundsen E, Moilanen I, Ebeling H, Yliherva A, Gissler M, Parviainen T, Tani P, Kawa R, Pisula E, Vicente A, Rasga C, Budişteanu M, Dale I, Povey C, Flores N, Jenaro C, Monroy ML, Primo PG, Charman T, Cramer S, Warberg CK, Canal-Bedia R, Posada M, Schendel D and Scattoni ML (2022) Autistic Adult Services Availability, Preferences, and User Experiences: Results From the Autism Spectrum Disorder in the European Union Survey. Front. Psychiatry 13:919234. doi: 10.3389/fpsyt.2022.919234

There is very little knowledge regarding autistic adult services, practices, and delivery. The study objective was to improve understanding of current services and practices for autistic adults and opportunities for improvement as part of the Autism Spectrum Disorder in the European Union (ASDEU) project. Separate survey versions were created for autistic adults, carers of autistic adults, and professionals in adult services. 2,009 persons responded to the survey and 1,085 (54%) of them completed at least one of the services sections: 469 autistic adults (65% female; 55% ≤35 years old), 441 carers of autistic adults (27% female; 6% ≤35 years old), 175 professionals in adult services (76% female; 67% in non-medical services). Top choices by autistic adults, carers or professionals for services best suiting their current needs were: residential services: "help in own home" (adults, carers of high independent adults, professionals), "fulltime residential facility" (carers of low independent adults); employment services: "job mentors" (adults, carers of high independent adults, professionals), "Sheltered employment" (carers of low independent adults); education services: "support in regular education setting" (all groups); financial services: financial support in lieu of employment ("Supplementary income for persons unable to have full employment" for adults, "full pension" for carers of low independent adults) or to supplement employment earnings for carers of high independent adults and professionals; social services: "behavior training" (adults) and "life skills training" (carers and professionals). Waiting times for specific services were generally $<$ 1 month or 1–3 months, except for residential services which could be up to 6 months; most professionals were uninformed of waiting times ($>$50% responded "don't know"). Five of seven residential services features recommended for autistic adults were experienced by $<$50% of adults. The knowledge of good local services models that work well for autistic adults was generally low across all services areas. The variation in services experiences and perceptions reported by autistic adults, carers, or professionals underscore the need to query all groups for a complete picture of community services availability and needs. The results showed areas for potential improvement in autistic adult services delivery in the EU to achieve recommended standards.

Keywords: autism spectrum disorder, adults, residential service, employment service, education service, financial service, social service

INTRODUCTION

Autism is characterized by deficits in social communication and interaction, and restricted/repetitive repertoires of behaviors, interests, and activities [Autism Spectrum Disorder, (1)] and usually is a lifelong condition (2–4). Despite the growing population of autistic youth aging into adulthood as well as newly diagnosed autistic adults, most studies on service use have been conducted investigating autistic children or young adults up until their late twenties (5, 6). In general, the research base in autistic adult services is underdeveloped which hampers efforts toward improvement of services provision and policymaking.

In adulthood, autistic persons often face challenges around services, such as lack of autism training of service providers and chaotic services management or, alternatively, having to pay for private services. Consequently, autistic adults or carers of autistic adults tend to express dissatisfaction with post-diagnostic support, interventions, and management of medical and psychiatric co-occurring conditions (7–15). A recent study explored autism service satisfaction and preferences of parents/guardians and autistic adults in Canada, France, Germany, Italy, and the United States (16) and the investigators found high rates of satisfaction for autism-specific early intervention and general day services but general dissatisfaction for job training and mixed-disability day services. Dissatisfaction could be one consequence of poor alignment between autistic services recommendations and actual experiences by users that has been reported in a few studies (8, 15, 17). Also, very few studies have explored the views of adult services by professionals versus autistic adults and carers [e.g., (15)].

The overall objective of the present study was to improve understanding of current services experienced by autistic adults and opportunities for improvement as part of the Autism Spectrum Disorder in the European Union (ASDEU) project. Specific study objectives were to examine perceptions and experiences of autistic adults, carers, and professionals on a variety of features of the overall services infrastructure (residential, employment, education, financial, social services) for autistic adults including: availability of public (versus private) services; whether the service they received was designed for autism specifically; what services were received versus what is perceived as most needed now; indicators of limited services availability (i.e., services waiting times); level of autism expertise in offices where services were applied for; alignment of user experiences of residential services with published guidelines for residential services; and users' awareness of good services models across all services types.

MATERIALS AND METHODS

Survey Development and Description

The ASDEU project conducted a survey on services based, in part, on a variety of published guidelines and recommendations regarding services for autistic adults [(18–21); Think Autism: Updating the 2010 Adult Autism Strategy]. The three versions of the survey targeted autistic adults; family/caregivers of autistic adults (NOT necessarily the carers of the adults who participated in this study themselves); and administrators/professionals/service providers for adults. Experts in all ASDEU sites reviewed the surveys and an autistic adult tested the adult version of the on-line survey. Written instructions were presented to the participants before they filled out the survey. Responders were asked to select answer choices that seemed to suit most closely with what they knew or had experienced and to answer to the best of their knowledge and experience. The survey questions were written using everyday language and avoided technical terms that might not be understood or not applicable across different countries. To ensure the reported information was recent, for each services section, only respondents who had applied for or had the service in the last 2 years were eligible to answer the section questions.

The present study used the following data: (1) demographic characteristics of responders, including 12 questions for the autistic adults, 9 for carers, and 7 for professionals; (2) residential services for autistic adults, including 11 questions for autistic adults, 11 for carers, and 6 for professionals; (3) employment services for autistic adults including 10 questions for autistic adults, 10 for carers, and 5 for professionals; (4) adult education services for autistic adults including 10 questions for autistic adults, 10 for carers, and 5 for professionals; (5) financial services for autistic adults including 10 questions for autistic adults, 10 for carers, and 5 for professionals; (6) social support services for autistic adults including 10 questions for autistic adults, 10 for carers, and 5 for professionals (**Supplementary Data 1**). For some of the questions regarding residential services provisions, the response options were designed to determine if the respondent's experiences with local services matched published recommendations (**Supplementary Data 10**). Supplementary Data 1 presents the survey questions and response options for the demographic characteristics and specific services questions examined for this study.

When asked to report availability and preference for services, the survey had two sets of questions for different services situations: whether a respondent had applied for a specific type of service in the last 2 years and (a) failed to get it or (b) received it. Respondents were then asked what type of service would best suit their current needs.

Recruitment and Survey Distribution

All ASDEU partners sent out survey invitations to participate to autism organizations (national, local, and voluntary) and service provider organizations (public and private, including residential facilities, job training, and education programs). Furthermore, these organizations were asked to share the survey links through their channels (e-newsletters, websites, or social media accounts). Also, the investigators at each site disseminated the surveys through their professional networks and on social media.

The survey was available online over 10.5 months in 2017. In mid-February, it was launched in three languages (English, Spanish, and Danish) and by mid-September 2017, in eight additional languages (French, Polish, Icelandic, German, Finnish, Italian, and Romanian, as well as Portuguese for the professional version); data for the analysis were collected until December 2017.

Each ASDEU site obtained local ethical approval before distributing the survey in their respective countries. All procedures were in accordance with the ethical standards of the institutional and/or national research committee. Responders read the information about the survey prior to start and gave their informed consent electronically. Personal identifying information was not collected. Data were analyzed in aggregated form.

Analysis Methods

The entire survey was completed or partially completed by 2,009 participants distributed as follows: 667 autistic adults, 591 carers of autistic adults, and 751 professionals. We excluded the 3.63% (*n* = 73, 21 autistic adults, 16 carers, and 9 professionals) of responders who partially completed the survey, resulting in a sample size of 1,963 responders (646 autistic adults, 575 carers, and 742 professionals). The response rate was 97.7%, minimizing the risk for non-response bias.

For the present study, only demographic characteristics, and responses specific to residential, educational, employment, financial or social services for autistic adults were analyzed. Other survey sections on autistic adults' diagnosis, health, and interventions are presented elsewhere (12, 13, 15).

For these analyses, distribution of responses (frequency, percent) from all three respondent groups regarding features of each type of service were analyzed separately; responses from the carer group for analysis were stratified on level of independence of the autistic adult under their care; for the questions on availability and preferences of services, responses from the autistic adult group were stratified on gender (**Supplementary Data 4B–8C**); we performed Chi-square tests (with Yates continuity corrections) on the affirmative answers to explore if autistic males vs. females vs. other gender/no gender answer differed in reporting the preference of services (**Supplementary Data 4C–8C**). We also repeated analyses of select questions – Have you tried (for adults)/Has the adult, or someone for the adult (for carers) tried to get a service at some time in the last 2 years? – to compare different countries of residence in terms of success of service availability/provision. For the questions on services, the number of respondents varied across the different service sections. A summary of the main findings is reported in **Table 1**.

RESULTS

Demographic Characteristics

For the demographic characteristics analysis, we considered responders to at least one of the services sections (*n* = 1,085). Responders were mostly women (autistic women: 308, 65%; carers: 361, 81%; professionals: 133, 76%), while the autistic adults cared for by carers were mainly men (319, 72%). Over half of the autistic adults (55%) were less than 35 years of age and 81%

TABLE 1 | Summary of the main findings.

Autistic adult	Carer	Professional
Demographic characteristics		
Mostly women, 65%	Autistic adults cared for by carers mainly men, 72%	Mostly women, 76%
Mostly less than 35 years of age, 55%	Mostly less than 35 years of age, 81%	NA
Primarily living in Denmark, 41%	Primarily living in Denmark, 27%	Primarily living in Denmark, 54%
	Autistic adults cared for by carers had hig or some level of independence, 50%	Most non-medical background, 69%
Organization of services		
Most applied for a residential service at a public office	Most applied for a residential service at a public office	NA
Availability and preferences of services		
Residential		
14% tried to get a residential service at some time in the last 2 years but failed	24% tried to get a residential service at some time in the last 2 years but failed	Most reported having knowledge of and work experience in residential services, 79%
29% were in a residential service now or had been at some time in the last 2 years	48% were in a residential service now or had been at some time in the last 2 years	"Help while living in a college or school dormitory" was reported less often as available, 42%
40% were satisfied with the residential service they currently had	31% carers of HI and 23% of LI were satisfied with the residential service they currently had	NA
"Help in own home" was the most frequent residential service got, failed, and needed	"Help in own home" was the most frequent residential service got, failed, and needed"; "full time residential facility" was the most frequent residential service got and failed for carers of LI	"Help in own home" and "full time residential facility with full apartment" were the most needed residential services
Employment		
12% tried to get an employment service at some time in the last 2 years but failed	13% tried to get an employment service at some time in the last 2 years but failed	About half of professionals reported having knowledge of and work experience in employment services
28% had an employment service now or had been at some time in the last 2 years	29% had an employment service now or had been at some time in the last 2 years	All services options except "employer programs to encourage employment of persons with autism" (19%), were reported to be available in the area by 44–77%
24% were satisfied with the employment service they currently had	20% carers of HI and 20% of LI were satisfied with the employment service they currently had	NA
"Specific counseling" was the most frequent employment service failed and "Internships or work placement" the most frequent got	"Internship or work placement" was the most frequent employment service failed and got for carers of HI; "Job mentors" was the most frequent employment service failed and "Sheltered employment and job mentors" was the most frequent employment service got for carers of LI	NA
"Job mentors" and "job placement specifically for autistic adults" were the service that would suit them best now (i.e., not satisfied with current service)	"Job mentors" and "job placement specifically for autistic adults" were the service that would suit them best now (i.e., not satisfied with current service) for carers of HI; "sheltered employment" and "job mentors" for LI	"Job mentors" and "job placement specific for autistic persons/employer programs to encourage employment of persons with autism" were the most needed employment services
Education		
6% tried to get an education service at some time in the last 2 years but failed	12% tried to get an education service at some time in the last 2 years but failed	37% reported having knowledge of and work experience in education services
14% had an education service now or had been at some time in the last 2 years	29% had an education service now or had been at some time in the last 2 years	All services options were claimed to be currently available by >65% of respondents except for "boarding school or college for adults with autism spectrum" (35%)
35% were satisfied with the education service they currently had	29% carers of HI and 34% of LI were satisfied with the education service they currently had	NA
"Mentorship or specialist support in a regular education setting" and "day school or college for adults with autism" were the most frequent education service got, failed, and needed	"Mentorship or specialist support in a regular education setting" and "day school or college for adults with autism" were the most frequent residential service got, failed, and needed	"Mentorship or specialist support in regular education settings" and "day school or college for adults with autism spectrum" were the most needed education services
Financial		
18% tried to get a financial service at some time in the last 2 years but failed	22% tried to get a financial service at some time in the last 2 years but failed	25% reported having knowledge of and work experience in financial services

(Continued)

TABLE 1 | (Continued)

Autistic adult	Carer	Professional
43% had a financial service now or had been at some time in the last 2 years	64% had a financial service now or had been at some time in the last 2 years	All services options were claimed to be currently available by >51% of respondents except for "special insurance to help pay for health care" (28%)
28% were satisfied with the financial service they currently had	21% carers of HI and 23% of LI were satisfied with the financial service they currently had	NA
"Supplementary income" was the most frequent financial service failed and "unemployment benefits" the most frequent got	"Support during school or job training" was the most frequent financial service failed for carers of HI; "caregiver supplementary income" for carers of LI; "full pension" was the most frequent financial service failed and got for carers of HI and LI	NA
"Supplementary income for persons unable to have full employment" and "full pension" were the most needed financial services	"Supported employment" was the most frequent financial service needed for HI; "full pension" for LI	"Supported employment" and "supplementary income for persons unable to have full employment" were the most needed financial services
Social support		
12% tried to get a social support service at some time in the last 2 years but failed	25% tried to get a social support service at some time in the last 2 years but failed	57% reported having knowledge of and work experience in social support services
31% had a social support service now or had been at some time in the last 2 years	36% had a social support service now or had been at some time in the last 2 years	"Life skills training" and "free time activities" were the most frequently selected options to be currently available
25% were satisfied with the social support service they currently had	12% carers of HI and 18% of LI were satisfied with the social support service they currently had	NA
"Free time activities" was the most frequent social support service failed and "life skill training" the most frequent got	"Free time activities" was the most frequent social support service failed for carers of HI; "life skill training" for carers of LI; "life skill training" was the most frequent social support service failed and got for carers of HI and LI, together with "free time activities" only for LI carers	NA
"Behavior training, for an individual," "peer to peer matching," "peer-to non-peer matching," and "life skills training" were the most needed social support services	"Peer-to non-peer matching" and "life skills training" were the most frequent social support service needed for HI; "life skills training," "behavior training for an individual," and "peer-to non-peer matching" for LI	"Life skills training," "free time activities," and "behavior training for individuals" were the most needed social support services
Alignment of user's experience with residential services guidelines		
<50% of autistic adults experienced: ways to get specialist care when needed, services coordination, activities to feel part of the community, the physical environment was adapted to their needs, and staff had specialist autism training	<50% of carers experienced: ways to get specialist care when needed, services coordination, support for employment, and support for independent living	Foreach recommended provision of residential services 63% of professionals said that it was in place or coming
Waiting times		
For the most services < 1 month or 1–3 months. Waiting times for residential services were more disparate and the time choices were more variable across the different types of residential services	For the most services < 1 month or 1–3 months. Waiting times for residential services were more disparate and the time choices were more variable across the different types of residential services	For the most part, did not know the waiting times for services and when they did report a waiting time it generally was not concordant with the adults' and carers' reports
Straff training		
<50% of autistic adults said that the staff of the office where to apply for services seemed knowledgeable about autism or autism services.	<50% of carers said that the staff of the office where to apply for services seemed knowledgeable about autism or autism services.	NA
50% or more said that only some or none of the staff seemed knowledgeable about autism spectrum or autism spectrum services	50% or more said that only some or none of the staff seemed knowledgeable about autism spectrum or autism spectrum services	NA
At the residential facility, 47% said that staff had special training in autism	At the residential facility, 59% said that staff had special training in autism	At the residential facility, 79% said that staff had special training in autism
Good local models		
<34% of adults knew of a good local service model. The domains with the lowest proportions of "yes" among adults were financial service, employment service, and adult education service	<34% of carers knew of a good local service model. The domain with the lowest proportions of "yes" among carers was financial service	Higher proportions of professionals knew of good local models, although for financial and social support service < 50% of professionals knew of a good local model
The domain with the highest proportions of "yes" for a good local model was the residential service	The domain with the highest proportions of "yes" for a good local model was the residential service	The domain with the highest proportions of "yes" for a good local model was the residential service

NA = Question not available for the correspondent group. HI, high independent adults; LI, low independent adults.

of the carers' adults were less than 35. Responders were primarily living in Denmark (410, 37%), France (147, 13%), Spain (125, 11%), Finland (109, 10%), Poland (88, 8%), Italy (79, 7%), and Iceland (71, 6%) and lived in cities that are not capital cities (758, 69%) (**Supplementary Data 2**).

Most of the autistic adult responders reported to be currently in a college/university education program (65, 61%) or had completed study at a college/university level (149, 40%). Over half (247, 52%) of the autistic adult responders were unemployed, and the most common reason for unemployment was having a disability that prevents them from having a job (80, 32%). Only 20% of the autistic responders were diagnosed between 16 and 25 years old, while the rest were 26 years of age or older when diagnosed (**Supplementary Data 2**).

About half of autistic adults cared for by carers had some level of independence (high level of independence, 42, 9%; some independence but needs support, 180, 40%), whereas the other half required a high level of support (needs a high level of support in daily living, 149, 33%; needs high level institution-like care, 70, 15%). Fifty-eight percent (n = 42) of the autistic adults cared for by carers were diagnosed with autism between 16 and 25 years old, while the rest were 26 years of age or older when diagnosed (**Supplementary Data 2**).

The most represented backgrounds of the professionals were teachers/pedagogues (45, 25%), social workers (44, 25%), and psychologists (20, 11%); 31% (n = 55) of professionals selected the option "Other" when asked to select their professional background. Most (69%) of the professionals had a non-medical background (**Supplementary Data 2**).

Organization of Services

Most adults and carers (60%) applied for service at a public office; almost all financial services applications (90%) were made at a public office. A large minority of social services applications (19.7% by autistic adults and 14.5% by carers) were made at charitable organizations (**Supplementary Data 3A**).

For employment, education or financial services, most adults and carers received the service from organizations NOT set up for autism specifically (largest percentages of respondents received the service at organizations for either all persons or persons with disabilities; autistic adults: 28.9% to 45.2%; carers: 18.5% to 71.9%) (**Supplementary Data 3B**). For residential and social services, however, 40% of adults and 50% of carers received the service from organizations set up for autism specifically (**Supplementary Data 3B**).

Availability of and Preferences for Services

Residential

Among respondents, 14.5% of adults (48 of 331; 12.9% females; 18% males; 13.3% other gender/gender no answer), or someone for the adult, and 24.2% (55 of 227) of carers tried to get a residential service at some time in the last 2 years but failed while 29.1% (138 of 474) of adults and 48.8% (217 on 444) of carers reported that they were in a residential service now or had been at some time in the last 2 years (**Figure 1** and **Supplementary Data 4B**). Autistic adults living in Italy (6, 16.6%) and Germany (1, 16.6%) and carers living in Finland (8, 22.2%) and Iceland (9, 22.5%) were those who have tried more often to get residential service and failed (**Supplementary Data 9**).

About 40.5% of adults but a lower percentage of carers (of high independence adults: 31.5%; of low independence adults: 23.9%) were satisfied with the residential service they currently had (**Figure 2** and **Supplementary Data 4A**).

For autistic adult respondents, "help in own home" was by far the most frequent residential service: over 41.1% of adults who tried but failed to get a residential service in the last 2 years, were trying to get "help in own home"; 53.1% of adults who had a residential service in the last 2 years had "help in own home" service (**Supplementary Data 4B**). The most frequent choice by autistic adults for a residential service best suiting their needs now (i.e., the adults who were not satisfied with what they had already) was "help in own home" (26.6%) (**Figure 2** and **Supplementary Data 4A**).

For the same three questions, the residential service profile of the adults of carers differed between adults of low and high independence. For carers of high independence adults, "help in own home" was the most common answer choice (**Supplementary Data 4B** – kind of residential service that was applied for, but failed: 34.2%; kind of residential service the adult had: 38.8%; residential service best suiting there needs now 32.8%), while "full time residential facility" was the most common answer choice when the adult was of low independence (**Supplementary Data 4B** – kind of residential service that was applied for, but failed: 26.0%; kind of residential service the adult got: 43.7%; **Supplementary Data 4A** – kind of service best suiting there needs now: 32.4%).

Similar distributions of responses on residential services availability and preferences were observed for autistic males and females (**Supplementary Data 4C**; X^2 = 10.1, p = 0.60, **Supplementary Data 4D**).

Most professionals (177 of 224; 79%) reported having knowledge of and work experience in residential services that are currently available for adults, including autistic adults. Each residential service option was said to be available in the area by about 56.2–78.8% of professionals, except for "help while living in a college or school dormitory" which was said to be available by only 42.5% of professionals (**Supplementary Data 4B**). Consistent with the autistic adults and carers, the top two choices by professionals for residential services most needed in their area were "help in own home" as well as "full time residential facility with full apartment" (respectively selected by 27.3 and 25.4% of professionals) (**Figure 2** and **Supplementary Data 4A**).

Employment

Among responders, 12.7% of the adults, or someone for the adult, (43 on 336; 12.5% females; 14.2% males; 7.1% other gender/no gender answer) and 13.3% (41 on 307) of carers tried to get an employment service at some time in the last 2 years but failed, while 28% (131 on 467) of the adults and the 29.3% (128 on 436) of the carers reported that had an employment service now or had been at some time in the last 2 years (**Figure 1** and **Supplementary Data 5B**). Autistic adults living in Italy (8, 22.2%)

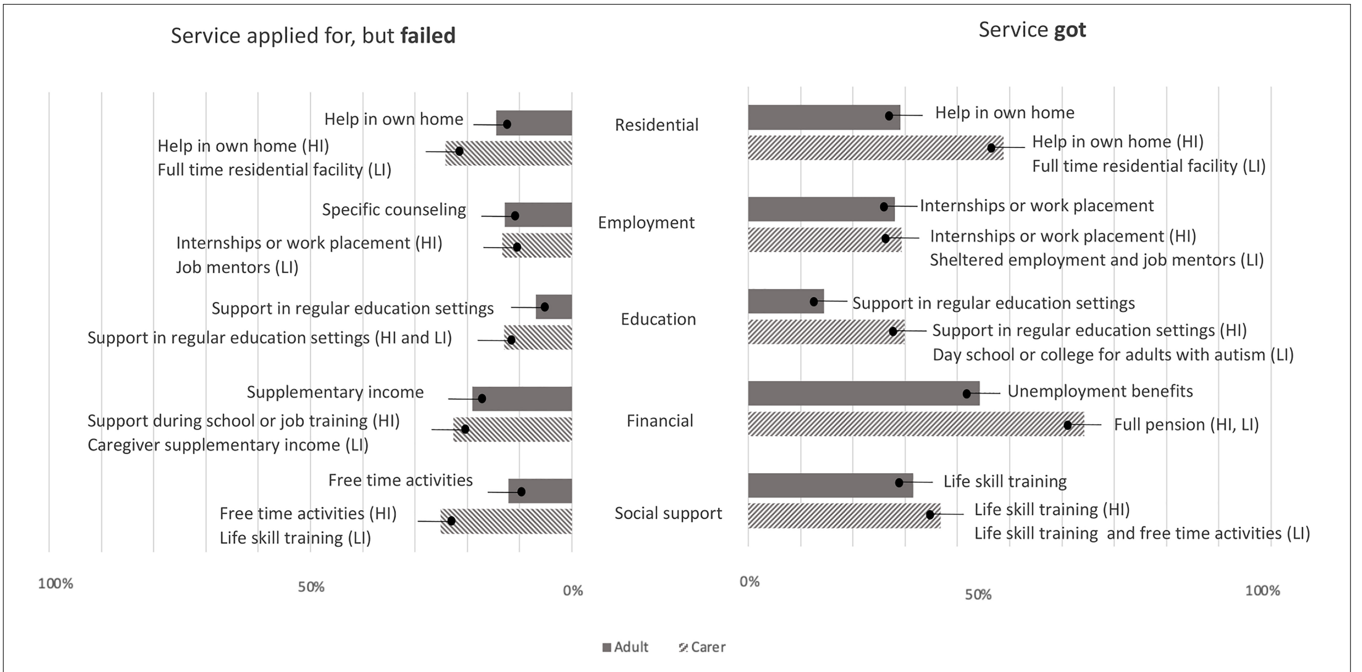


FIGURE 1 | Services that had been applied for but failed to get or Services got reported by autistic adults and carers. Values expressed as percentages. Shown in the text boxes are the most frequently selected specific service option within each service area reported by autistic adults and carers of high independent adults (HI) and low independence adults (LI).

and Germany (1, 16.6%) and carers living in Finland (5, 13.8%) were those who have tried more often to get employment service and failed (**Supplementary Data 9**).

A quarter or less of adults (24.1%) and carers (of high independence adults: 20.7%; of low independence adults: 20.4%) were satisfied with the employment service they currently had (**Figure 3** and **Supplementary Data 5A**).

For adults who tried to get a service and failed, the top two services they sought were "employment counseling specific for persons with autism spectrum" (17.1%) and "job mentors" (16.1%). For adults who had an employment service, the most common services they got were "internships or work placement" (21.6%) and "job mentors" (18.7%) (**Supplementary Data 5B**). When asked what service that would suit them best now (i.e., not satisfied with current service), the top choices by the adults were: "job mentors" (18.1%) and "job placement specifically for autistic adults" (13.5%) (**Figure 3** and **Supplementary Data 5A**).

For carers, the top two choices differed by the adult's level of independence. For a service that the carers tried to get and failed, carers of high independence adults were more often trying to get "internships or work placement" (16.6%) and "community job center" (14.2%) while carers of low independence adults were trying to get "job mentors" (26.8%) or "employment counseling specifically for autistic persons" (17%). For carers who had a service, the most frequent services the adult got among carers of high independence adults was "internships or work placement" (21.5%) and "job mentors" (19.6%) while carers of low independence adults got "sheltered employment" (18.1%) or "job mentors" (18.1%) (**Supplementary Data 5B**). For services that would suit best the adult now (i.e., not satisfied with current service), carers of high independence adults selected more often "job mentors" (24.8%) and "job placement specifically for autistic adults" (12.9%) while carers of low independence adults selected "sheltered employment" (19.5%) and "job mentors" (18.6%) **Supplementary Data 5A**).

Similar distributions of responses on employment services availability and preferences were observed for autistic males and females (**Supplementary Data 5C**; $X^2 = 14.7$, $p = 0.68$, **Supplementary Data 5D**).

About half of the professionals reported having knowledge of and work experience in employment services that are currently available for adults, including autistic adults. For professionals, all of the 11 services options except "employer programs to encourage employment of persons with autism" (19.1%), were reported to be available in the area by 44–77% of professionals (**Supplementary Data 5B**). For professionals, the top two choices of services most needed now were: "job mentors" (18.1%) and "job placement specific for autistic persons/employer programs to encourage employment of persons with autism" (15.3%) (**Figure 3** and **Supplementary Data 5A**).

Education

Among respondents, 6.8% of adults (27 of 393; 5.7% females; 10.4% males; 7.1% other gender/no gender answer), or someone for the adult, and 12.9% (39 of 302) of the carers tried but failed to get an adult education service at some time in the last 2 years; 14.5% (67 of 462) adults and 29.9% (129 of 431) of the carers reported that they had an adult education service now or at some time in the last 2 years (**Figure 1** and **Supplementary Data 6B**). Autistic adults living in Italy (7, 19.4%) and Republic of

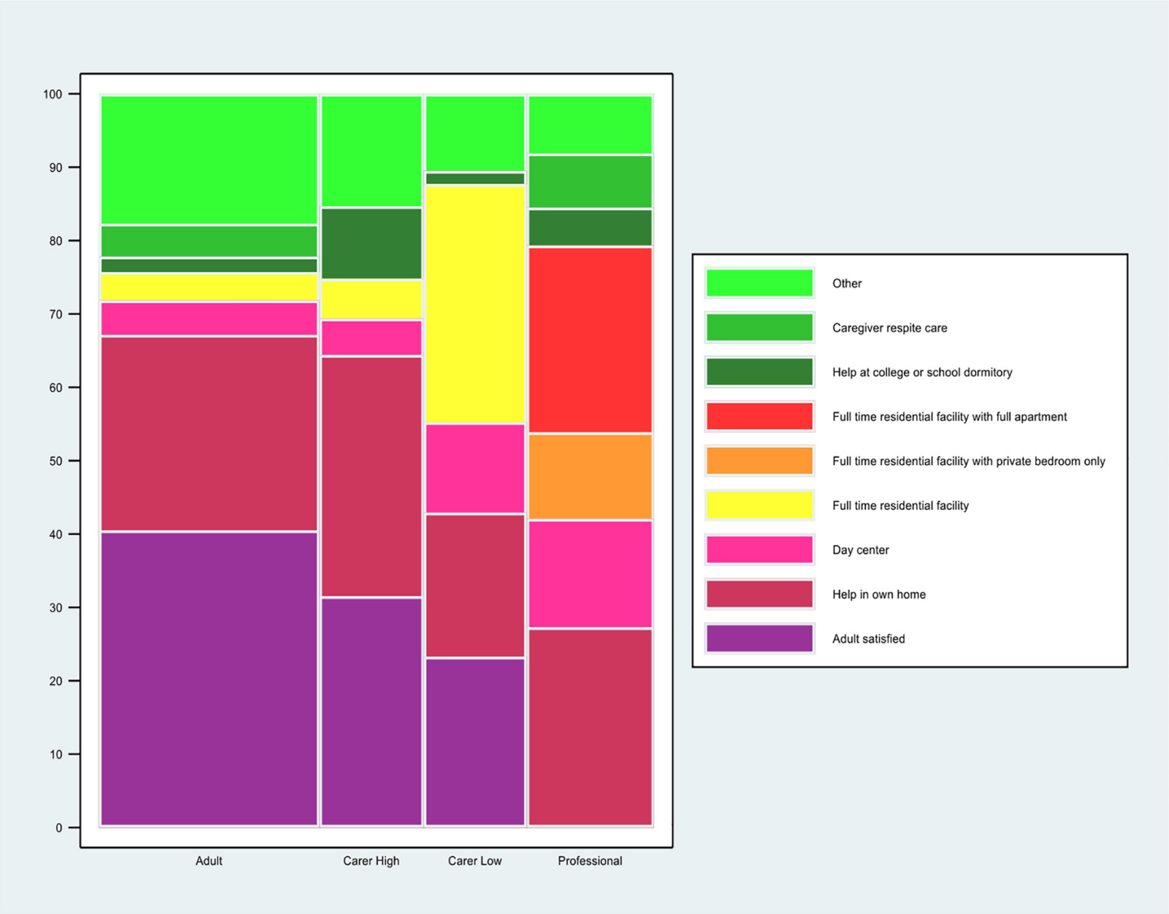


FIGURE 2 | Preferred residential service. The question for the autistic adults was: *"If you could choose a residential service that fits your needs best now, what would you choose? (Please, tick 1 box)."* The question for the carers of autistic adults was: *"If you could choose a residential service that fits the adult's needs best now, what would you choose? (Please, tick 1 box)."* The carers' data are stratified by high and low level of independence of the autistic adult. The question for the professional was: *"Which 2 types of residential services do you think are most needed for autistic adults in the (geographical) area where you work now? (Please, tick 2 boxes)."* Professionals who selected "*I prefer not to make a choice*" are not included in the calculation of % for each of the other answer choices. The %s for the other answer choices are based on the professionals who made a choice, *n* = 271 (301–30 = 271). *This% is calculated on the total sample of the professionals' responders (*n* = 301). Data are for all adults and carers of adults.

Ireland (2, 15.3%) and carers living in Republic of Ireland (2, 20.0%), Finland (5, 13.8%), and France (9, 13.4%) were those who have tried more often to get education service and failed (**Supplementary Data 9**).

About a third of adults (35.8%) and carers (of high independence adults: 29.1%; of low independence adults: 34.1%) were satisfied with the education service they currently had (**Figure 4** and **Supplementary Data 6A**).

For adults and carers, the most frequently selected education service option across all three questions (tried and failed; had a service; what service would best suit now) was "mentorship or specialist support in a regular education setting" while "day school or college for adults with autism" was consistently a close second choice. The option that was consistently least frequently selected across all 3 questions and all respondent groups was "boarding school or college for autistic adults" (**Figure 4** and **Supplementary Data 6A,B**).

Similar distributions of responses on education services availability and preferences were observed for autistic male and female (**Supplementary Data 6C**; $X^2 = 9.2$, $p = 0.32$, **Supplementary Data 6D**).

About a third (37%) of the professionals reported having knowledge of and work experience in adult education services that are currently available for adults, including autistic adults. For professionals, all services options were claimed to be currently available by >65% of respondents except for "boarding school or college for adults with autism spectrum" (only 35.5% of professionals said it was currently available) (**Supplementary Data 6B**). For professionals, the 2 options most frequently selected as most needed now were "mentorship or specialist support in regular education settings" (42.3%) and "day school or college for adults with autism spectrum" (36.9%) (**Figure 4** and **Supplementary Data 6A**).

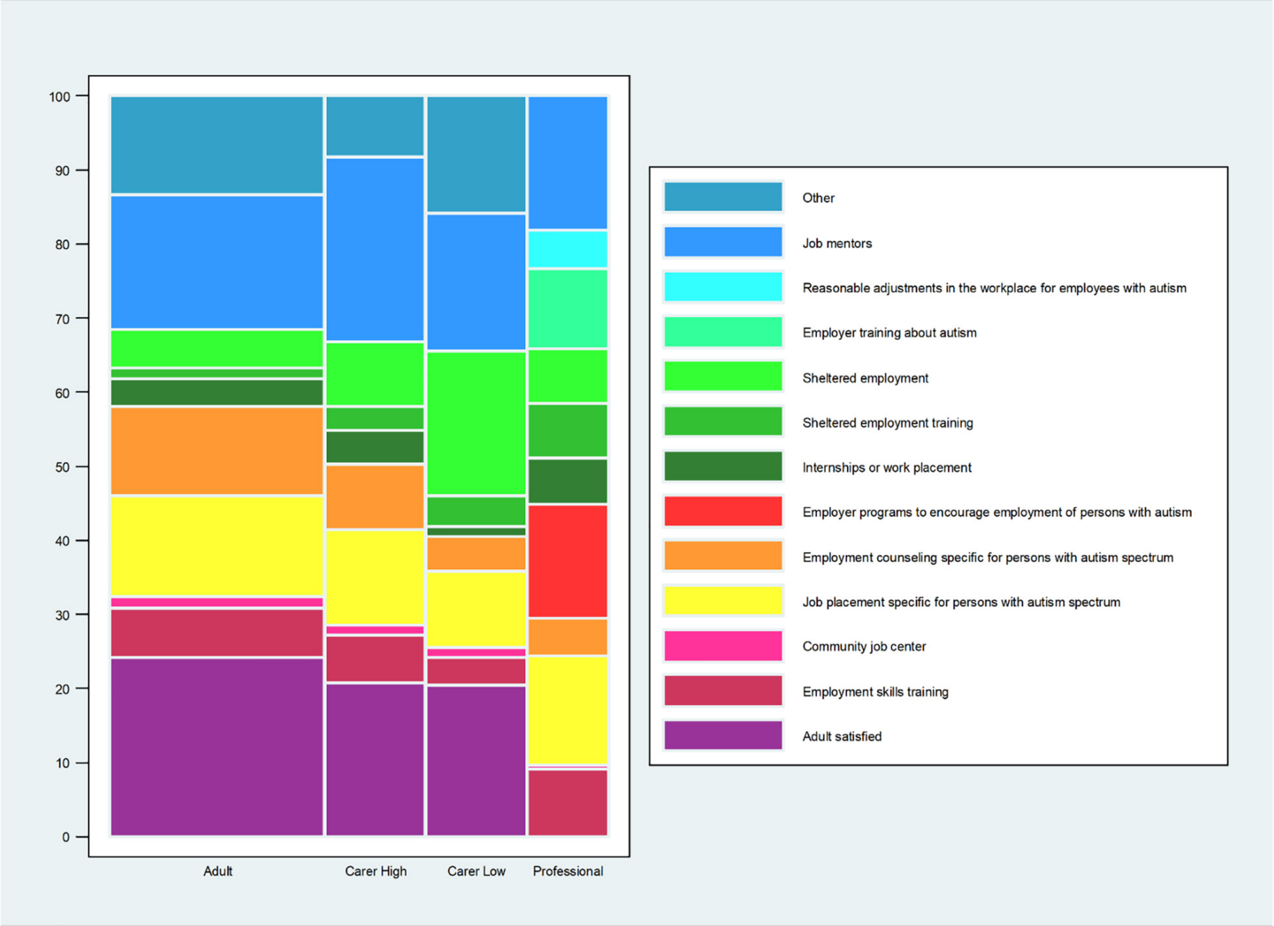


FIGURE 3 | Preferred employment service. The question for the autistic adults was: *"If you could choose an employment service that fits your needs best now, what would you choose? (Please, tick 1 box)."* The question for the carers of autistic adults was: *"If you could choose an employment service that fits the adult's needs best now, what would you choose? (Please, tick 1 box)."* The carers' data are stratified by high and low level of independence of the autistic adult. The question for the professional was: *"Which 2 types of employment services do you think are most needed for autistic adults in the (geographical) area where you work now? (Please, tick 2 boxes)."* Professionals who selected "*I prefer not to make a choice*" are not included in the calculation of% for each of the other answer choices. The%s for the other answer choices are based on the professionals who made a choice, *n* = 176 (198–22 = 176). *This% is calculated on the total sample of the professionals' responders (*n* = 198). Data are for all adults and carers of adults.

Financial

Among respondents, 18.9% of adults (49 of 258; 18.5% females; 22% males), or someone for the adult, and 22.6% (34 of 150) of the carers tried and failed to get a financial service at some time in the last 2 years while 43.2% (197 of 455) of the adults and 64.2% (270 of 420) of the carers reported that they had financial service now or had one at some time in the last 2 years (**Figure 1**). Autistic adults living in Spain (5, 22.7%) and France (13, 21.3%) and carers living in Republic of Ireland (1, 10.0%), Denmark (12, 9.8%) and Italy (3, 9.3%) were those who have tried more often to get financial service and failed (**Supplementary Data 9**).

About a third of adults (28.7%) but fewer carers (of high independence adults: 21.8%; of low independence adults: 23.1%) were satisfied with the financial service they currently had (**Figure 5** and **Supplementary Data 7A**).

For adults, the most frequently selected financial service options that they tried to get and failed were "supplementary income for persons unable to have full employment" (18.4%) or "full pension" (13.1%) (**Supplementary Data 7B**). For the service the adult got the most frequent options were "unemployment benefits" (22.3%) and "full pension" (16.2%) (**Supplementary Data 7B**). The most frequent options that would best suit their needs now were "supplementary income for persons unable to have full employment" (17.4%) or "full pension" (12.3%) (**Figure 5** and **Supplementary Data 7A**).

For carers the selected options differed depending on the level of their adult's independence. For carers of high independence adults, the top selected option for a financial service they tried to get and failed was "stipend/support during school or job training" (26%) while for carers of low independence adults "caregiver supplementary income" (42.8%) was the top choice. The most frequently selected financial service option across questions for a service that the adult got was "full pension" for both carers of high independence adults (19.5%) and low independence

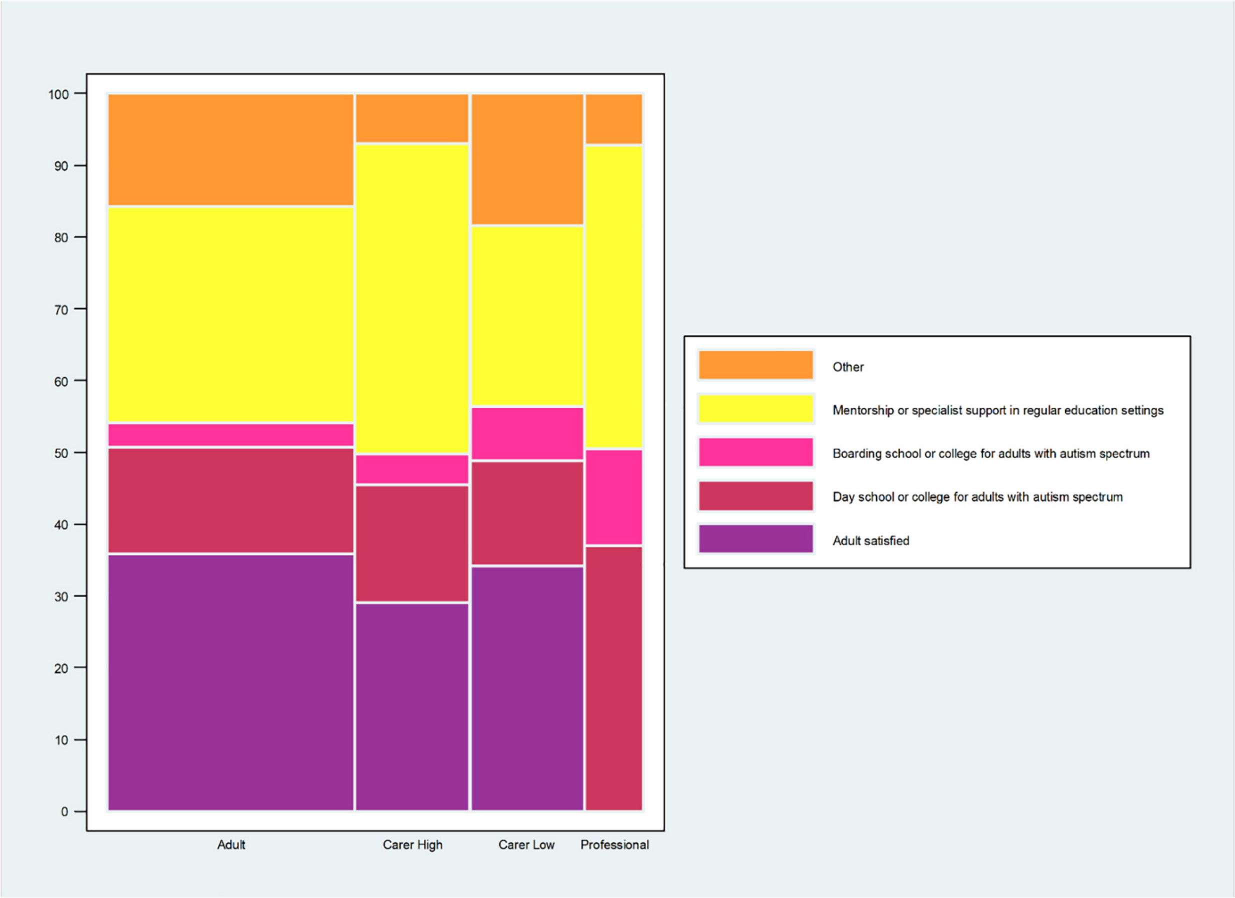


FIGURE 4 | Preferred adult education service. The question for the autistic adults was: *"If you could choose an education service that fits your needs best now, what would you choose? (Please, tick 1 box)."* The question for the carers of autistic adults was: *"If you could choose an education service that fits the adult's needs best now, what would you choose? (Please, tick 1 box)."* The carers' data are stratified by high and low level of independence of the autistic adult. The question for the professional was: *"Which 2 types of education services do you think are most needed for autistic adults in the (geographical) area where you work now? (Please, tick 2 boxes)."* Professionals who selected "*I prefer not to make a choice*" are not included in the calculation of% for each of the other answer choices. The %s for the other answer choices are based on the professionals who made a choice, *n* = 111 (124–13 = 111). *This% is calculated on the total sample of the professionals' responders (*n* = 124). Data are for all adults and carers of adults.

adults (39.2%) (**Supplementary Data 7B**). For carers of high independence adults, the most frequent option that would best suit their needs now was "supported employment" (21.3%) and for carers of low independence adults "full pension" (25.6%) (**Supplementary Data 7A**).

Gender analysis showed a significant difference in preference responses for financial services ($X^2 = 7.4$, $p = 0.005$, **Supplementary Data 7D**). About 20% of females preferred "Supplementary income for persons unable to have full employment," while only 12.1% of males selected this option. Whereas "Supported employment" was more often selected by males (14.9%) than females (7.1%; **Supplementary Data 7D**).

About a quarter of the (25.6%) professionals reported having knowledge of and work experience in financial services that are currently available for adults, including autistic adults. For these professionals, all services options were claimed to be currently available by >51% of respondents except for "special insurance to help pay for health care" (only 28.8% of professionals said it was currently available; **Supplementary Data 7B**). For professionals, the 2 options most frequently selected as most needed were "supported employment" (26.9%) and "supplementary income for persons unable to have full employment" (19.2%) (**Figure 5** and **Supplementary Data 7A**).

Social Support

Among responders, 12% of adults (37 of 306; 12.3% females; 11.0% males; 16.6% other gender/no gender answer), or someone for the adult, and 25% (66 of 263) of the carers tried and failed to get a social support service at some time in the last 2 years; 31.5% (142 of 450) of the adults and 36.7% (153 of 416) of the carers reported that had a social support service now or had one at some time in the last 2 years (**Figure 1**). Autistic adults living in France (10, 16.3%) and Spain (3, 13.6%) and carers living in Iceland (10, 25.0%), Italy (7, 21.8%), Republic of Ireland (2, 20.0%), and France (13, 19.4%) were those that have tried more often to get social support service and failed (**Supplementary Data 9**).

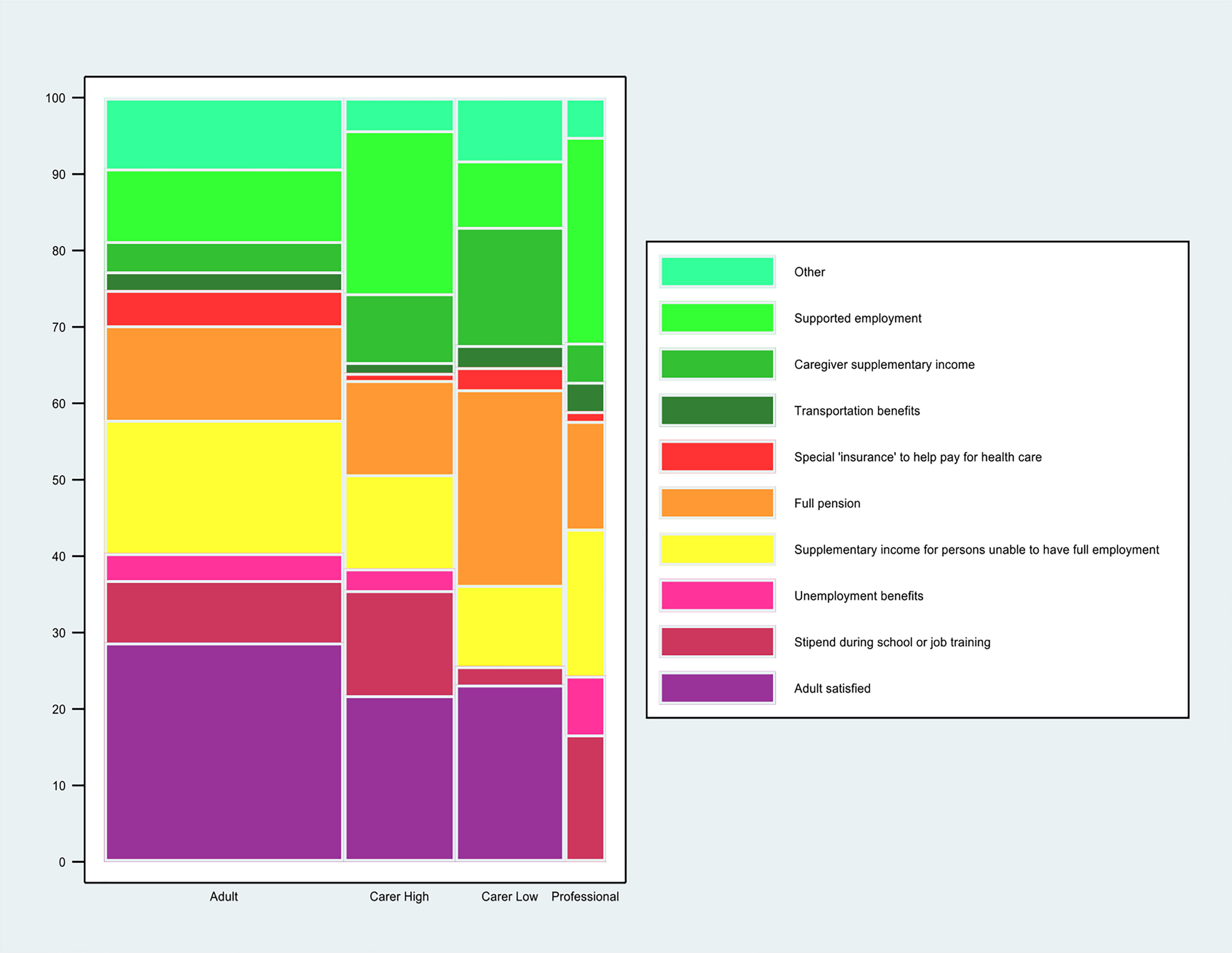


FIGURE 5 | Preferred financial service. The question for the autistic adults was: *"If you could choose a financial service that fits your needs best now, what would you choose? (Please, tick 1 box)."* The question for the carers of autistic adults was: *"If you could choose a financial service that fits the adult's needs best now, what would you choose? (Please, tick 1 box)."* The carers' data are stratified by high and low level of independence of the autistic adult. The question for the professional was: *"Which 2 types of financial services do you think are most needed for autistic adults in the (geographical) area where you work now? (Please, tick 2 boxes)."* Professionals who selected "*I prefer not to make a choice*" are not included in the calculation of% for each of the other answer choices. The %s for the other answer choices are based on the professionals who made a choice, *n* = 78 (88–10 = 78). *This% is calculated on the total sample of the professionals' responders (*n* = 88). Data are for all adults and carers of adults.

About a third of adults (25.9%) but fewer carers (of high independence adults: 12%; of low independence adults: 18.4%) were satisfied with the social support service they currently had (**Figure 6** and **Supplementary Data 8A**).

For both adults and carers - of both high and low independence adults - the two most frequently selected choices for social services they tried to get and failed, or currently have, were: "life skills training" and "free time activities" (**Supplementary Data 8B**). In contrast, when asked what services options would suit them best now, adults selected "behavior training, for an individual" and "peer to peer matching" (11.4%), as well as "peer-to non-peer matching" (11.0%) and "life skills training" (10.1%) (**Supplementary Data 8A**). For carers of high independence adults, their top choices for this question were "peer-to non-peer matching" as well as "life skills training" (17.8%). For carers of low independence adults, their top two choices were "life skills training" (22.8%), "free time activities" (14.5%) in addition to "behavior training for an individual" (14.0%) and "peer-to non-peer matching" (12.6%) (**Supplementary Data 8A**).

Similar distribution of responses on social support services availability and preferences were observed for autistic males and females (**Supplementary Data 8C**; X^2 = 28.2, *p* = 0.10, **Supplementary Data 8D**).

Over half (57.2%) of the professionals reported having knowledge of and work experience in social support services that are currently available for adults, including autistic adults. The same two service types selected by autistic adults (tried to get and failed, or currently have: "life skills training" and "free time activities") were by far the most frequently selected options to be currently available by the professionals (both said to be currently available by 80% of professionals) (**Supplementary Data 8B**). The

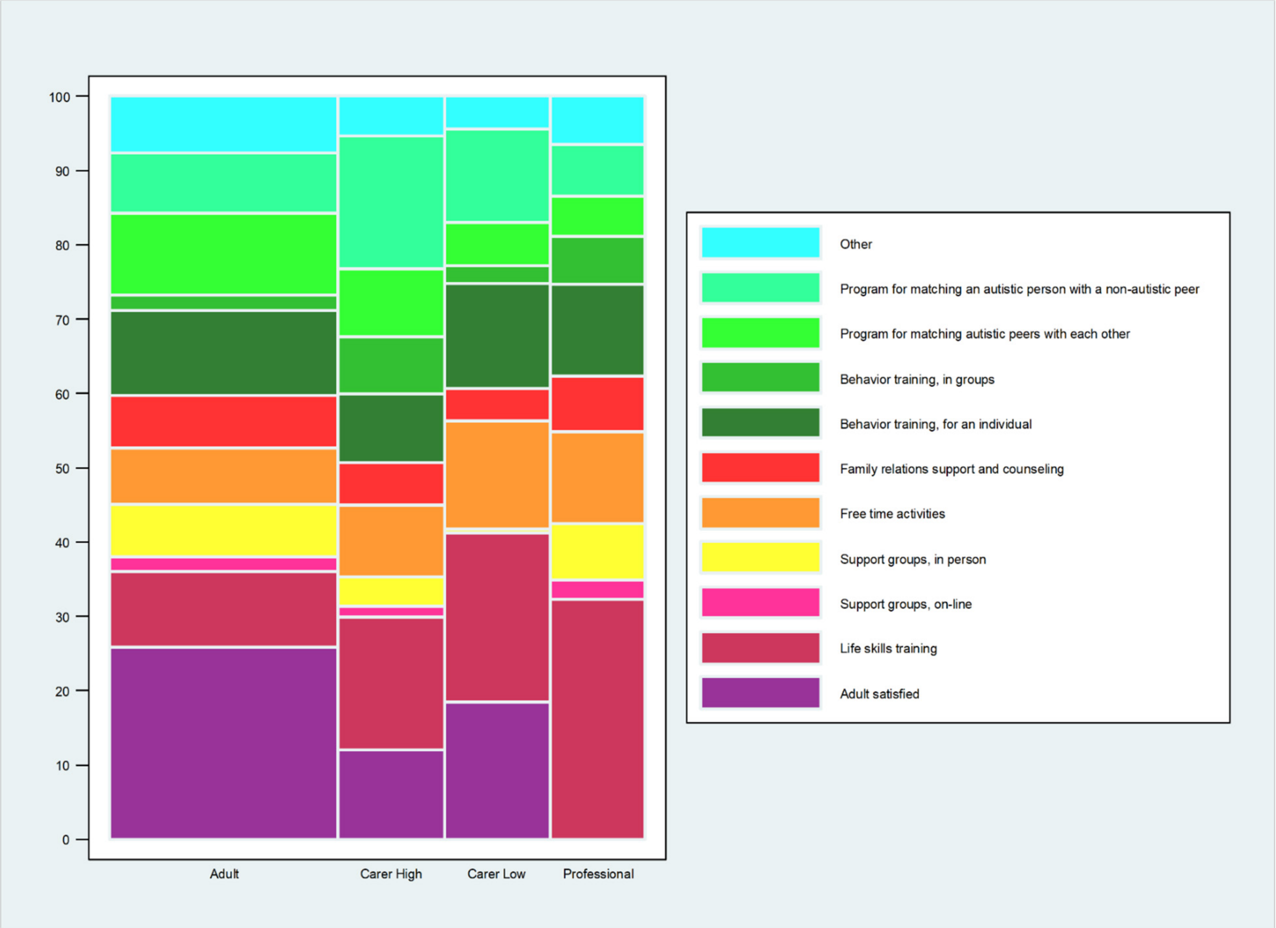


FIGURE 6 | Preferred social support service. Data are for all adults and carers of adults. The question for the autistic adults was: "*If you could choose a social support service that fits your needs best now, what would you choose? (Please, tick 1 box).*" The question for the carers of autistic adults was: *"If you could choose a social support service that fits the adult's needs best now, what would you choose? (Please, tick 1 box)."* The carers' data are stratified by high and low level of independence of the autistic adult. The question for the professional was: *"Which 2 types of social support services do you think are most needed for autistic adults in the (geographical) area where you work now? (Please, tick 2 boxes)."* Professionals who selected "*I prefer not to make a choice*" are not included in the calculation of% for each of the other answer choices. The %s for the other answer choices are based on the professionals who made a choice, *n* = 186 (207–21 = 186). *This% is calculated on the total sample of the professionals' responders (*n* = 207).

professionals' choices for what was most needed was consistent with that of the adults and carers: "life skills training" (by far the most frequently selected option at 32.2% of professionals), "free time activities" and "behavior training for individuals" (12.3%; **Figure 6** and **Supplementary Data 8A**).

Alignment of User's Experience With Residential Services Guidelines

For each recommended provision of residential services (i.e., structured activities for the residents, activities for the residents to feel part of the community, opportunities for the residents to go into different places in the community, a physical environment that is adapted to the needs of adults with autism spectrum, staff with specialist autism spectrum training, ways to get specialist care when it is needed, ways to coordinate services with other providers in the area, if needed, support for employment, support for independent living), 63% of professionals said that it was in place or coming.

Somewhat lower proportions of adults and carers reported that they experienced each recommended service provision with their residential service. More than 50% of carers reported: structured activities (63.4%), staff had specialist autism training (59.2%), the adult had opportunities to go into the community (58.8%), physical environment was adapted to the adult's needs (58.8%), and the adult had activities to feel part of the community (55%) (**Supplementary Data 10**).

However, less than 50% of autistic adults or carers experienced: ways to get specialist care when needed (adults: 18.2%, carers: 44.9%), services coordination (adults: 32.1%, carers: 40.7%), support for employment (carers only, 37.5%), and support for independent living (carers only, 44.4%). Additionally, less than 50% of adults experienced the following (although many also said the item was not applicable to them): activities to feel part of the community (39.4%), the physical environment was adapted to their needs (38.6%), and staff had specialist autism training (47.4%) (**Supplementary Data 10**).

Waiting Times

Adults' and carers' reports regarding waiting times for employment, education, financial and social services were very highly concordant and were for the most part < 1 month or 1–3 months. Adults' and carers' reports regarding waiting times for residential services, however, were more disparate and the time choices were more variable across the different types of residential services. Professionals, for the most part, did not know the waiting times for services (50% or more selected "don't know") and when they did report a waiting time it generally was not concordant with the adults' and carers' reports (**Supplementary Data 11**).

Staff Training

At the offices where autistic adults and carers went to apply for a given service (residential, employment, education, financial, or social), less than 50% of both groups said that the staff seemed knowledgeable about autism or autism services. On the other hand, 50% or more said that only some or none of the staff seemed knowledgeable about autism spectrum or autism spectrum services.

At the residential facility itself, 47.4% of adults and 59.2% of carers said that staff had special training in autism (**Supplementary Data 12**), while 79% of professionals answered "yes" to this feature: Do you have knowledge of and work experience in residential services that are currently available for adults, including autistic adults?

Good Local Models

Across the five different services domains, the survey included a question asking the respondent if they knew of a good local model for the service, for example, in the residential service area, employment or education for autistic adults. Supplementary Data 13 presents the results from these questions, summarized over all five domains. Across each of the five service area domains, generally less than 34.7% of adults and carers knew of a good local service model. The domain with the lowest proportions of "yes" among adults (9.9%) and carers (14.6%) was financial service; employment service (9.5%) and adult education service (10%) were also low among autistic adults. Higher proportions of professionals knew of good local models, although for two of the five domains less than 50% of professionals knew of a good local model (financial service: 36.5%; social support service: 47.4%). The domain with the highest proportions of "yes" for a good local model was the residential service domain (autistic adults: 17.48%; carers: 34.7%; professionals: 72.6%). Another striking feature was that large proportions of respondents answered "don't know" to the questions, indicating that knowledge of good local models of service is not high, even among professionals (**Supplementary Data 13**).

DISCUSSION

The availability of autism services and unmet needs should be tracked to inform ongoing, coordinated system actions where service users and providers are key and active players in the process. In this study, we asked autistic adults, carers, and professionals about what they want, have, or need regarding services. The study results inform our understanding of several general features of the services infrastructure for autistic adults with the added advantage of looking at a variety of service types. For example, adults and carers were most likely (over 60%) to apply for a service at a public office although, according to the majority of respondents, the staff at the organizations where they applied for a given service did not seem knowledgeable about autism. The employment, education, and financial services they received were typically NOT set up for autistic persons specifically while residential and social services most likely were. These results should be noted as a gap in the service provision and are reinforced by the Cascio and Racine (16) results where most respondents preferred autism-specific services, especially for autism-specific support groups, residential services, and social/recreational groups as well as autism-specific early intervention and general day services.

While all service types had been sought for or received by large proportions of adults or carers (although < 50% in each case) in the last 2 years, the largest proportions of respondents had sought (and failed) or received financial services.

Answers on success of service provision, varied considerably across different countries and difficulties emerged in specific countries. For example, responders living in Italy reported more often that they had tried to get a service and failed (autistic adults: residential, employment, and education services; carers: financial and social support services). Responders living in France reported this challenge for financial and social support services (autistic adults), education and social support services (carers). Responders living in the Republic of Ireland reported this challenge for education (autistic adults and carers), financial and social support services (carers). Sample sizes by country were also quite variable, which could be contributing to the variability in the results. Uneven sample size across different countries with different health/social care systems means that the overall summary mean averages may not apply more broadly across Europe and we need to know more about access to health and social care provision in the countries underrepresented in the current survey.

There were fairly consistent results across all respondent groups in terms of the specific services which they had sought for, received, or would best suit them now, e.g., "help in own home" among the different type of residential services, whereas respondents' satisfaction with their current service varied widely by type of service and respondent group: the highest proportion was found among the autistic adults regarding satisfaction with their residential service and the lowest was found among carers of high independence adults regarding satisfaction with their social services. There were very similar distributions of responses by gender of the autistic adults regarding services availability and preferences, except in the case of financial services.

Residential Services

Forty percent of the autistic adults were satisfied with their current residential situation and "help in own home" was by far the most common and preferred residential service choice for autistic adults. Availability and preference/need for residential services of carers appeared to depend in part on the level of independence of the adult: "help in home" for high independence and "full time residential facility" for carers of low independence adults. The proportion of carers satisfied with what they had was low (31% for high independent adults; 23% low independence adults). Perhaps reflecting an under-met need, professionals also most often selected "help in own home" as well as "full time residential facility with full apartment" as the residential service most needed. In the United States (US), it has been observed that it is more common for autistic adults to live in a family member's home and less common to live in agency apartments, in their own home or an "other living arrangement" (22). Nevertheless, in a long-term prospective follow-up study of a population-based cohort (n = 120), very few families (n = 3, 3.6%) wanted to keep their loved ones at home as long as possible (23). An interview study suggested that what was considered "best" depended on the family member in question: mothers perceived that the adult living in the family home was the best option for the family, while living in a residential facility was the best arrangement for the autistic adult (24). From the adult's family perspective, 12 families (13%) in the Billstedt et al. (23) study reported the need for respite care "to cope with the situation, to provide a welcome break, to help with transition from home and to enable the individual and his or her family to get used to separations" (23).

Employment Services

For adults and carers, "job mentors" was the employment service most likely to be selected across the different questions of employment service availability or preference/need. According to the professionals, the service option that was least likely to be currently available was "job placement specific for autistic persons/employer programs to encourage employment of persons with autism," while the top choice for what was most needed was "job mentors." Other studies have reported the importance of "worksite peer mentors" who are constantly present at work to help the autistic adult with social interactions and other problems (25–27) (versus job mentors who are not always present at the job and may be less knowledgeable about work requirements and social interactions/environment; (25)). Another study highlighted the success of a close cooperation between the autistic adult job coaches and employers in exploring targeted job opportunities and finding job duties appropriate to the adult's abilities (28).

Adult Education Services

For adults and carers, "mentorship or specialist support in a regular education setting" was the education service most likely to be selected across the different questions of education service availability or preference/need. For carers of low independence adults, "day school or college for adults with autism" was the education service most likely to be selected among the kind of adult educational service the adult got. Along similar lines, the service options that the professionals reported were selected as top choices for what was most needed were "mentorship or specialist support in regular education settings" and "day school or college for adults with autism spectrum." The preferences for mentorship in regular education settings or autism-specific facilities likely reflect the support needs to address challenges of autistic students in higher education settings that require increasing independence, organization and time management, social relationships, unexpected changes, and sensory and academic demands (29–36). Other investigators have advocated for autism spectrum-specific support provided by higher education organizations (37, 38).

Financial Services

For adults and carers, the most common financial services of choice seemed to concern financial support in lieu of employment ("full pension") or to supplement employment earnings, either by the adult or carers, or for support during education/job training. For professionals, the top choices for needed financial services also revolved around employment: "supported employment" or "supplementary income for persons unable to have full employment." The focus in these results on financial support due to no or limited earnings from employment is interesting in view of a recent literature review of the high costs associated with autism which highlighted a considerable array of potential costs: medical and healthcare service costs, therapeutic costs, (special) education costs, costs of informal care by family/caregivers, costs of accommodation, respite care, and out-of-pocket expenses, as well as costs of lost productivity by the adult or family/caregivers (39). Thus, perhaps the shortfalls in income from employment that limit the ability to pay for their diverse needs are a main financial concern of autistic adults or their carers. This concern is reflected regardless of the gender of the autistic respondent.

Social Support Services

Although "life skills training" and "free time activities" were consistently chosen by most adults, carers, and professionals across the questions of social services availability and preference/need, another top preference was "behavior training for an individual" and, for carers of higher independence adults, different forms of "peer to peer matching." While during adolescence, autistic people may show increased interest in social relationships and in developing social skills [e.g., (40, 41)], most individuals continue to show social impairment in adolescence and adulthood (42, 43). This study's results appear to underscore the perceived need to improve poor social skills and to have targeted social skills services integrated into the care pathway of autistic adults.

Alignment With Residential Services' Guidelines

The National Institute for Health and Care Excellence autism guidelines [(21), updated in 2021] advises that the residential environment should be "structured to support and maintain a collaborative approach between the autistic person and their family, partner or carer(s) for the development and maintenance of interpersonal and community living skills" (21). The NICE guidelines also concern the residential care activities, care environments and care staff characteristics. The present study investigated the user and professionals' experiences around these aspects.

From the perspective of the adults and carers in this study, only 2 of 8 recommendations were experienced by the majority (>50%) of the adults and only 5 of 10 recommendations were experienced by the majority (>50%) of carers: structured activities for the residents and opportunities for the residents to go into different places in the community (for adults and carers), activities for the residents to feel part of the community, a physical environment that is adapted to the needs of adults with autism spectrum, staff with specialist autism spectrum training (for carers). The other recommended services provisions experienced by less than half of respondents were "ways to get specialist care when needed," "services coordination," "support for employment," "support for independent living," "activities to feel part of the community," "residential physical environment was adapted to their needs," and "residential staff had specialist autism training." The present results parallel the findings of Scattoni et al. (15) where a lack of alignment was observed between the user experiences and guidelines on recommended characteristics for post-diagnostic support for autistic adults (15).

Waiting Times for Services

According to the adult and carer responses, waiting times for residential services seemed to be more variable than waiting times for employment, education, financial, or social services; the waiting times for the latter were for the most part less than 1 month or 1 to 3 months. Professional respondents were not well informed about waiting times for these types of services (majority answering "Don't know"). There is little literature on services for autistic adults waiting times; the available literature focuses on child services waiting times. Caldwell and Heller (44) reported that families on waiting lists for services for developmental disabilities showed more unmet needs and lower service satisfaction than families supported by services. If longer waiting times indicate both a large demand and limited supply of the service, then this study's results indicate a most limited availability for residential services for autistic adults that appears to be larger than that for other types of services.

Professionals, for the most part, reported that they did not know the waiting times for services and when they did report a waiting time it generally was not in parallel with the users' reports. Users should be accompanied by an autism-trained care manager through the process of requesting and waiting for residential service - which should be as short as possible. In addition, professionals should be aware of the time that users and their families spend awaiting care so that they can best accommodate them in the service.

Service Staff Training

The perception of more than half of autistic adults and carers was that only some or none of the employees in offices where services are applied for seemed knowledgeable about autism or autism services. The value of staff training was supported by a study of McDonnell et al. (45) that showed that a 3-day training course increased residential social care and day service staff confidence in managing aggression in autistic people. The present study appears to indicate considerable shortfalls in training of services staff for autistic adults even though staff training improves service quality for the clients and benefits the staff persons themselves. It is particularly important that employees in the offices where autistic adults and their carers apply for services know about autism and its needs in adulthood so that they can best refer and advise them about the services available locally.

Good Local Service Models

Although knowledge of good local services models that work well for autistic adults was generally low across all services areas, residential services had the highest proportions of respondents with positive responses. Large proportions of respondents answered "don't know" to the questions, indicating that knowledge of good local models of service is not high, even among professionals. These results underscore both the complex nature of the services infrastructure needed for autistic adults and opportunities for improvement.

This generally low positive response rate may reflect a possible lack of good services models for autistic adults in the local community or, possibly, a critical information gap among both users and professionals. It is recommended that local care pathways are understandable, accessible, acceptable for users and providers, consider the person's knowledge and understanding of autism and its care and be appropriate to the local communities [(21), updated in 2021; 2014], and relevant professionals should know local autism care pathways and the way to access services (46).

Limitations

The present results should be interpreted in view of the study limitations. First, the survey data were collected by convenience sampling thus it is likely that there may be selection bias since only people already in a support network and with internet access had access to the survey. Second, most of the autistic adult respondents were female [as found in on-line surveys in general; (47)], thus the results may poorly reflect the autistic males' experiences and preferences. Third, most of the responders were living in cities that are not capital cities having <1.000, 1.000–20.000 or 20.000–100.000 inhabitants, thus the results may apply to specific contexts, with less services availability than found in capital cities. Fourth, the survey did not ask for important clinical information from the responders (e.g., the psychosocial functioning, illness severity or current treatment) apart from the question for the carers regarding level of independence of the autistic adult asked of the carers. Future studies should seek to ask participants for more detailed information about their clinical background to determine the impact on services use. Fifth, age specific analysis (e.g., different experiences with service options by age at diagnosis) was not feasible due to sample size limitations, especially because data were also stratified by gender. Finally, although we considered the level of independence of the carer's autistic adult, we could not fully account for the developmental level of the autistic adult which is a strong predictor of service usage and needs (48).

CONCLUSION

The ASDEU survey sheds light on the state of different types of autistic adult services in 11 countries of the European Union from the perspective of autistic adults, carers and professionals. The results underscore the highly diverse needs and preferences of the autistic community such that the different residential, employment, educational, financial, and social services options that adults or their carers tried to get and failed, or succeeded in receiving, or believed would better suit their current needs varied depending on the respondent group and the level of independence of the autistic adult in question. Although there was some concordance between professional and adult/carer responses in terms of the availability and preferences/needs of different services options, the differences are also important to note since they highlight the different perspectives of the professional providers versus the adult/carer services users. Thus, for services policy planning, it is important to consider the perspectives of all these types of respondents when assessing autistic adult services needs and gaps in the community (49).

The results also highlight specific gaps in knowledge: lack of autism knowledge and training in services office staff, shortfalls in meeting published recommendations around residential services (which may reflect lack of knowledge about them by the service provider), professionals' lack of awareness of services waiting times, and lack of knowledge by all respondent groups of good local services models that work well for autistic adults. Filling these knowledge gaps may be important steps toward more equitable service delivery and better support for the autistic adult community.

DATA AVAILABILITY STATEMENT

The datasets presented in this article are not readily available because we can only share aggregated data (not individual level), and any data based on a small sample size (<5) may not be shared as an extra data privacy precaution. Requests to access the datasets should be directed to DS, diana.schendel@ph.au.dk and MS, marialuisa.scattoni@iss.it.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Each ASDEU site obtained local ethical approval. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

MLS, TS, MM, and DS: formal analysis. MM, MLS, and DS: writing – original draft preparation. MP: funding acquisition for the survey dissemination and data collection and analysis (DGSANCO) and principal investigator. MLS: funding acquisition for the Italian participation at the survey, data analysis and writing ("Osservatorio Italiano per il monitoraggio dei disturbi dello spettro autistico" and "I disturbi dello spettro autistico: attività previste dal decreto ministeriale del 30.12.2016") and principal investigator. All authors contributed to the conceptualization, investigation, and writing – review and editing and have read and agreed to the published version of the manuscript.

FUNDING

The present study has been funded by the European Parliament and managed by the European Union, DG SANTE (Ref. SANCO/2014/C2/035), the Italian Ministry of Health projects "Osservatorio Italiano per il monitoraggio dei disturbi dello spettro autistico" (Fasc. 1S49), and the collaboration agreement with the Ministry of Health – General Directorate of Health Prevention and General Directorate of Health Programming, registered by the Court of Auditors on 3.6.2019 with note no. 1-2105 for the implementation of the project "Definizione di criteri e modalità di utilizzo del fondo per la cura di soggetti con disturbo dello spettro autistico" (Fasc. 5S13).

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2022.919234/full#supplementary-material

REFERENCES

1. American Psychiatric Association. *Diagnostic and Statistical Manual of Mental Disorders*. 5th ed. Washington, DC: American Psychiatric Association (APA) (2013).
2. Howlin P, Goode S, Hutton J, Rutter M. Adult outcome for children with autism. *J Child Psychol Psychiatry.* (2004) 45:212–29. doi: 10.1111/j.1469-7610.2004.00215.x
3. Howlin P, Moss P, Savage S, Rutter M. Social outcomes in mid-to later adulthood among individuals diagnosed with autism and average nonverbal IQ as children. *J Am Acad Child Adolesc Psychiatry.* (2013) 52:572–81. doi: 10.1016/j.jaac.2013.02.017
4. Woolfenden S, Sarkozy V, Ridley G, Williams K. A systematic review of the diagnostic stability of autism spectrum disorder. *Res Autism Spectr Disord.* (2012) 6:345–54. doi: 10.1016/j.rasd.2011.06.008
5. Bishop-Fitzpatrick L, Hong J, Smith LE, Makuch RA, Greenberg JS, Mailick MR. Characterizing objective quality of life and normative outcomes in adults with autism spectrum disorder: an exploratory latent class analysis. *J Autism Dev Disord.* (2016) 46:2707–19. doi: 10.1007/s10803-016-2816-3
6. Shattuck PT, Roux AM, Hudson LE, Taylor JL, Maenner MJ, Trani JF. Services for adults with an autism spectrum disorder. *Can J Psychiatry.* (2012) 57:284–91. doi: 10.1177/070674371205700503

7. Anderson C, Butt C. Young adults on the autism spectrum: the struggle for appropriate services. *J Autism Dev Disord.* (2018) 48:3912–25. doi: 10.1007/s10803-018-3673-z
8. Crane L, Batty R, Adeyinka H, Goddard L, Henry LA, Hill EL. Autism diagnosis in the United Kingdom: perspectives of autistic adults, parents and professionals. *J Autism Dev Disord.* (2018) 48:3761–72. doi: 10.1007/s10803-018-3639-1
9. Hurt L, Langley K, North K, Southern A, Copeland L, Gillard J, et al. Understanding and improving the care pathway for children with autism. *Int J Health Care Qual Assur.* (2019) 32:208–23. doi: 10.1108/IJHCQA-08-2017-0153
10. Jones L, Goddard L, Hill EL, Henry LA, Crane L. Experiences of receiving a diagnosis of autism spectrum disorder: a survey of adults in the United Kingdom. *J Autism Dev Disord.* (2014) 44:3033–44. doi: 10.1007/s10803-014-2161-3
11. Lewis LF. A mixed methods study of barriers to formal diagnosis of autism spectrum disorder in adults. *J Autism Dev Disord.* (2017) 47:2410–24. doi: 10.1007/s10803-017-3168-3
12. Micai M, Ciaramella A, Salvitti T, Fulceri F, Fatta LM, Poustka L, et al. Intervention services for autistic adults: an ASDEU study of autistic adults, carers, and professionals' experiences. *J Autism Dev Disord.* (2021) 52:1623–1639. doi: 10.1007/s10803-021-05038-0
13. Micai M, Ciaramella A, Salvitti T, Fulceri F, Fatta LM, Poustka L, et al. Autistic adult health and professional perceptions of it: evidence from the ASDEU project. *Front Psychiatry.* (2021) 12:614102. doi: 10.3389/fpsyt.2021.614102
14. Raymond-Barker P, Griffith GM, Hastings RP. Biographical disruption: experiences of mothers of adults assessed for autism spectrum disorder. *J Intellect Dev Disabil.* (2018) 43:83–92. doi: 10.3109/13668250.2016.1262011
15. Scattoni ML, Micai M, Ciaramella A, Salvitti T, Fulceri F, Fatta LM, et al. Real-world experiences in autistic adult diagnostic services and post-diagnostic support and alignment with services guidelines: results from the ASDEU study. *J Autism Dev Disord.* (2021) 51:4129–46. doi: 10.1007/s10803-021-04873-5
16. Cascio MA, Racine E. Autism service preferences of parents/guardians and autistic adults in five countries. *Autism Res.* (2022) 15:570–85. doi: 10.1002/aur.2667
17. Mukaetova-Ladinska EB, Stuart-Hamilton I. Adults with autism spectrum disorder (ASD): service user perspective on ageing transition. *Columbia Int Publ Am J Autism*. (2016) 2:1–11. doi: 10.7726/aja.2016.1001
18. Autism Europe. *Towards a Better Quality of Life: The Rights of Ageing People With Autism*. (2013). Available online at: http://www.autismeurope.org/files/files/2012-European-Parliament-event/ageingreport-en-sml.pdf (accessed July 28, 2021).
19. Kendall T, Megnin-Viggars O, Gould N, Taylor C, Burt LR, Baird G. Management of autism in children and young people: summary of NICE and SCIE guidance. *BMJ.* (2013) 347:f4865. doi: 10.1136/bmj.f486523985309
20. National Audit Office. *Supporting People With Autism Through Adulthood.* (2009). Available online at: http://www.nao.org.uk/report/supporting-people-with-autism-through-adulthood/ (accessed January 25, 2019).
21. NICE. *Autism Quality Standard. England*. (2012). Available online at: https://www.nice.org.uk/guidance/cg142/documents/autistic-spectrum-conditions-in-adults-full-guideline2 (accessed July 28, 2021).
22. Hewitt AS, Stancliffe RJ, Hall-Lande J, Nord D, Pettingell SL, Hamre K, et al. Characteristics of adults with autism spectrum disorder who use residential services and supports through adult developmental disability services in the United States. *Res. Autism Spectr. Disord.* (2017) 34:1–9. doi: 10.1016/j.rasd.2016.11.007
23. Billstedt E, Gillberg IC, Gillberg C. Aspects of quality of life in adults diagnosed with autism in childhood: a population-based study. *Autism.* (2011) 15:7–20. doi: 10.1177/1362361309346066
24. Krauss MW, Seltzer MM, Jacobson HT. Adults with autism living at home or in non−family settings: positive and negative aspects of residential status. *J Intellect Disabil Res.* (2005) 49:111–24. doi: 10.1111/j.1365-2788.2004.00599.x
25. Coleman DM, Adams JB. Survey of vocational experiences of adults with autism spectrum disorders, and recommendations on improving their employment. *J Vocat Rehabil.* (2018) 49:67–78. doi: 10.3233/JVR-180955
26. Hurlbutt K, Chalmers L. Employment and adults with asperger syndrome. *Focus Autism Other Dev Disabil.* (2004) 19:215–22. doi: 10.1177/10883576040190040301
27. Müller E, Schuler A, Burton BA, Yates GB. Meeting the vocational support needs of individuals with asperger syndrome and other autism spectrum disabilities. *J Vocat Rehabil.* (2003) 18:163–75.
28. Mawhood L, Howlin P. The outcome of a supported employment scheme for high-functioning adults with autism or asperger syndrome. *Autism.* (1999) 3:229–54. doi: 10.1177/1362361399003003003
29. Fleischer AS. Alienation and struggle: everyday student-life of three male students with asperger syndrome. *Scand J Disabil Res.* (2012) 14:177–94. doi: 10.1080/15017419.2011.558236
30. Gelbar NW, Smith I, Reichow B. Systematic review of articles describing experience and supports of individuals with autism enrolled in college and university programs. *J Autism Dev Disord.* (2014) 44:2593–601. doi: 10.1007/s10803-014-2135-5
31. Kapp SK, Gantman A, Laugeson EA. Transition to adulthood for high-functioning individuals with autism spectrum disorders. In: Mohammadi MR editor. *A Comprehensive Book on Autism Spectrum Disorders*. London: InTech (2011). p. 451–78. doi: 10.5772/21506
32. Pinder-Amaker S. Identifying the unmet needs of college students on the autism spectrum. *Harv Rev Psychiatry.* (2014) 22:125–37. doi: 10.1097/HRP.0000000000000032
33. Madriaga M. 'I avoid pubs and the student union like the plague': students with asperger syndrome and their negotiation of university spaces. *Child Geogr.* (2010) 8:39–50. doi: 10.1080/14733280903500166
34. Madriaga M, Goodley D. Moving beyond the minimum: socially just pedagogies and asperger's syndrome in UK higher education. *Int J Incl Educ.* (2010) 14:115–31. doi: 10.1080/13603110802504168
35. Taylor MJ. Teaching student with autistic spectrum in HE. *Educ Train.* (2005) 47:484–95. doi: 10.1108/00400910510626330
36. Van Hees V, Moyson T, Roeyers H. Higher education experiences of students with autism spectrum disorder: challenges, benefits and support needs. *J Autism Dev Disord.* (2015) 45:1673–88. doi: 10.1007/s10803-014-2324-2
37. Friedman NDB, Warfield ME, Parish SL. Transition to adulthood for individuals with autism spectrum disorder: current issues and future perspectives. *Neuropsychiatry.* (2013) 3:181–92. doi: 10.2217/NPY.13.13
38. Van Bergeijk E, Klin A, Volkmar F. Supporting more able students on the autism spectrum: college and beyond. *J Autism Dev Disord.* (2008) 38:1359–70. doi: 10.1007/s10803-007-0524-8
39. Rogge N, Janssen J. The economic costs of autism spectrum disorder: a literature review. *J Autism Dev Disord.* (2019) 49:2873–900. doi: 10.1007/s10803-019-04014-z
40. Mesibov GB, Handlan S. Adolescents and adults with autism. 2nd ed. In: Cohen DJ, Volkmar FR editors. *Handbook of Autism and Pervasive Developmental Disorders*. New York, NY: John Wiley & Sons (1997). p. 309–22.
41. Volkmar FR, Klin A. Social development in autism: historical and clinical perspectives. In: Baron-Cohen S, Tager-Flusberg H, Cohen DJ editors. *Understanding Other Minds: Perspectives From Autism*. New York, NY: Oxford University Press (1995). p. 40–55.
42. Church C, Alinsanski S, Amanullah S. The social, behavioral, and academic experiences of children with asperger syndrome. *Focus Autism Other Dev Disabil.* (2000) 15:12–20. doi: 10.1177/108835760001500102
43. Seltzer MM, Krauss MW, Shattuck P, Orsmond GI, Swe A, Lord C. Changes in the symptoms of autism in adolescence and adulthood. *J Autism Dev Disord.* (2003) 33:565–81. doi: 10.1023/B:JADD.0000005995.02453.0b
44. Caldwell J, Heller T. Longitudinal outcomes of a consumer-directed program supporting adults with developmental disabilities and their families. *Intellect Dev Disabil.* (2007) 45:161–73.
45. McDonnell A, Sturmey P, Oliver C, Cunningham J, Hayes S, Galvin M, et al. The effects of staff training on staff confidence and challenging behavior in services for people with autism spectrum disorders. *Res Autism Spectr Disord.* (2008) 2:311–9. doi: 10.1016/j.rasd.2007.08.001
46. NICE. *Autism Quality Standard. England.* (2014). Available online at: https://www.nice.org.uk/guidance/qs51/resources/autism-pdf-2098722137029 (accessed March 4, 2020).

47. Smith G. *Does Gender Influence Online Survey Participation: A Record-Linkage Analysis of University Faculty Online Survey Response Behavior*. ERIC Document Reproduction Service No. ED 501717. San Jose, CA: San José State University (2008)

48. Maenner MJ, Smith LE, Hong J, Makuch R, Greenberg JS, Mailick MR. Evaluation of an activities of daily living scale for adolescents and adults with developmental disabilities. *Disabil Health J*. (2013) 6:8–17. doi: 10.1016/j.dhjo.2012.08.005

49. Shattuck PT, Garfield T, Roux AM, Rast JE, Anderson K, Hassrick EM, et al. Services for adults with autism spectrum disorder: a systems perspective. *Curr Psychiatry Rep*. (2020) 22:13. doi: 10.1007/s11920-020-1136-7

Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers | Frontiers in Psychiatry

SYSTEMATIC REVIEW
published: 29 June 2022
doi: 10.3389/fpsyt.2022.914750

Early Autism Intervention Components Deliverable by Non-specialists in Low- and Middle-Income Countries: A Scoping Review

***Lavangi Naithani*[1], *Caitlin Goldie*[2], *Abhipreet Kaur*[1], *Charlotte Butter*[2], *Shweta Lakhera*[1], *Kathy Leadbitter*[2†] and *Gauri Divan*[1*†]**

[1] Sangath, Goa, India, [2] Division of Neuroscience and Experimental Psychology, The University of Manchester, Manchester, United Kingdom

Introduction: The past decade has seen key advances in early intervention for autistic children in high-income countries, with most evidence based on specialist delivery of interventions. The care gap seen in low- and middle-income countries (LMIC) remains close to 100%. A key challenge in addressing this care gap concerns the paucity of specialists available to deliver services. Task-sharing provides an important potential solution; there is a need to identify interventions that are suitable for scaled-up delivery through task-sharing in low-resourced settings. We aimed to conduct a scoping review to identify studies which reported autism intervention delivered by non-specialists within LMIC and, using established frameworks, specify intervention components with evidence of successful non-specialist delivery.

Methods: A scoping literature search, conducted within four databases, generated 2,535 articles. Duplicates were removed, followed by screening of titles and abstracts, with 10% double-rated for reliability. 50 full text articles were then screened independently by two raters. Articles were included if studies: (a) were conducted in LMIC; (b) included samples of autistic children (age < 10); (c) evaluated psycho-social interventions delivered by non-specialists; (d) reported child outcomes; and (e) were peer-reviewed full-texts in English. Two established frameworks – @Practicewise and NDBI-Fi framework - were then used to ascertain the commonly delivered components of these interventions.

Results: Two studies met the inclusion criteria. Both studies evaluated parent-mediated interventions delivered by non-specialists in South Asia. Through the two frameworks, we identified elements and techniques that had been delivered successfully by non-specialists.

Conclusion: There is evidence from two acceptability and feasibility trials that non-specialists can be trained to deliver some intervention elements and techniques within parent-mediated interventions, with good fidelity and acceptability and evidence of effectiveness. The review points up the lack of a widespread evidence base in this area

Edited by:
Diana Schendel,
Drexel University, United States

Reviewed by:
Aldina Venerosi,
National Institute of Health (ISS), Italy
Olivia Herrington,
Yale University, United States

***Correspondence:**
Gauri Divan
gauri.divan@sangath.in

† These authors share last authorship

Specialty section:
This article was submitted to Autism,
a section of the journal
Frontiers in Psychiatry

Received: *07 April 2022*
Accepted: *23 May 2022*
Published: *29 June 2022*

Citation:
Naithani L, Goldie C, Kaur A, Butter C, Lakhera S, Leadbitter K and Divan G (2022) Early Autism Intervention Components Deliverable by Non-specialists in Low- and Middle-Income Countries: A Scoping Review. Front. Psychiatry 13:914750. doi: 10.3389/fpsyt.2022.914750

and need for further research in low resourced settings, including well-powered trials and mechanistic analyses to identify active ingredients. A focus on the pre-requisites for non-specialist delivery is critical to reduce inequity and provide universal health coverage within resource-constrained health systems.

Keywords: low- and lower-middle-income countries, non-specialist delivery, autism intervention components, autism spectrum disorder (ASD), low resource setting

INTRODUCTION

Autism Spectrum Disorder, more commonly referred to as autism, is a lifelong neurodevelopmental disability characterized by impairments in social communication, restricted, repetitive interests and behaviors, and differences in sensory processing, with the presence of co-occurring intellectual disability in up to a third of individuals (1). A review of global prevalence reported 1–1.8% (2), with similar findings in studies from low- and middle-income countries (LMIC), with prevalence rates of 1.04 and 1.1% in Sri Lanka and India, respectively (3, 4). In the majority of LMIC, there exist a number of obstacles to accessing care which include the low awareness of the condition and the lack of access to detection and treatment services. Many LMIC have little or no policy implementation and underfunded health care systems for child and adolescent services (5, 6). This results in a significant "detection" gap in the identification of autism in young children, as well as a "treatment gap" with a lack of evidence-based interventions for the children who do receive a diagnosis.

Low- and middle-income countries have a significant proportion of their populations in the younger demographic age groups. This, coupled with large detection and treatment gaps, makes the need to identify interventions that can be scaled up a critical component of the right to care for young children with autism but also for the attainment of the United Nations Sustainable Development Goal 3. This goal aims to ensure the healthy lives and the promotion of health for all at all ages (7). While some LMIC settings do have services, these are predominantly focused in metropolitan areas and remain impacted by the scarcity of specialists with expertise in child development, including developmental pediatricians, child psychiatrists, speech and language therapists or occupational therapists (6). There is a need for radical innovations to help scale up services in these low resource contexts. Across many areas of health, including within treatment for mental health conditions like depression, innovations have used the process of "task-sharing" to address this dearth of specialist care (8). This approach aims to systematically skill up a non-specialist or para-professional with intervention-specific knowledge and skills, such that they are competent to facilitate the delivery of an evidence-based intervention in the community. Task-sharing is usually supported by a strong supervisory framework: the non-specialist has focused knowledge in a specific intervention or area of care, but more expert advice or care is readily available in the event of complex cases or scenarios. This approach allows an intervention to be delivered by a non-specialist, with the mobility to reach out to families in the community. Thus task-sharing with accessible non-specialist community-based facilitators overcomes barriers such as the limitation of specialist services, the expenses of such services, and the geographical distance to services.

The aim of this study was to conduct a scoping review and identify interventions for young children with autism that have evidence of feasibility, acceptability and effectiveness when delivered by non-specialists in LMIC. We adopted a narrow definition of "non-specialist" informed by the availability of personnel for task sharing to deliver care to children and families in LMIC settings (9), rather than the workforce available within low-resourced settings in high-income countries. These providers are typically frontline community health workers (10) who are not specialists in child development, autism, or mental health but who are already working on the ground within local communities. Examples include the Accredited Social Health Activists working in Delhi (11), Female Community Health Volunteers in Nepal (12), and Community Health Volunteers working in Kenya (13). Other potential providers include community volunteers or parent/disability "champions". Our definition of specialist providers included those specialist professionals largely unavailable within LMIC: pediatricians, psychiatrists, nurse practitioners, psychologists, speech and language therapists, occupational therapists, physical therapists, and students/trainees of these professions. We focused on interventions delivered within a family context rather than school-based or teacher-delivered interventions, which tend to center around educational outcomes. In addition, we sought to address inconsistency in the definition around non-specialist delivery found within the literature. We differentiated between non-specialist-*mediated* interventions, and non-specialist *delivery*, the latter being the focus of our search. The former includes a broad range of approaches in which a practitioner delivers the intervention to a dyad (e.g., parent-child, sibling-child, peer-child) and the agent of mediation between the practitioner and autistic child (e.g., parent, sibling, peer) is considered a non-specialist. Some reviews (14) identified the parent receiving the intervention as the non-specialist and included all studies of parent-mediated interventions even where the intervention was delivered to the parent by a specialist. To meet our definition, parent-mediated interventions were included only when delivery to the parent-child dyad was by a non-specialist. This was a purposeful distinction to ensure that the focus was on interventions deliverable to families by non-specialists in LMIC. We also excluded interventions which were delivered by both specialists and non-specialists in tandem (15).

Following the identification of studies meeting our criteria, we sought to characterize the common elements (defined as a therapeutic activity or strategy) and techniques (defined as skills used during a session to deliver an element) of interventions that have been delivered by non-specialists under supervision and evaluated in LMIC countries. We were guided by two published frameworks: (1) the model proposed by Chorpita et al. (16) for delineating common ingredients of interventions for young children and adolescents which allows for the design of scalable evidenced modular interventions; (2) the framework developed by Frost and colleagues (17) through an expert-led Delphi approach, which aims to characterize the common elements and components of naturalistic developmental behavioral interventions (NBDIs) for autism.

METHODS

We followed the five-step framework for scoping reviews suggested by Levac and colleagues (18). We firstly defined the research question and then identified the relevant studies based on predetermined inclusion and exclusion criterion. We then charted and summarized the data. We describe these steps in detail below.

An OVID literature search was conducted within four databases (APA PsychInfo, APA Psycharticles, Ovid Medline + Epub Ahead of Print, Embase) to identify relevant studies. Search terms are shown in **Table 1**. This search identified 1,721 articles after 808 duplicates were identified and removed within OVID.

These articles were imported into Rayyan (19), a free web application designed for collaborative reviews. Rayyan identified 24 potential duplicates. These were checked manually and 6 were deleted. To establish inter-rater reliability prior to the title and abstract screen, 10% (173) were screened by all five reviewers, maintaining blind to each other's inclusion/exclusion decisions. Agreement was reached for 130 articles (75%). Disagreements and uncertainties were resolved through discussion and clarification of terminologies and inclusion/exclusion criteria with support from two senior authors, until consensus was reached. Of these articles, 166 were excluded and 7 included for full-text review. The remaining 1,542 articles were divided up between five raters (AK, SL, LN, CB, and CG). All raters had at least a relevant undergraduate degree (CB, CG) or Masters degrees or equivalent (LN, AK, and SL) and were working within autism intervention research projects. The raters screened the title and abstracts, applying the inclusion/exclusion criteria shown in **Table 2**. This resulted in 1,499 articles being excluded and 43 articles put forward for a full text review. Together with the articles used for inter-rater reliability, a total of 1,671 articles were excluded and 50 articles (7 from reliability screen, plus 43 from individual screen) were put forward for full text review. The full breakdown of exclusion reasons across both stages can be found in the PRISMA (**Figure 1**).

The full-text review of the 50 articles was carried out independently by two raters from the pool of five raters, applying the same inclusion/exclusion criteria. There were 6 conflicts at this stage and these were resolved through discussion with the wider coding team. This led to the exclusion of 48 articles and the inclusion of 2 articles.

TABLE 1 | OVID search terms.

Category	Search terms	Search fields
Condition	autis*, pervasive	In title
Age-group	Child*	Anywhere in article
Article Type	Primary research	
LMIC context, global health or culture	LMIC*, LAMIC*, low resource*, low-income, limited resource*, middle-income, glob*, cultur*	Anywhere in article
Delivery agent	Paraprofessional*, para-professional*, semi-professional*, lay, frontline, front-line, non-specialist*, task*sharing, aide*, assistant*	Anywhere in article
Focus on intervention	care, treatment*, therap*, intervention*, train*, service*, group*	In abstract, keywords or title

The asterix is used as a standard method of describing search terms in a review.

The next stage involved reviewing 18 articles recommended through key informants and bibliographic searches. These were divided between the 5 raters who, again, applied the same inclusion/exclusion criteria. These reviews produced two articles, which were the same articles that resulted from the search.

Common Components

Chorpita and Daleiden (20) developed a framework for identifying the components of psychosocial interventions for mental health conditions in children and adolescents which had demonstrated "favorable" results in effectiveness evaluations. The framework is descriptive in nature and does not seek to pinpoint the critical or "active" elements of these interventions. These identified elements have been collated into a coding manual called @PracticeWise (21). We examined the elements and techniques mapped in the @Practicewise common elements framework (20), and calibrated these broad definitions to encompass the needs of autism interventions along with the common components defined for the NBDI fidelity measure (17). LN and GD mapped the elements and techniques by reading the description of the intervention in the publication and manuals, plus drawing upon their professional understanding of the interventional approaches. We defined *elements* as a therapeutic activity or strategy and *techniques* as a skill that a therapist uses before or during the session to deliver an element (16).

RESULTS

Two publications met our full search criteria, see **Figure 1**. Both these papers reported well-designed evaluations of interventions for young autistic children delivered in LMICs by non-specialists with no previous training in child development. Characteristics of the studies, interventions, and delivery agents are summarized in **Table 3**. Rahman et al. (22) trialed a parent-mediated

TABLE 2 | Inclusion/exclusion criteria.

Domain	Inclusion criteria	Exclusion criteria
Language	In English language	Not in English language
Publication type	Published and peer-reviewed empirical articles	Reviews Chapters from books Correction notices Study protocols Guidelines and clinical protocols Unpublished dissertations/theses
Publication date	Published 2006–2021	Published before 2006
Research setting	Low or middle income country	High income country
Sample	Sample (recipients of intervention) includes children with a diagnosis of autism or parents/caregivers of young children with a diagnosis of autism Sample includes only or mainly children under 10 years Sample > 10 participants	Sample (recipients of intervention) is medical or educational professionals Sample contains only adults or only/mainly children above the age 10 Sample < 10 participants
Study design	Feasibility or pilot randomized controlled trials Large randomized controlled trials Pre- and post-intervention design	Epidemiological studies Cross-sectional studies Surveys Purely qualitative studies Case studies or case series Descriptive
Outcome measurement	Studies that include measurement of child outcomes relating to core features of autism (e.g., social communication, interaction, play and behavior)	Studies which report feasibility/acceptability but not outcomes Studies which report only parent/caregiver outcomes School based interventions focused on curriculum outcomes
Intervention evaluated	Psycho-social intervention targeting core features of autism	Interventions solely targeting non-core features of autism, e.g., sleep, motor control Technological interventions, e.g., robots, computer avatars etc. Pharmacological interventions Biomedical treatments e.g., diets, hyperbaric O2 etc.
Delivery agents	Intervention delivered by non-specialists (see definition)	Intervention delivered completely or partly by specialists, trainees or students of professional courses or teachers Intervention is self-directed Child peer-delivery or -mediation

social communication-focused intervention (Parent mediated intervention for Autism Spectrum Disorders in South Asia: PASS) delivered by non-specialist health workers, at home or in a clinic, in Rawalpindi, Pakistan and Goa, India. The intervention was an adaptation of the UK Pre-school Autism Communication Therapy (PACT), which was modified for delivery by non-specialists working within a competency-based training and supervision cascade led by specialists (23). The "non-specialists" were bachelor level graduates with no prior exposure to child development or autism. Precisely as in the original PACT intervention, they used video-feedback methods to enhance the parents' sensitivity to their children's non-verbal and verbal communication and refine the relevance of their responses to their child (known as parental "synchrony"). Parents were also encouraged, in later stages of the intervention process and where appropriate, to introduce action routines, repetitive language and pauses with the aim of aiding the child's communication development. Implementation of strategies was encouraged between therapy sessions. Children with a diagnosis of autism aged 2–9 years were randomly assigned (1:1) to the PASS intervention ($n = 32$) or the treatment as usual (TAU) condition ($n = 33$), with controls for age (<6 or >6), functional impairment and treatment center location (Rawalpindi or Goa). The outcome measures for this initial effectiveness trial included parental synchrony and child communicative initiations (measured on the Dyadic Communication Measure for Autism) (24), parent reports of child adaptation and language use (measured on the Vineland Adaptive Behavior Scale (25), and the McArthur Communicative Development Inventory (26), respectively), the verbal and non-verbal aspects of social communication (measured on the Communication and Symbolic Behavior Scales Developmental Profile) (27) and maternal depressive symptoms (measured using a Patient Health Questionnaire-9) (28). The intervention proved feasible and acceptable for participants (only 9% of participants were lost to follow up). Additionally, and importantly, the intervention was delivered by the non-specialists with high and sustained therapist fidelity to the intervention manual (89% of intervention sessions meeting the pre-defined threshold for acceptable fidelity). For outcomes, in comparison to TAU, PASS increased parental synchrony and the initiation of communication by the children in the dyad, but child-specific outcomes beyond dyad were not affected. Other results are reported in **Table 3**.

During the PASS trial it became apparent that co-occurring non-autistic difficulties were often un-treated due to the lack of other generic child development and health services in the community, and that these co-occurring problems such as those associated with sleep, toilet training, parental wellbeing, and dietary restrictions, could hinder access to the core PASS technique. A number of additional modules were therefore devised for incorporation when necessary, on the basis of the reasonable effectiveness evidence for each co-occurring difficulty but simplified such that they could be within the competence of non-specialists. The implementation of these was based on a clinical decision algorithm which helped the provider and parent identify the families' priority co-occurring condition and the most feasible strategies that families could implement for their child. After three sessions of the PASS social communication intervention, the non-specialist health workers supported the parent to identify the co-occurring concerns most problematic for the child and family and suggested relevant advice and strategies. Subsequent sessions were then extended to include both core social communication strategies

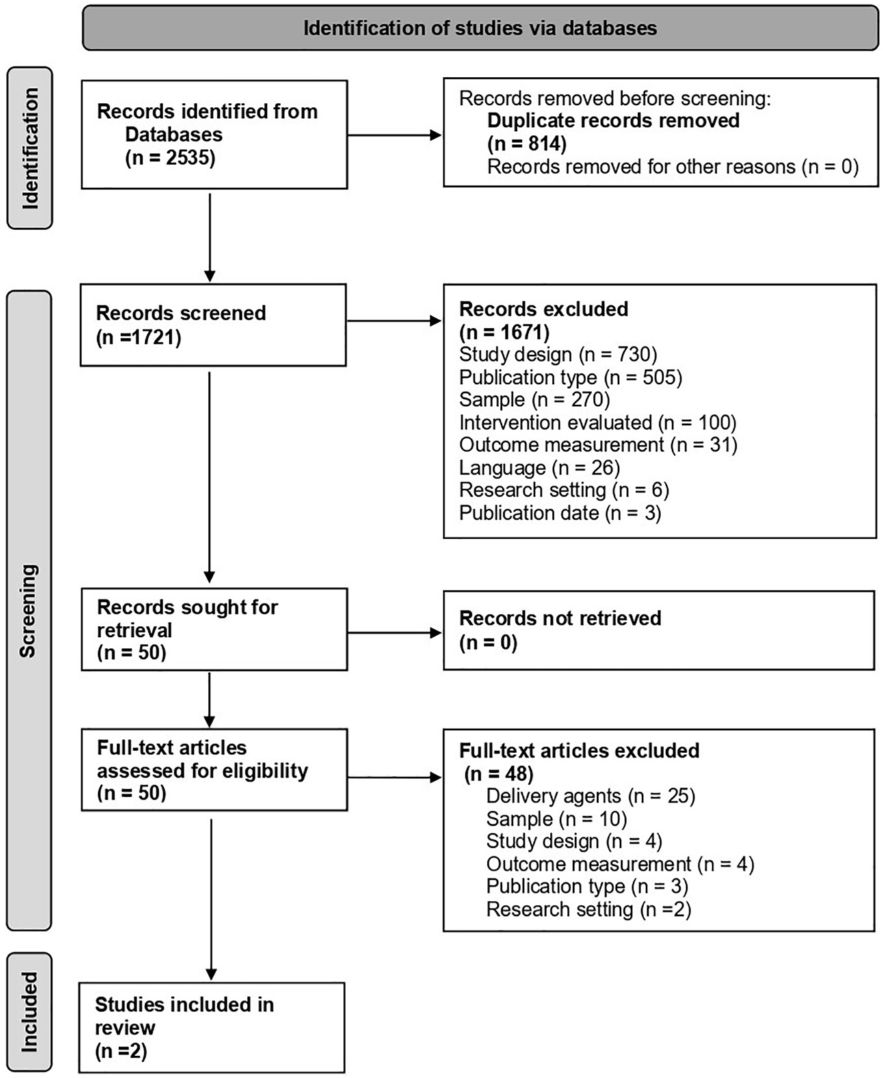


FIGURE 1 | PRISMA flow diagram of the study selction process.

and supportive strategies as appropriate and parents were given handouts to use between sessions for the latter. These sections of the session were delivered as distinct interventions, and the separate "Plus" section was only delivered after three foundational social communication sessions were complete. The distinction between core PASS and the "Plus" elements is clarified in **Table 4**. In distinction to the collaborative video-feedback facilitation of the core PASS intervention for autistic development, some of the "Plus" modules for co-occurring problems contained practical strategies for the management of day-to-day challenges to caregiving and family functioning (for instance, functional analysis of toileting and sleep difficulties). Written home programs for both components supported families to practice strategies collaboratively set during the session.

Divan and colleagues (29) evaluated this "PASS PLUS" intervention in a small effectiveness evaluation, delivered one-on-one by non-specialist health workers in family homes, this time in rural Kolhapur, India. Children with a diagnosis of autism aged 2–9 years were randomly assigned to the PASS PLUS intervention (n = 21) or usual care condition (n = 19) with stratification for age ($<$5 or $>$5) and functional impairment. In practice, the Plus modules were delivered to 79% of the dyads (n = 15/19), with 4 families (21%) stating no expressed needs for further support. The most commonly delivered modules in decreasing order were for behavioral challenges (n = 12), sensory difficulties (n = 7), toileting support (n = 5), parental well-being (n = 4), sleep problems (n = 3) and feeding difficulties (n = 2). The study demonstrated high feasibility and intervention sessions met high therapist fidelity thresholds, with 92% of the core PASS procedures meeting fidelity thresholds for mandatory manualized content, and 96% meeting thresholds for quality. This was lower for the supplementary Plus modules, with fidelity thresholds to these manual components being met in 75% of sessions, and 81% meeting quality thresholds. The intervention

TABLE 3 | Key characteristics of studies identified by the scoping review.

	Rahman et al. (22)	**Divan et al.** (29)
Study characteristics		
Location of research	South Asia (Rawalpindi, Pakistan and Goa, India)	South Asia (Kolhapur, India)
Design	Single-blind effectiveness RCT	Single-blind effectiveness RCT
Name of intervention	PASS with treatment as usual	PASS-Plus with treatment as usual
Comparison	Treatment as usual (educational provision, some speech and language therapy)	Treatment as usual (educational provision, some speech and language therapy)
Follow-up period	8 months	9 months
Child diagnosis	Professional diagnosis of ASD	Professional diagnosis of ASD
Age of children	2–9 years	2–9 years
Number of participants	65	40
Intervention characteristics		
Supervision	Group and peer supported	Group and peer supported
Intervention Approach	Interventionist to Parent	Interventionist to Parent
Length of intervention	Fortnightly, 12 sessions over 6 months	Fortnightly, 12 sessions over 6 months
Treatment target	Parent-child synchrony and child initiations	Autism symptom severity, parent-child synchrony, shared attention and child initiations
Intervention Techniques	Video-feedback of parent-child play interactions and psychoeducation (see **Tables 4**, **5**)	Video-feedback of parent-child play interactions, psychoeducation, and separate section addressing commonly co-occurring conditions, with discussions, collaborative goal setting and supportive illustrated handouts to support low literacy (**Tables 4**, **5**).
Study results		
Feasibility, acceptability and fidelity	- Good feasibility for delivery within both Goa and Rawalpindi contexts and in children up to age 9, including those with high support needs - Good acceptability - Low attrition- 10% was allowed for; actual rate was 9% - High therapist fidelity to the intervention manual sustained throughout the trial	- High feasibility for delivering the intervention in a rural setting - Good acceptability - Attrition/attendance: 89% of participants received at least 3 sessions and 68% received the maximum 12 - High therapist fidelity to the core PASS intervention manual; lower fidelity for the Plus modules in terms of lay health workers tailoring to families' needs
Parent-child dyadic communication	Dyadic Communication Measure for Autism (DCMA) (24) Parental synchrony: Adjusted Mean Difference (AMD) 0.25; 95% CI 0.14 to 0.36; Effect Size (ES) 1.61 Proportion of child communication initiations with parent: AMD 0.15; 95% CI 0.04 to 0.26; ES 0.99 Child-parent mutual shared attention: AMD −0.16; 95% CI –0.26 to –0.05; ES −0.70	Dyadic Communication Measure for Autism (DCMA) (24) Parental synchrony: AMD 0.35; 95% CI 0.18 to 0.52; ES 3.97 Proportion of child communication initiations with parent: AMD 0.17; 95% CI 0.03 to 0.32; ES 1.02 Child-parent mutual shared attention: AMD 0.10; 95% CI −0.07 to 0.27; ES 0.5 Co-morbid symptoms: AMD −9.0; 95% CI −24.26 to 6.26; ES 0.32
Parental mental health	Patient Health Questionnaire-9 (28) AMD 0·95; 95% CI –1·38 to 3·27; ES 0.27	Patient Health Questionnaire-9 (28) AMD −4.55; 95% CI −8.52 to −0.58; ES 0.76
Child adaptive behavior	Vineland Adaptive Behavior Scale (VABS) (25) Adaptive Behavior Composite: AMD –0·93; 95% CI –3·53 to 1·68) ES –0.08	Vineland Adaptive Behavior Scale (VABS) (25) Adaptive Behavior Composite: AMD 0.67; 95% CI −3.80 to 5.15; ES 0.06
Child communication and language	McArthur Communicative Development Inventory (26) Receptive subscale: AMD 0·19; 95% CI –0·56 to 0·94; ES 0·07 Expressive subscale: AMD 0·03; 95% CI –0·46 to 0·52; ES 0·01 Communication and Symbolic Behavior Scales Developmental Profile (27) Total score: AMD –0·68; 95% CI –8·79 to 7·42; ES –0·02	N/A
Child behavior	N/A	Developmental Behavior Checklist (DBC) (30) Total score: AMD −9.00; 95% CI −24.26 to 6.26; ES 0.32
Child autism symptoms	N/A	Brief Observation of Social Communication Change (BOSCC) (31) AMD −2.42; 95% CI −7.75 to 2.92; ES 0.22

TABLE 4 | List of common components based on the @PracticeWise framework (20).

S.No	Element/ Technique	Source	Practice element label	Definition	Rahman et al. (22)	Divan et al. (29)
1.	Technique	©Practicewise	Accessibility Promotion	Any strategy used to make services convenient and accessible or to proactively enhance treatment participation. This might include: • "appointment reminders" (e.g., telephone, postal, text message or email confirmations and reminders • availability of on-site child care • flexible scheduling, e.g., after-hours scheduling, drop-in appointments • location, e.g., holding sessions at a local school, the family's home, or other convenient sit • transportation, e.g., to appointments, bus tokens, gas money	Home based delivery, appointment reminders telephone confirmation	As in PASS
2.	Element	©Practicewise	Activity Selection	The identification of specific positive activities in which the child can participate outside of therapy, with the goal of promoting or maintaining involvement in rewarding and enriching experiences.	Collaborative identification of activities for home practice	As in PASS
3.	Element	©Practicewise	Caregiver Coping	Exercises or strategies designed to enhance caregivers' ability to deal with stressful situations.	No	"Plus" component only: Supportive strategies for parental well-being including referrals for at risk parents
4.	Element	Additional	Communication Skills _Social Communication	Strategies to support the development of joint/shared attention and joint engagement	Video feedback technique to identify and encourage parent behaviors to support social communication during dyadic play	As in PASS
5.	Element	Additional	Communication Skills Language expansion	Strategies to support language expansion	Video feedback technique to identify and support opportunities for language expansion during dyadic play	As in PASS
6.	Element	Additional	Family Engagement	Strategies that are directed beyond the caregiver-child dyad to help wider family members to understand the intervention and support intervention strategies which are being generalized	Initial home visit where family members are supported to increase their understanding of the impairments in autism and the goals of the intervention	As in PASS
7.	Element	©Practicewise	Functional Analysis/Behavior Management	The study of antecedents and consequences impacting a behavior designed to yield a functional understanding of that behavior. This analysis is typically tested through controlled manipulation of antecedents and consequences to verify the formulation.	No	Plus component only: Supporting caregivers to consider the environment around challenging behaviors, and to address contributing factors.
8.	Technique	Additional	Goal Setting-Caregivers	The explicit selection of a therapeutic goal set for the parent for the purpose of working toward achieving that goal. This often involves repeated assessment of the successful approximation of the goal.	Home practice goals collaboratively set with caregiver based on discussions during the session.	As in PASS
9.	Technique	Additional	Handouts	The use of written or illustrated materials to support psycho-education and strategies that caregivers can use to promote specific changes in the child's behavior	No	Plus component only: Illustrated handouts designed to support understanding and addressing common co-occurring conditions

(Continued)

TABLE 4 | (Continued)

S.No	Element/ Technique	Source	Practice element label	Definition	Rahman et al. (22)	Divan et al. (29)
10.	Technique	Additional	Homework- Review	Review of homework set in the previous therapy session	Home practice goals reviewed at each session	As in PASS
11.	Technique	Additional	Record/diary	Caregiver or non-specialist maintaining records or a diary of child's behavior or strategies/home program practiced	Written record of key observations from the session along with goals for home practice maintained	As in PASS
12.	Element	©Practicewise	Play Therapy	The use of play as a primary strategy for therapeutic change. This may include the use of play as a strategy for clinical interpretation.	Dyadic play as the basis of the video feedback for social communication intervention	As in PASS
13.	Technique	©Practicewise	Praise- Caregivers	The training of caregivers, teachers, or others involved in the social ecology of the child in the administration of social rewards to promote desired behaviors. This can involve praise, encouragement, affection, or physical proximity.	Validation of strategies used during play and for engagement with home practice is a technique used as a social reward for the caregiver	As in PASS
14.	Element	©Practicewise	Problem Solving	Training in the use of techniques, discussions, or activities designed to bring about solutions to targeted problems, usually with the intention of imparting a skill for how to approach and solve future problems in a similar manner. Includes components such as brainstorming, choosing a solution, and evaluating the results.	Collaborative problem solving to support caregiver to engage with strategies during home practice	As in PASS. Plus component only: Additionally to support adopting strategies to manage co-occurring conditions
15.	Technique	©Practicewise	Psychoeducation - Caregiver	The formal review of information with the caregiver (Socratic or otherwise) about the development of the child's problem and its relation to a proposed intervention. This often involves an emphasis on the caregiver's role in either or both. This can include multiple media (e.g., a video about mental health problem).	Initial engagement with the family and caregivers aims at gaining an insight of the families' understanding of autism and their aspirations of the intervention process.	As in PASS Plus component only: Additionally understanding the family's interpretation and reasons for co-occurring conditions
16.	Technique	Additional	Relationship/Rapport Building-Caregiver	Strategies in which the primary aim is to increase the quality of the relationship between the therapist and caregiver. Can include active listening and empathy	Reflective video-feedback and collaborative goal setting aims at building a positive therapeutic alliance	As in PASS
17.	Element	©Practicewise	Relaxation	Techniques or exercises designed to induce physiological calming, including muscle relaxation, breathing exercises, imagery, meditation, and similar activities.	No	Plus component only: Supportive relaxation techniques as a first step to help support caregiver well-being
18.	Technique	©Practicewise	Tangible Rewards	The training of parents, teachers, or others involved in the social ecology of the child in the contingent administration of tangible rewards to promote desired behaviors. This can involve tokens, charts, or record keeping, in addition to direct (i.e., first order) reinforcers.	No	Plus component only: Supporting caregivers to consider tangible rewards which can motivate their child while they use techniques to address co-occurring conditions
19.	Technique	Additional	Video feedback	Reviewing caregiver-child interaction videos and providing feedback to the caregiver on the child's behavior, the impact of the caregivers actions on the child's behavior and suggestions on alternative ways of interacting.	Reflective video-feedback and collaborative goal setting is the core methodology of this intervention	As in PASS.

was reported to be acceptable, and with regards to attendance, 89% of participants in the trial arm received at least the minimum 3 sessions and 68% received the maximum 12. For outcomes, the results showed that there was no evidence that additional delivery of these rather different strategies had a negative effect on the effectiveness of the core PASS video-aided communication intervention; the positive changes in parent synchrony and child initiations in the dyad were replicated as in PASS (22). In addition, PASS PLUS showed some evidence of a trend toward improvement in parental mental wellbeing (as measured by the PHQ-9), (28) but did not lead to significant changes, relative to the control group, in child outcomes such as co-occurring symptom levels (as measured by the Developmental Behavior Checklist, DBC) (30) or autism symptom severity as measured by the Brief Observation of Social Communication Change (BOSCC) (31). A large scale-up effectiveness and cost-effectiveness evaluation of this PASS Plus intervention is currently underway using delivery by frontline workers embedded in the established health system in urban India (32). The Communication-centered Parent mediated treatment for Autism Spectrum Disorders in South Asia ("COMPASS") trial in this way represents the first large scale effectiveness test of an intervention for autism in a LMIC.

Common Components

Reviewing the described interventions in the publications along with the knowledge of the intervention, common components across the identified interventions are represented in **Tables 4**, **5**.

DISCUSSION

This scoping review set out to identify interventions for young autistic children that have been delivered by non-specialists in low- and middle-income countries (LMIC) with evidence of feasibility, acceptability, and effectiveness. It was hoped that the review would elucidate the intervention elements that have evidence of successful delivery in LMIC by non-specialist providers and offer indications for the future direction in the design, mechanistic evaluation, and delivery of scalable interventions for low resource settings. This review identified only two studies that have evaluated the effectiveness of interventions for young autistic children and their families delivered by non-specialists in LMIC. Both these studies were conducted by teams that included senior authors of this review and both evaluated interventions that were parent-mediated and primarily communication-focused, where the non-specialist had no direct contact with the child beneficiary. There is, therefore, a striking lack of diverse empirical evidence to guide scalable intervention design and delivery within the countries of the world in which most of the world's autistic children live.

Further clinical research in LMIC is urgently needed to provide evidence that is directly applicable to the key needs of LMIC health systems, to inform task-sharing and non-specialist delivery of autism interventions. This will help reduce inequity and achieve universal coverage within health systems which have an acute paucity of human and economic resources.

This review adopted a narrow definition of non-specialist providers to accurately reflect the most abundant workforce resource available to task-sharing approaches within LMIC: frontline community health workers, community volunteers or parent/disability champions. This focus allows the results to be more directly applicable to these settings. Other reviews (33) have investigated "non-specialist" delivery but adopted a wider definition of practitioners who are not relevant to the many LMIC settings that we are discussing. Frontline community health workers provide advice and support around issues such as maternity care, nutrition, immunization, child growth and development. They are often educated through high school but are not necessarily graduates; they have no formal healthcare or child development training but receive focused training around specific issues. They often live within the communities they serve and therefore likely to be trusted by the community. These frontline workers are an invaluable and flexible resource that can be trained, supervised, and incentivized to deliver interventions to children with neurodevelopmental disabilities and their families.

In this review we used two pre-established frameworks to classify intervention components, within the two interventions identified. We showcase that non-specialists – supported by a carefully established and implemented supervisory framework - can deliver a range of intervention elements and techniques to parents of young autistic children, with preliminary evidence of feasibility, acceptability, and effectiveness. Six elements from the @Practisewise framework (20) and four elements from the NDBI framework (17) along with three additional elements, were shown to be successfully delivered in this way. These included supporting parents to understand the communication development of their autistic child and to use strategies (e.g., language expansion, communication temptations) to support communication and language development. Similarly, we identified four techniques from the @Practisewise framework (20) and four from the NDBI framework (17) along with five additional techniques, including collaborative goal setting, parental psychoeducation, and the use of positive social or tangible rewards. It is important to note that we have not identified any tests of whether non-specialists could deliver intervention components directly with the autistic child or whether they could deliver interventions targeting other developmental domains or more complex co-occurring conditions, such as anxiety. These are important foci for future work. Another important clarification is that we are not proposing that the elements and techniques that we have identified are *active ingredients.* This would require in-depth mechanistic studies of each element and their interactions with each other and with outcomes (34, 35). A priority for future research should be to identify such active ingredients but also to evaluate the extent to which those ingredients can be delivered by non-specialists. Our conclusions on components must be tempered by the fact that they are based on a very small number of studies testing two interlinked interventions. However, this analysis can help clinicians and researchers to select from these evidence-based intervention components which have proved to be feasible for non-specialist delivery to inform

TABLE 5 | List of common components based on the NDBI-Fi framework (11).

S. No.	Components	Brief Description	Rahman et al. (22)	Divan et al. (29)
1.	Face-to-Face (Technique)	• Child and adult facing each other • Child and adult on similar level	The parent is encouraged to take a position which allows them to observe their child's communication signals	As in PASS
2.	Follow child's lead (Element)	• Child and adult are both active participants in child-chosen activity	The parent is encouraged to allow the child to choose the toy they would like to play with	As in PASS
3.	Positive affect (Technique)	• Adult uses positive affect • Adult matches affect to child's sensory needs	The parent is encouraged to be positively involved in the child's play and validate the child's choices during the interaction	As in PASS
4.	Modeling language (Element)	• Adult adjusts language to the child's developmental level	The parent is encouraged to recognize their child's communication level and match it during play	As in PASS
5.	Responding to communication (Element)	• Adult verbally responds to child's communication behaviors by repeating, clarifying, or expanding	The parent is encouraged to recognize their child's communication and initially repeat their outputs. As the child's communication increases the parent is encouraged to expand the language contingently	As in PASS
6.	Communicative temptations (Elements)	• Adult creates situations to elicit communication from the child followed by a brief period of expectant waiting	As the child and parent build a to and fro play and communication routine, expectant pauses are introduced	As in PASS
7.	Frequency of direct teaching (Technique)	• Adult directs the child to demonstrate new or emerging skills	No	Plus component only: The parent is encouraged to adopt techniques that can help build the child's adaptive skills for independent functioning such as toileting, eating and sleeping.
8.	Quality of direct teaching (Technique)	• Adult uses high-quality teaching strategies (e.g., clear instructions, when a child is motivated, contingent reinforcement).	No	Plus component only: The parent promotes desired behaviors of the child through strategies such as positive reinforcement, clear instructions, and social stories.

scalable autism interventions implemented through the task-sharing approach.

An important question concerns the equivalence of task-sharing approaches in comparison to specialist-delivered approaches. The task-sharing approach has evidence of feasibility and acceptability within the context of adult mental health across diverse low resource settings (36–39) and some supportive evidence of equivalence. For example, a head-to-head comparison within a depression treatment study (40) found that a simpler psychological treatment delivered using the task-sharing approach had the same effect as a costlier intervention delivered by specialists. This suggests that well-designed training and supervision cascades led by highly qualified and expensive experts can support less qualified individuals to deliver complex interventions with similar effectiveness but to more people and at a lower cost. We do not have any directly comparable results on task-sharing versus specialist delivery for autism interventions and this is an important focus for future research if we are to ensure health equity across populations.

The Lancet commission on the Future of Care and Clinical Research for Autism (41) recognizes the prohibitive costs and lack of access to evidence-based interventions in most of the world. The authors recommend a stepped care personalized approach to care, (5, 42) and we would suggest that interventions containing the common components identified here could be evaluated as WHO tier 3 interventions for delivery in low resource settings, allowing more complex cases to be triaged up to specialist care. As part of this stepped care approach, non-specialist delivery could be used to widen access to support, potentially benefiting many additional children, including those with other neurodevelopmental disabilities (**Figure 2**).

That said, even these common components delivered in one setting may have challenges in others, which may include the professional and personal workload of the frontline worker. Additionally, there will need to be contextual adaptations based on community perceptions and needs along with structural systemic changes to support career progression of frontline workers who may be trained to deliver such complex interventions.

In addition to our two identified papers, there were studies which did not meet our inclusion criteria, but which may also inform decisions around non-specialist delivery. We have included a couple of informative examples here. Hamdani and colleagues (43) conducted a large (n = 540) single-blinded, cluster randomized controlled trial in rural Rawalpindi, Pakistan. The trial evaluated the "FaNs for Kids" intervention compared

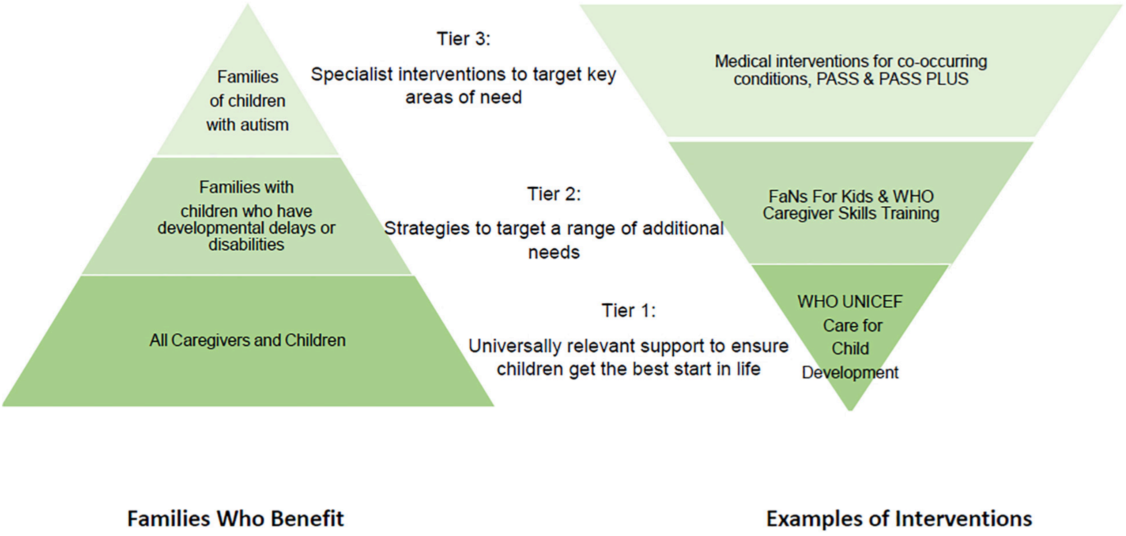


FIGURE 2 | Stepped care approach to supporting children based on the WHO Nurturing Care Framework.

to enhanced treatment-as-usual for parents of children aged 2–12 with a range of developmental delays and disabilities. The sample included children with communication disabilities but not formally diagnosed autism and, for this reason, did not meet the inclusion criteria of our search. FaNs for Kids was a group-based, psycho-education intervention adapted from the WHO Mental Health Gap Intervention Guidelines (mhGap-IG). The programme targeted child development and functioning (communication, socioemotional development, co-occurring conditions, and motor difficulties), as well as caregiver psychological well-being, and was delivered by "family champions," volunteers with family experience of disability, under the supervision of master trainers. The intervention was technologically assisted: caregivers accessed training videos on a tablet device and then met in nine group-based sessions to discuss different scenarios and build management plans around their child's needs. The study found that FaNs for Kids resulted in improved health-related quality of life in caregivers, but there were no significant improvements on child outcomes, relative to the control group.

Through this study, Hamdani and colleagues demonstrated that technology can be an effective medium to deliver psychosocial interventional components to parents within low resourced settings. This highlights the potential held by technology-assisted interventions to scale-up intervention delivery in such settings which because of the standardized messaging can help maintain program fidelity as well as ensure intervention dosage by non-specialists. In PASS (22) and PASS Plus (29) non-specialists also make use of digital technology, through the use of video feedback techniques. Within an ongoing trial of the PASS Plus intervention delivered by frontline health workers in Delhi, India, digital technology is being harnessed further to overcome the challenges presented by the COVID-19 pandemic and enable ongoing training and supervision of the workforce and intervention delivery to families using video-conferencing and telephonic technology (44). Lakhera and colleagues evaluated the feasibility of this digitally assisted delivery of PASS Plus and the acceptability of virtually adapted sessions to 50 families of children with autism, 34 families who transitioned from receiving home based delivery of PASS Plus, and 16 families who received virtual delivery only. Though there were several barriers to virtual delivery of sessions, including a family preference for home-based delivery, the lack of access to the internet and lack of digital literacy, most families and non-specialist providers adapted successfully to the remote delivery of the intervention. Such innovations of technology – if integrated within the existing health care system – may help to leap-frog barriers to scaling up service delivery.

A second programme of research that did not meet our inclusion criteria but that may also inform decisions around non-specialist delivery is the World Health Organization Caregiver Skills Training programme (CST) (45). The CST is a trans-diagnostic, family centered intervention designed to be delivered by non-specialists and to fit within Tier 2 of the World Health Organisation (WHO) stepped-care model (i.e., targeted support for children with identified developmental needs; see **Figure 2**) and thereby bridge the treatment gap for children in low resource settings (46). The CST combines group sessions for caregivers with home visits to tailor the intervention to each family and set individual goals for each child (47). The intervention is informed by a range of approaches including psycho-education, social communication interventions, functional analysis, positive parenting, and self-care methods with a strong foundation in the WHO-UNICEF Nurturing Care Framework (48). At the point of this scoping review, there were no published systematic evaluations of effectiveness using non-specialist providers. However, feasibility and acceptability studies have been conducted (49, 50), and we have been informed of evaluations of non-specialist delivery under review. The intervention has been pre-piloted through specialist workers in Italy and is now being field tested, including with non-specialist providers, in more than 30 countries around the world (50).

The strengths of this publication are that we have conducted a rigorous scoping review and have used two frameworks to classify intervention components allowing us to identify key techniques and strategies that non-specialists have delivered with evidence of effectiveness in three settings of urban and rural South Asia to families of young children with autism. A further strength concerns the explicit definition of non-specialist adopted in this review. However, this process has highlighted the acute lack of empirical data that is directly applicable to the needs of LMIC health systems, and is an urgent indication for more relevant research to support families of children with autism in the most underserved populations of the world.

DATA AVAILABILITY STATEMENT

The original contributions presented in this study are included in the article/supplementary material, further inquiries can be directed to the corresponding author.

AUTHOR CONTRIBUTIONS

GD and KL conceived the idea for the research manuscript and defined the inclusion criteria for the search. LN, CG, AK, CB, and SL screened abstracts and full-text manuscript based on the search criteria on Rayyan software with support from GD and KL. LN, CG, CB, AK, KL, and GD drafted the first version of the manuscript and contributed to subsequent versions. All authors read and approved the final manuscript.

FUNDING

This work has been conducted with the support of a Medical Research Council, UK grant. Grant Reference MR/R006164/1. We acknowledge funding from the UK MRC/Wellcome/DfID and Joint Global Health Trials Award: MR/R006164/1.

ACKNOWLEDGMENTS

We would like to thank Supriya Bhavnani, Jonathan Green, and Stephanie Shire for their invaluable inputs into this study.

REFERENCES

1. Baio J, Wiggins L, Christensen DL, Maenner MJ, Daniels J, Warren Z, et al. Prevalence of autism spectrum disorder among children aged 8 years—autism and developmental disabilities monitoring network, 11 sites, United States, 2014. *MMWR Surveill Summ.* (2018) 67:1. doi: 10.15585/mmwr.ss6706a1
2. Elsabbagh M, Divan G, Koh YJ, Kim YS, Kauchali S, Marcín C, et al. Global prevalence of autism and other pervasive developmental disorders. *Autism Res.* (2012) 5:160–79. doi: 10.1002/aur.239
3. Perera H, Wijewardena K, Aluthwelage R. Screening of 18–24-month-old children for autism in a semi-urban community in Sri Lanka. *J Trop Pediatr.* (2009) 55:402–5. doi: 10.1093/tropej/fmp031
4. Arora NK, Nair MK, Gulati S, Deshmukh V, Mohapatra A, Mishra D, et al. Neurodevelopmental disorders in children aged 2–9 years: population-based burden estimates across five regions in India. *PLoS Med.* (2018) 15:e1002615. doi: 10.1371/journal.pmed.1002615
5. Divan G, Bhavnani S, Leadbitter K, Ellis C, Dasgupta J, Abubakar A, et al. Annual research review: achieving universal health coverage for young children with autism spectrum disorder in low-and middle-income countries: a review of reviews. *J Child Psychol Psychiatry.* (2021) 62:514–35. doi: 10.1111/jcpp.13404
6. World Health Organisation. *Mental Health Atlas 2020.* Geneva: World Health Organisation (2021).
7. United Nations. *Goal 3, The Sustainable Development Goals Report.* (2021). Available online at: https://sdgs.un.org/goals/goal3 (accessed February 28, 2022).
8. Raviola G, Naslund JA, Smith SL, Patel V. Innovative models in mental health delivery systems: task sharing care with non-specialist providers to close the mental health treatment gap. *Curr Psychiatry Rep.* (2019) 21:1–3. doi: 10.1007/s11920-019-1028-x
9. Singla DR, Kohrt BA, Murray LK, Anand A, Chorpita BF, Patel V. Psychological treatments for the world: lessons from low-and middle-income countries. *Annu Rev Clin Psychol.* (2017) 13:149–81. doi: 10.1146/annurev-clinpsy-032816-045217
10. Van Ginneken N, Tharyan P, Lewin S, Rao GN, Meera SM, Pian J, et al. Non-specialist health worker interventions for the care of mental, neurological and substance-abuse disorders in low-and middle-income countries. *Cochr Database Syst Rev.* (2013) 2013: CD009149.
11. Mane Abhay B, Khandekar Sanjay V. Strengthening primary health care through Asha workers: a novel approach in India. *Primary Health Care.* (2014) 4:2167–1079.
12. Shrestha R, Barbaro J, Dissanayake C. Changes in knowledge on the signs of autism in young children (11–30 months) among female community health volunteers in Nepal. *J Autism Dev Disord.* (2022) 52:219–39. doi: 10.1007/s10803-021-04944-7
13. Aseyo RE, Mumma J, Scott K, Nelima D, Davis E, Baker KK, et al. Realities and experiences of community health volunteers as agents for behaviour change: evidence from an informal urban settlement in Kisumu, Kenya. *Hum Resour Health.* (2018) 16:1–2. doi: 10.1186/s12960-018-0318-4
14. Naveed S, Waqas A, Amray AN, Memon RI, Javed N, Tahir MA, et al. Implementation and effectiveness of non-specialist mediated interventions for children with autism spectrum disorder: a systematic review and meta-analysis. *PLoS One.* (2019) 14:e0224362. doi: 10.1371/journal.pone.0224362
15. Nair MK, Russell PS, George B, Prasanna GL, Bhaskaran D, Leena ML, et al. CDC Kerala 9: effectiveness of low intensity home based early intervention for autism spectrum disorder in India. *Indian J Pediatr.* (2014) 81:115–9. doi: 10.1007/s12098-014-1474-8
16. Chorpita BF, Daleiden EL, Weisz JR. Identifying and selecting the common elements of evidence based interventions: a distillation and matching model. *Ment Health Servic Res.* (2005) 7:5–20. doi: 10.1007/s11020-005-1962-6
17. Frost KM, Brian J, Gengoux GW, Hardan A, Rieth SR, Stahmer A, et al. Identifying and measuring the common elements of naturalistic developmental behavioral interventions for autism spectrum disorder:

development of the NDBI-Fi. *Autism.* (2020) 24:2285–97. doi: 10.1177/1362361320944011

18. Levac D, Colquhoun H, O'Brien KK. Scoping studies: advancing the methodology. *Implement Sci.* (2010) 5:1–9.
19. Ouzzani M, Hammady H, Fedorowicz Z, Elmagarmid A. Rayyan—a web and mobile app for systematic reviews. *Syst Rev.* (2016) 5:1–0. doi: 10.1186/s13643-016-0384-4
20. Chorpita BF, Daleiden EL. Mapping evidence-based treatments for children and adolescents: application of the distillation and matching model to 615 treatments from 322 randomized trials. *J Consult Clin Psychol.* (2009) 77:566–79. doi: 10.1037/a0014565
21. PracticeWise. *Psychosocial and Combined Treatments Coding Manual*. Satellite Beach, FL: PracticeWise (2012).
22. Rahman A, Divan G, Hamdani SU, Vajaratkar V, Taylor C, Leadbitter K, et al. Effectiveness of the parent-mediated intervention for children with autism spectrum disorder in south Asia in India and Pakistan (PASS): a randomised controlled trial. *Lancet Psychiatry.* (2016) 3:128–36. doi: 10.1016/S2215-0366(15)00388-0
23. Divan G, Hamdani SU, Vajartkar V, Minhas A, Taylor C, Aldred C, et al. Adapting an evidence-based intervention for autism spectrum disorder for scaling up in resource-constrained settings: the development of the PASS intervention in South Asia. *Glob Health Action.* (2015) 8:27278. doi: 10.3402/gha.v8.27278
24. Aldred C, Green J, Adams C. A new social communication intervention for children with autism: pilot randomised controlled treatment study suggesting effectiveness. *J Child Psychol Psychiatry.* (2004) 45:1420–30. doi: 10.1111/j.1469-7610.2004.00848.x
25. Sparrow SS, Cicchetti DV, Balla DA. *Vineland Adaptive Behavior Scales*. 2nd ed. Livonia, MN: Pearson Assessment (2006).
26. Fenson L, Marchman VA, Thal DJ, Dale PS, Reznick JS. *MacArthur-Bates Communicative Development Inventories*. 2nd ed. Baltimore, MD: Paul H. Brookes (2007).
27. Wetherby AM, Prizant BM. *Communication and Symbolic Behavior Scales: Developmental Profile*. Baltimore, MD: Paul H. Brookes (2002).
28. Kroenke K, Spitzer RL, Williams JB. The PHQ-9: validity of a brief depression severity measure. *J Gen Intern Med.* (2001) 16:606–13. doi: 10.1046/j.1525-1497.2001.016009606.x
29. Divan G, Vajaratkar V, Cardozo P, Huzurbazar S, Verma M, Howarth E, et al. The feasibility and effectiveness of PASS plus, a lay health worker delivered comprehensive intervention for autism spectrum disorders: pilot RCT in a rural low and middle income country setting. *Autism Res.* (2019) 12:328–39. doi: 10.1002/aur.1978
30. Einfeld SL, Tonge BJ. *Manual for the Developmental Behaviour Checklist (Primary Carer Version)*. Melbourne, VIC: Monash University Centre for Developmental Psychiatry (1994).
31. Grzadzinski R, Carr T, Colombi C, McGuire K, Dufek S, Pickles A, et al. Measuring changes in social communication behaviors: preliminary development of the brief observation of social communication change (BOSCC). *J Autism Dev Disord.* (2016) 46:2464–79. doi: 10.1007/s10803-016-2782-9
32. Sangath. *Communication-Centred Parent-Mediated Treatment for Autism Spectrum Disorder in South Asia (COMPASS)*. Delhi: Sangath (2018).
33. Reichow B, Servili C, Yasamy MT, Barbui C, Saxena S. Non-specialist psychosocial interventions for children and adolescents with intellectual disability or lower-functioning autism spectrum disorders: a systematic review. *PLoS Med.* (2013) 10:e1001572. doi: 10.1371/journal.pmed.1001572
34. Green J, Garg S. Annual research review: the state of autism intervention science: progress, target psychological and biological mechanisms and future prospects. *J Child Psychol Psychiatry.* (2018) 59:424–43. doi: 10.1111/jcpp.12892
35. Gulsrud AC, Hellemann G, Shire S, Kasari C. Isolating active ingredients in a parent-mediated social communication intervention for toddlers with autism spectrum disorder. *J Child Psychol Psychiatry.* (2016) 57:606–13. doi: 10.1111/jcpp.12481
36. Ali BS, Rahbar MH, Naeem S, Gul A, Mubeen S, Iqbal A. The effectiveness of counseling on anxiety and depression by minimally trained counselors: a randomized controlled trial. *Am J Psychother.* (2003) 57:324–36. doi: 10.1176/appi.psychotherapy.2003.57.3.324
37. Chatterjee S, Chowdhary N, Pednekar S, Cohen A, Andrew G, Andrew G, et al. Integrating evidence-based treatments for common mental disorders in routine primary care: feasibility and acceptability of the MANAS intervention in Goa, India. *World Psychiatry.* (2008) 7:39–46. doi: 10.1002/j.2051-5545.2008.tb00151.x
38. Chibanda D, Mesu P, Kajawu L, Cowan F, Araya R, Abas MA. Problem-solving therapy for depression and common mental disorders in Zimbabwe: piloting a task-shifting primary mental health care intervention in a population with a high prevalence of people living with HIV. *BMC Public Health.* (2011) 11:828. doi: 10.1186/1471-2458-11-828
39. Rahman A, Malik A, Sikander S, Roberts C, Creed F. Cognitive behaviour therapy-based intervention by community health workers for mothers with depression and their infants in rural Pakistan: a cluster-randomised controlled trial. *Lancet.* (2008) 372:902–9. doi: 10.1016/S0140-6736(08)61400-2
40. Richards DA, Ekers D, McMillan D, Taylor RS, Byford S, Warren FC, et al. Cost and outcome of behavioural activation versus cognitive behavioural therapy for depression (COBRA): a randomised, controlled, non-inferiority trial. *Lancet.* (2016) 388:871–80. doi: 10.1016/S0140-6736(16)31140-0
41. Lord C, Charman T, Havdahl A, Carbone P, Anagnostou E, Boyd B, et al. The lancet commission on the future of care and clinical research in autism. *Lancet.* (2022) 399:271–334. doi: 10.1016/S0140-6736(21)01541-5
42. Green J, Leadbitter K, Ainsworth J, Bucci S. An integrated early care pathway for autism. *Lancet Child Adolesc Health.* (2022) 6:335–44. doi: 10.1016/S2352-4642(22)00037-2
43. Hamdani SU, Suleman N, Akhtar P, Nazir H, Masood A, Tariq M, et al. Effectiveness of a technology-assisted, family volunteers delivered, brief, multicomponent parents' skills training intervention for children with developmental disorders in rural Pakistan: a cluster randomized controlled trial. *Int J Ment Health Syst.* (2021) 15:1–7. doi: 10.1186/s13033-021-00476-w
44. Lakhera S, Roy SG, Laithani N, Sangwan P, Azar Z, Menon S, et al. Adaptation of a parent mediated communication intervention for autism for virtual delivery, in India. In: *Proceedings of the International Society for Autism Research*. (2021).
45. Salomone E, Pacione L, Shire S, Brown FL, Reichow B, Servili C. Development of the WHO caregiver skills training program for developmental disorders or delays. *Front Psychiatry.* (2019) 10:769. doi: 10.3389/fpsyt.2019.00769
46. World Health Organisation. *WHO Caregivers Skills Training for Families of Children With Developmental Delays and Disorders*. (2022). Available online at: https://www.who.int/teams/mental-health-and-substance-use/treatment-care/who-caregivers-skills-training-for-families-of-children-with-developmental-delays-and-disorders (accessed March 3, 2022).
47. Tekola B, Girma F, Kinfe M, Abdurahman R, Tesfaye M, Yenus Z, et al. Adapting and pre-testing the World Health Organization's caregiver skills training programme for autism and other developmental disorders in a very low-resource setting: findings from Ethiopia. *Autism.* (2020) 24:51–63. doi: 10.1177/1362361319848532
48. World Health Organization, United Nations Children's Fund, World Bank Group. *Nurturing Care for Early Childhood Development: a Framework for Helping Children Survive and Thrive to Transform Health and Human Potential*. Geneva: World Health Organisation (2018).
49. Sengupta K, Shah H, Ghosh S, Sanghvi D, Mahadik S, Dani A, et al. World Health Organisation-caregiver skills training (WHO-CST) program: feasibility of delivery by non-specialist providers in real-world urban settings in India. *J Autism Dev Disord.* (2021) 51:1–8. doi: 10.1007/s10803-021-05367-0
50. Salomone E, Ferrante C, Salandin A, Ferrara F, Torchio E, Foletti G, et al. Acceptability and feasibility of the World Health Organization's

caregiver skills training implemented in the Italian national health system. *Autism*. (2021) 25:13623613211035228. doi: 10.1177/13623613211035228

Conflict of Interest: LN, AK, SL, and GD were employed by the not-for-profit Sangath. GD has led on the development and evaluation of the PASS and PASS Plus interventions and has received funding from Autism Speaks US and Grand Challenges Canada for this work. KL was also supported by an Autism Speaks grant for work on these interventions.

The remaining authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers | Frontiers in Psychiatry

TYPE Original Research
PUBLISHED 19 July 2022
DOI 10.3389/fpsyt.2022.907093

“We are doing the best we can to bridge the gap” - service provider perspectives of educational services for autism spectrum disorder in South Africa

Sarosha Pillay[1,2]*, Madeleine Duncan[1] and Petrus J. de Vries[2]

[1]Department of Health and Rehabilitation Sciences, University of Cape Town, Cape Town, South Africa, [2]Centre for Autism Research in Africa, Division of Child & Adolescent Psychiatry, University of Cape Town, Cape Town, South Africa

Background: The South African education system is increasingly unable to meet the growing needs of children with autism spectrum disorder (ASD). Recent studies in the Western Cape, one of the better resourced provinces in South Africa, showed that the pathway to care for children with ASD was an inconsistent and lengthy process, and that many children with ASD waited for extended periods to get access to an appropriate school placement. It is therefore clear that scalable and sustainable solutions are required to improve access to appropriate education for children with ASD.

Methods: Here we performed a qualitative study using thematic analysis of ten multi-sectorial ASD service provider interviews in the Western Cape Province to examine provider perspectives and proposed solutions to meet the educational needs of children with ASD.

Results: Provider perspectives were grouped in three categories: “bridging the gap across the spectrum and lifespan”, “gaps to bridge”, and “building bridges”. The first category captured provider perspectives of the service-related needs inherent to a diagnosis of ASD. The second category summarized service provider views of the challenges associated with providing services to children with ASD and the third category captured provider perspectives on potential actions to improve ASD education services delivery in the province. The overarching theme that emerged was “We’re doing the best we can to bridge the gap”.

Conclusion: Participants provided ten key recommendations for service strengthening that may lead to contextually relevant innovations to meet the educational needs of children with ASD in the province. Findings from this study has direct relevance to other South African provinces and may have relevance to improve pathways and reduce service delivery gaps also in other low-and-middle-income countries.

KEYWORDS

autism spectrum disorder, service provider perspectives, education systems, low-and-middle-income countries, service delivery

OPEN ACCESS

EDITED BY
Diana Schendel,
Drexel University, United States

REVIEWED BY
Hirokazu Kumazaki,
Kanazawa University, Japan
David Griessel,
University of the Free State,
South Africa

*CORRESPONDENCE
Sarosha Pillay
sarosha.pillay@uct.ac.za

SPECIALTY SECTION
This article was submitted to Autism, a section of the journal Frontiers in Psychiatry

RECEIVED 29 March 2022
ACCEPTED 24 June 2022
PUBLISHED 19 July 2022

CITATION
Pillay S, Duncan M and de Vries PJ (2022) “We are doing the best we can to bridge the gap” - service provider perspectives of educational services for autism spectrum disorder in South Africa. *Front. Psychiatry* 13:907093. doi: 10.3389/fpsyt.2022.907093

Introduction

Autism Spectrum Disorder (ASD) is a complex developmental disorder that affects 1–2% of the world's population at varying degrees and is characterized by a range of impairments in the areas of social communication, learning and behavior (1). The quality of life of many individuals with ASD and their families are significantly impacted by these impairments (2–4) and individuals with ASD may require services across the lifespan to minimize and manage some of the core features of ASD and co-occurring conditions (5, 6).

High-income countries (HIC) are typically better resourced to meet the needs of individuals with ASD and their families. However, even in some HIC there have been reports on challenges in service delivery, most notably in the areas of access to early diagnostic evaluations (7), policy implications for early intervention and support for school-aged children with ASD (8), and inclusion and employment of individuals with ASD (9).

Olusanya et al. (10, 11) reported that 95% of all children with developmental disabilities including ASD live in low-or middle-income countries (LMIC) (10, 11), yet there is little information on how these children are identified, evaluated, treated and educated (12–15). In a scoping review of all autism research in sub-Saharan Africa, Franz and colleagues identified that less than 1% of the world's autism research had taken place in Africa, and that no studies had examined health or education systems for children with autism (15).

South Africa has a population of 58.8 million people of diverse cultural and socio-economic backgrounds (16). It is an upper-middle income country with the highest Gini co-efficient indicating vast socio-economic disparities between rich and poor (17). High Gini co-efficient is characteristic of many LMICs, including India and most Latin-American countries (18). South Africa therefore has a socio-economic profile that is very representative of the needs of the majority of the world's population.

Vast disparities across social class and racial lines exist in access to public health and education services due, amongst others, to the socio-political legacy of apartheid (19–21). In South Africa, children with disabilities including those diagnosed with ASD are most at risk of not having their health, social and educational needs met due to reliance on state-funded services used by the majority of the population (22). The South African special education sector is a product of the apartheid era where children were historically classified according to race and disability (22) and children with specific disabilities could only be enrolled at the few available schools allocated to the disability. Although there have been efforts to correct these legacies of apartheid, in 2016 there were 119 403 children with disabilities attending 455 schools across the country (23) and an estimated 597,953 children with disabilities reported to be out of schools (24). The limited state-funded services for children with special education needs in South Africa therefore tend to be oversubscribed resulting in long periods of waiting for access.

In a study of the educational system for ASD in the Western Cape Province of South Africa, Pillay and colleagues (25) set out to identify all school-aged children with a diagnosis of ASD in the Western Cape Education Department database. Out of more than 1 million children, only 940 children with a diagnosis of ASD were identified, representing a rate of 0.08%. Based on a conservative ASD prevalence at 1%, the finding suggested a more than 10-fold under-identification of ASD in schools in the Western Cape province (25). Apart from the low numbers, the authors also identified very low rates of co-occurring diagnoses in the sample, complex and confusing pathways to diagnosis and treatment, and, surprisingly, found that 89% of children with an ASD diagnosis were in special educational placements (25). The authors next proceeded to search for those children waiting for a school placement in the province, and identified 744 children, with 266 (36%) of them being of legal school-going age, but not in education (26). Fifty two percent (52%) of children had been waiting for schools for more than a year (26). To compound the emerging picture of ASD in the province, the authors found a 76% increase in children with ASD in school between 2012 and 2016 (25), and a 276% increase in children on the "waiting list" for the same period (26). The findings from these earlier studies made it clear that, even in one of the better-resourced provinces of South Africa, the educational system was not able to meet the needs of children with ASD and their families.

In health systems research, Gilson and colleagues (27) pointed to the importance of understanding any given system in a "whole-system" way, including the "hardware" elements (e.g., human resources, infrastructure, financing), the 'software" elements (e.g., ideas and interests, relationships and power, values and norms), and the interaction between these hardware and software elements. Applying the whole-system concept to the educational system in the Western Cape province of South Africa, and in the context of previous work by Pillay and colleagues on hardware elements, we recognized the importance of exploring also the software elements in the education system in order to generate strategies that may support strengthening of the system. Pillay and colleagues (25, 26) examination of the hardware elements of the education system reported the rates, demographic, disability and educational profile of children with ASD both in schools and those waiting for schools in the province. These data provided a starting point for understanding the landscape of ASD education in the Western Cape and the authors proposed that engagement with stakeholders would be necessary next steps for developing a more comprehensive picture of the ASD situation in the province.

In an effort to complement the "hardware" findings of Pillay and colleagues, we therefore set out to examine the 'social" elements of the system by seeking the perspectives of service providers, a key stakeholder group, in the Western Cape province. Our overarching aim was to describe their views of existing services for children with ASD in the Western Cape, and their recommendations for improvements to existing service systems.

Methods

Design

A qualitative pragmatic research methodology (28) was used to collect descriptive data arising from the realities faced by ASD service providers.

Participants and procedures

Purposive and snowball sampling was used to identify and recruit a broad range of service providers from the government, private and voluntary sectors. To be eligible, potential participants had to have first-hand experience of delivering ASD educational and other services, and knowledge of the waiting list for these services in the Western Cape province. Participants were invited by email or telephonically to participate. Written informed consent was obtained from all participants.

Data collection

Individual semi-structured interviews of approximately 45–60 min were conducted by the first author. Interviews were conducted in English and were digitally recorded. The interview guide included broad, open-ended questions around service provider perspectives of existing services in the province and their proposed solutions to improve ASD service delivery. Clarification probes were used to ensure the following research questions were being answered: "what do service providers think about the current unmet education needs of children with ASD in the Western Cape province and what recommendations do they have for addressing these needs?" The interview was terminated when the interviewer and interviewee felt that data saturation was reached.

Data analysis

Audio-recorded interviews yielded 7 h and 30 min of data that were transcribed verbatim by the first author into textual form for thematic analysis (29). NVivo version 12 was used for data storage, management and first level inductive coding to identify units of meaning expressed by the service providers. Second level coding of inductively identified codes was done manually and involved grouping codes into sub-categories and categories of meaning from which an overarching theme emerged. Thematic synthesis described by Thomas and Harden (30) was used where the primary researcher identified the sub-categories, categories, and theme and consensus was reached through discussions with the second author, an experienced qualitative researcher. All proposed subcategories, categories and themes were then presented to the third author for discussion until consensus was reached.

Scientific rigor

The creditability and transferability of the data (31) were enhanced by the primary researcher's prolonged engagement and participant observation in the field that enabled the pertinent use of probes during the data gathering interviews. Participants verified the accuracy of the information reflected in the findings through a rigorous process of member checking (32). Rigor was also enhanced through data triangulation from different sources namely interviews, researcher field notes and document reviews.

Ethical approval

Ethical approval was granted by the University of Cape Town Human Research Ethics Committee (HREC reference: 072/2016) as well as the Western Cape Department of Education (reference: 20150727-1712).

Community participation

No individuals with ASD or their family members were directly involved in this study. Ten service providers who work with individuals with ASD and their families were involved.

Results

Demographic characteristics of participants

The characteristics of included participants are shown in Table 1. A total of ten participants across different professional groups, sectors, departments and base for work (urban/rural) were included.

Thematic analysis

Table 2 provides a summary of the main theme, categories and sub-categories identified in analysis. The overarching theme that emerged in answering the research questions was "*we are doing the best we can to bridge the gap*". The theme reflected service providers' awareness of the discrepancies that exist between the scope and nature of the ASD educational service demands and their capacity (either individually or collectively) to meet these demands. Providers expressed perspectives about the scope and nature of ASD service needs in three discreet capacity-related categories: (1) *bridging the gap across the spectrum and lifespan*, (2) *gaps to bridge,* and (3) *building bridges*. The first category captured provider perspectives of service-related factors inherent in the ASD diagnosis and intervention. That is, providers acknowledged the wide-ranging, lifelong and changing needs of children with ASD and their families. The second category summarized service provider views of the range of structural constraints that limited their capacity to meet service needs. The third category captured provider perspectives on potential actions to "build bridges" that might reduce the demand-capacity divide.

The theme, categories and sub-categories as presented in Table 2 will next be discussed with representative quotes.

TABLE 1 Participant characteristics.

Participant number	Service provider	Sector	Urban/Rural
P1	Medical doctor	Health	Urban
P2	Special school principal	Education	Urban
P3	Special school deputy principal	Education	Urban
P4	Occupational therapist	Education	Rural
P5	Educational psychologist	Education	Urban/Rural
P6	Educator	NGO	Urban/Rural
P7	Parent advocate	NGO	Urban
P8	ASD educational consultant	Private	Urban/Rural
P9	Medical doctor	Health	Urban
P10	Psychologist	Education	Urban/Rural

NGO, non-government organization.

Category 1: Bridging the gap across the spectrum and lifespan

Service providers spoke about the need for ASD services to be available to individuals with all levels of severity of ASD throughout the different stages of their life. Four sub-categories emerged: lifespan factors, disability factors, curriculum factors, and policy factors.

Lifespan factors

Given the lifespan implications of the diagnosis, service providers felt that the right type of service was essential to support the individual with ASD across the different stages of life and that these services were generally lacking:

> *"That's what we need, cradle to grave provision…And I do think it would be great if the Western Cape could do that…if it was possible" (P8)*

TABLE 2 Summary of main theme, categories and sub-categories.

Theme	Categories	Sub-categories	Description of codes that constituted the sub-category
We're doing the best we can to bridge the gap	Bridging the gap across the spectrum and lifespan	Lifespan factors	The lifespan needs of persons with ASD
		Disability factors	How the diagnostic label of ASD can be a barrier to services
		Curriculum factors	The fit between the learning potential and educational needs of each child with ASD and what they are being taught
		Policy factors	Participant views on the regulatory, policy and political factors influencing ASD services in the province
	The gaps to bridge	Resource constraints	The challenges in human and infrastructural resources
		Contextual constraints	Issues of litigation arising from unmet service needs, equity of access and the burden of public health care
		Competence constraints	The perceived competencies and skill-set of decision-makers, policy-makers and planners
	Building bridges	Leadership	The importance of leadership to address the challenges in ASD education
		Capacity building	Recommended actions that would strengthen the range and quality of resources
		Co-ordination	System efficiency through managed interagency collaboration
		Innovation	Novel actions for improved service delivery

Disability factors

This sub-category contained perspectives about the diagnosis of autism acting as an inherent barrier to service access and inclusion:

> *"If I didn't know you were autistic, I might still provide services for you, but now, oh but you come with that label, oh you're ASD, sorry then you fall into that stream... You see, and for me, that is then a disservice to the child" (P9)*
>
> *"The question was asked to me by somebody yesterday, about, why is autism so fearful? Everybody cringes when they hear you have to take learners with autism, why can't they just go to other schools?" (P2)*

Service providers stressed the right to participation in education for all children with ASD regardless of the severity of disability. Some expressed concern that ASD children presenting with high levels of needs were denied access to educational services because of the amount of individualized functional support that they require. These children are referred to "special care centers" (centers for children with severe-to-profound intellectual disability and associated ASD) where the education-related intervention that they receive is not always optimal:

> *"... but the special care centers can't necessarily cater for our children with autism, who are very busy, and they don't present the same as the other children at the special care centers. The disability is just so different. So what happens to those children who can't be supported in an autism school, and needs even higher level of support?" (P5)*

Curriculum factors

The quality of education for children with ASD was a big concern with some service providers feeling that the curriculum did not prepare young adults with ASD for vocation after school:

> *"... where we are focusing on skills, we're focusing on really mundane skills... And I think situations where our teenagers and our young adults being taught, for example, to put windscreen wipers into boxes... that's not a career" (P7)*

The need for differentiation of the mainstream curriculum to accommodate and optimally support the learning potential of all children with ASD according to their developmental needs was stressed:

> *"I think that there might be a lot of children who are in autism schools, the traditional autism schools, who could potentially find a home in mainstream schools, if there was the willingness to say "you know what? This child is not going to be attending English, it's going to go to Maths, and then he's going to sit in his or her own little space for the next hour". I don't know, I mean obviously there are massive kind of logistical requirements, and you've got a schooling system already under strain..." (P10)*

Policy and political factors

This sub-category included perspectives on the moral and legislative right to education for all children irrespective of ability:

> *"sorry, I take the extreme view, every child has a right to education in the constitution, and they should all be in school, and I know they're not... Everybody else can fight for their piece of cake, but we need to say, if we're not looking after the most needy citizens, then what are we as a society?" (P8)*

Concerns were raised that ignorance about the diagnosis and its service requirements rendered policy-makers ineffective. Service providers felt that government stakeholders and policy-makers did not have a good understanding of the ASD situation in the province and that people who did know (such as service providers, individuals with ASD and their families) were not included in policy decision-making processes, leading to "knee-jerk" short-term rather than strategic long-term actions:

> *"I think they just don't know. Honestly, my opinion is that the policy makers and even some of the people who are writing the adapted curriculums have never set foot in a classroom. Some of the people who are even writing the SIAS* document, for example, which I think is a brilliant document, have never stepped into a classroom. And you can't... you can't do this... you can't make decisions for the people on the ground if you've never been on the ground" (P7)*
>
> *Policy on Screening, Identification, Assessment and Support (SIAS) provides a framework for standardized procedures to facilitate the inclusion of children who require additional support in schools.
>
> *"... they* [policy makers] are *reactionary. I understand why, I understand the pressures that they experience and they have to endure, but they have knee-jerk reactions all the time" (P3)*

Perspectives on policy compliance and the political agenda behind policy development and implementation were raised. Service providers felt that the lack of transparency around the ASD waiting list had unfortunate consequences for the children with ASD:

> *"I call it a political game as well… we can't get away from it… I'm not saying the one is better than the other* [political party], *but it's politics. And unfortunately, the children suffer. And once again it comes back to my point of, I question, is it in the best interest of the children?" (P3)*
>
> *"I suspect that it had become something that would potentially be politically very, very uncomfortable, were it to arrive in the public domain; that in reality, that we were able somehow to generate a list of children who weren't in schools for autism, versus available proper spaces, it would look like a disaster." (P10)*

Category 2: Gaps to bridge

The second category described resource, contextual and competence constraints that were creating service gaps that needed to be bridged in order for ASD services in the Western Cape to meet current and future needs.

Resource constraints

Resource constraints referred to challenges in human and infrastructural resources, and the impact thereof on a) service provider wellbeing, and b) the ability of the education system to provide critical early intervention services.

With the growing demand for ASD educational services, service providers felt the current infrastructure was not keeping up with the need for customized physical space:

> *"I don't see any significant moves, plans, to provide that infrastructure. And when I talk infrastructure, I mean the hard buildings, people, you know, not just a programme, but the physical facilities to provide in this growing need, that is just getting more and more. You know, we're already battling with a backlog, we're sitting with this backlog… But it's not just trying to catch up, we have to provide for the ever growing number" (P9)*

Human resources including people with the necessary skills and willingness to work with children with ASD was seen as a major gap that needs to be bridged:

> *"I think what we're lacking is people. People qualified to work in it, and not just people qualified, people passionate about working in it. And I think this isn't a field to go into for a nine to five job. You go into this field, it's hard work, the kids do have their challenges, and as gorgeous as they are, they aren't without tough days, and what we're lacking is people who want to work with that. So, for me, that's the biggest resource we're missing, is people. People motivated to work in the field" (P7)*

In the absence of appropriate and adequate resources, it was felt that the increasing pressures put on existing ASD schools by the education department to place more children in a classroom to alleviate the waiting list would have negative consequences on the quality of education and on the mental health of the staff:

> *"I'm concerned about the quality of education that we're going to deliver from here on in. I'm very concerned about that because our staff members will be burnt out." (P3)*

Participants were of the opinion that resource constraints also contributed to the constant delays in establishing essential early intervention programmes. With the pressures that the education department faced in providing education for school-aged children with ASD, early intervention programmes were being neglected:

> *"I think [it] is absolutely heart-breaking, never mind heart-breaking, it's also a human right being denied, in my opinion, that's early intervention… it's not available, because they need to deal with the waiting list." (P8)*
>
> *"I think the communication support for children, knowing that there's some critical windows, in young lives. And I'm talking about the two to four / five year age group, that missing period of communication support." (P10)*

Contextual constraints

The context within which the growing ASD waiting list exists was highlighted. Service providers acknowledged that education for children with ASD was only one of the public health challenges that the education department was faced with:

> *"But I think people are working hard, and I think one shouldn't underestimate the amount of work that the Education Department must be looking at, and even Early Childhood Development. Because remember, we're just looking at autism. We have huge numbers… I have children with cerebral palsy and other special needs, who are also struggling to get services" (P1)*

Some service providers indicated that the waiting list was a useful tool for monitoring the need for services "*it is a fair system*" (P3) while others questioned its purpose:

> *"I think that initial idea with the waiting list, was... it was meant well, but it turned out to be a disaster. I think there*

> *exists, quite a huge misunderstanding, a lack of knowledge, regarding the... what is the waiting list? Waiting for what? Waiting for placement? Waiting for assessment? Waiting for a chair? What are we waiting for? And to me, it is sad... as much as I understand the need, that, we have to put people on a list to get the services to them, I disagree with them having to be on a list, especially the numbers that we're talking about..." (P10)*

Concerns about children on the waiting list not receiving any intervention while they wait for a placement at a school was raised:

> *"And that confusion, I think, contributes to part of the problem. Some think, no they are being serviced somewhere, so they're okay. And then, yet there are others that are also on the waiting list, but they're sitting in the home, so they're out of school, and nothing happens with them, for years on end" (P9)*

Service providers felt that children from lower-income homes with parents who did not complain or who were foreign nationals were more at a disadvantage:

> *"I think there's been a huge move of people into the Western Cape. And wherever you see immigrant groups, large numbers of children from the DRC*, there's been children from Somalia, and they are all trying to access services. These children are born in this country, so it's not a case that they've come from other places to access services, they were born here, but they are needing services and education. So I think we are having to extend the number... or increase the number of... or capacity of autism units" (P1)*
>
> **DRC – Democratic Republic of Congo*

Competency constraints

Participants attributed the current situation in service delivery to lack of capacity rooted in ignorance, denial and poor planning. The competence of decision makers in assessing the urgency of the ASD situation, strategic planning and taking action to manage the situation in the province was raised:

> *"No, there's a better word - ignorance. It's a pervasive ignorance in the society, about the need and the problem that we're facing. So yes, you know, speaking of other things, country-wide, everything on the news at the moment, is mismanagement, poor planning, but for me it's poor planning of Eskom*, poor planning of resources.. but for me, it's less.. it's not just poor planning, it is a not knowing, it wasn't even aware of this coming" (P10)*
>
> *South African electricity utility company

Service providers raised concerns that government stakeholders were not doing enough to manage the situation despite warnings from service providers. One service provider explained:

> *"so I don't know whether anybody could foresee it, but those that do... in the last six years, the same noises have been made, but I don't see a balancing. The writing was on the wall, people have been putting these things in meetings and in presentations and saying, listen, wake up, there's something coming, and I didn't see the counter to that." (P10)*

Service providers felt that decision-makers did not acknowledge the urgency of the ASD situation. The increasing number of children being diagnosed with ASD that would require services was not taken into consideration for future planning:

> *"... well maybe it's living in denial, maybe it's just simply saying, can't deal with that now, got other crisis to deal with, fighting fires within the moment, but not making long term plans.... I don't think there's enough future planning, projections made, and strategic planning toward those projections." (P9)*

Service providers felt that there was no clear sense of who "owned" the problem and who accepted accountability and responsibility. Instead, there was a tendency to shift responsibility and not generate solutions to the problem:

> *"... who's problem is it? Is it the country's problem? Is it that community's problem? Is it a social... you know, is it a worldwide problem? Is it the government's problem? Who's problem is this? And if we're now particularly talking about autism, autism is on the rise, who's problem is it? And can we shrug shoulders and say, well it's not my problem, you know, I'm doing this kind of work, that's your problem, find help for it... So we all sort of put these little blinkers on, and I know I'm making a sweeping statement, but it is easier, than to then live in the denial, and say, you know what? I'm doing the best I can, just to get my yard clean, and leave that one with the problem, to deal with their problem. Yet that person, has questions, has need for support, where... who's door, do they knock on to? I don't know, I don't have that answer. It's a philosophical one." (P10)*

Category 3: Building bridges

The third category described service provider acknowledgment of the efforts made by the current leadership and their suggestions for improved ASD service delivery in four sub-categories: leadership, capacity-building, co-ordination and innovation.

Leadership

Despite the numerous challenges alluded to in the preceding categories, service providers felt that the public sector services in the Western Cape province were taking the lead and doing much better than the rest of the country in terms of ASD service delivery:

> *"Contrary to popular belief, the Western Cape is streets ahead of the rest of the country, and for many reasons. And the first reason is, the coordinated way in which things are done. There is a consolidated database (waiting list), which is maintained very efficiently, extremely efficiently, with all sorts of information about each child in that database. It's a tracking mechanism for where a child is, what happens to the child, etc., etc. It is a fair system…Fair in the sense that you obviously have to look at children first, who are school going age, those children need to be prioritized, it is first come first serve, it's not about financial status, socio-economic structure, culture doesn't play a role" (P3)*

Capacity-building

The expertise and experience necessary for working with children with ASD was raised. Service providers felt that increasing the competence in school staff would result in improved inclusion of children with ASD in the educational system:

> *"The biggest shift they* [teachers] *need to make, is to know that in this class, you need to prepare a child for life, and not teaching math and literacy. And that's a teacher thing. Teachers are born and trained to teach math and literacy. And to make that shift, to helping you to wipe your nose, is also part of learning and getting you ready…So don't feel like you're not doing your work when you're not teaching, you're playing all day…" (P4)*

Participants proposed that the multidisciplinary educational outreach teams that were appointed by the Western Cape Education Department mainly to support the ASD schools and units across the province could support children with ASD in mainstream schools as well and advocate for better inclusion of these children in the broader education system:

> *"There needs to be a service like the outreach, but it's not just for special schools, not just for screening. It should be, here we've got a service, a referral is your service, we'll come and help this teacher to include this child in her class. And that's what I was doing for nearly 2 years, and it works. But you've got to get the staff trained, you've got to get them on the same page, they're going to accept this child. And it's a whole school issue, it's whole school philosophy." (P9)*

The provision of essential home programmes, early intervention services and family support programmes could assist in bridging the service gap while children with ASD wait for school placement:

> *"I think if we could have more home programme type things, like they have here at* [an ASD school], *that would maybe be useful. So then while children are waiting on the waiting list, they at least have some intervention in the meantime as well, or some extra support…If it was an ideal world also supporting the whole family, because often I think parents struggle, the siblings struggle, so the whole family could be supported. That would be ideal" (P5)*

Planning and coordination

Participants advocated for long-term planning of sustainable solutions that ensured coordinated intersectoral actions to provide educational opportunities for children with ASD:

> *"In the Western Cape, our biggest drawback is planning. I think we are all doers. It feels like everyone wants to jump in and do, and I'm not a planner, that's maybe why it's frustrating for me as well, I'm also a doer, I see the problem, I want to fix it. But we need to take a step back, because especially, I've looked on the waiting list yesterday, especially with the young ones coming, that are identified, is someone planning for what's coming? Is someone looking at what is on their way? Or are we just trying to figure out what to do with the ones that we have now?" (P4)*
>
> *"Research has shown, where the best services across the world is rendered, is where you have very close inter-departmental collaboration. And that is, I think, one of the reasons why we struggle, it is almost non-existent." (P3)*

Innovation

Participants advocated for innovation that would design contextually-relevant educational opportunities for every child with ASD to be among their typically-developing peers. They felt that innovative ideas could emerge from inter-professional collaboration in the design of early intervention packages drawing on international best practice examples:

> *"If it's an ideal world, I'd like to have a child in a school…and the neurotypical children need to learn about how to engage with an autistic child. However, there are those children that need, and if you're asking ideal world, then I'm saying there must be way more facilities. Not only big schools with three hundred of these children together; smaller, small facilities with fifty children in, ten classes of five each, you know."* (P9)

Service providers acknowledged that ASD is a particularly challenging condition to manage and by drawing on the experiences of other countries, the Western Cape could develop a contextually relevant model for ASD service delivery as one service provider explained:

> *"I think, with intelligent people who know the landscape, to craft something that is necessarily pragmatic. There are going to be disappointments, it's an incredibly challenging condition, that the most developed countries in the world don't get right. But what are the core things that we can draw in, and build on.... let's make a concerted effort, to go look at international trends of developed countries, where they have trialed, and tried, and tested, and what is working and what does not work."* (P10)

Recommendations from participants for service strengthening of ASD education in the Western Cape

Table 3 shows a summary of the ten key recommendations made by participants for service strengthening of ASD education in the province. Narrative comments on these will be incorporated into the discussion.

TABLE 3 Participant recommendations for service strengthening of ASD education in the Western Cape.

No.	Service provider recommendations
1	Introduce early intervention and support to children younger than compulsory school going age on the waiting list
2	Support children and their families at home or in the community long before children require formal education
3	Rethink inclusive education so that the majority of children with ASD may not require special school placements
4	Change the stigma and perception about autism in mainstream education at primary, secondary and tertiary level
5	Develop training programmes for educators to enhance their skills to work with learners/students with ASD
6	Rethink curricula for children with ASD to be flexible, based on the needs of individual learners/students, and, where appropriate, focus on a meaningful range of life skills
7	Include users/carers and people with ASD into policymaking, curriculum development and training at all levels
8	Balance the need for more special educational provisions and improved access to mainstream settings for children with ASD
9	Avoid "knee-jerk" responses to service needs by development of long-term, integrated policies, plans and actions
10	Learn from international best practice examples to develop contextually appropriate solutions to meet the educational needs of children with ASD in South Africa

Discussion

The purpose of this study was to examine "software" elements of ASD educational services in the Western Cape province of South Africa that might firstly, complement the "hardware" data reported to date and secondly, inform systems strengthening through service re-organization, policy review, and the development of best practices in ASD services. Earlier studies of the hardware of ASD educational services in the Western Cape province of South Africa identified a range of structural challenges – low identification rates of ASD, low identification of co-occurring diagnoses, complicated and inconsistent pathways to diagnoses and, concerningly, an observation that 89% of all children in school with a known diagnosis of ASD were in special school settings (25). In addition, the authors identified a large "waiting list" of children in need of special educational ASD placements, more than a quarter of which were of legal school-going age (26).

Using qualitative data from ten highly diverse ASD experts in the province, the findings suggested that participants perceived ASD services in the Western Cape as doing "the best we can to bridge the gap" despite the complexities of ASD population needs and prevailing contextual circumstances. However, in spite of doing "the best we can", the majority of participants expressed significant concern about various hardware (e.g., limited human resources, infrastructure, training) and software (e.g., lack of priority of ASD and other disabilities, knee-jerk responses, ignoring early intervention) elements and made a number of recommendations to strengthen education services in the province.

Participants acknowledged the complexity of the autism spectrum that inherently poses unique service delivery challenges. Vohra and colleagues (33) concurred that individuals with ASD experience more barriers in accessing services compared to individuals with other developmental disabilities or mental health conditions. Lai and Weiss (34) argued that the variable and lifelong nature of ASD makes planning for services challenging, pointing out that individuals with ASD have normative age-dependent service needs, including timely access to identification and diagnostic services, early intervention, school services, after-school and adult services. The participants in this study felt that all levels of services for individuals with ASD were lacking in the Western Cape.

Services providers pointed out that children were on a waiting list but that there were no systems in place to provide intervention or support for these children and their families while they waited. Studies have shown that early identification, diagnosis and intervention is essential for minimizing some of the core features of ASD and thereby positively influencing the functional trajectory of the disorder throughout schooling into adulthood (35, 36). The lack of early intervention services during the critical period of development has significant consequences for language, social and cognitive development with more financial costs relating to long term care and poor prospects of employment (5). A waiting list initiative to find and intervene in young children with ASD could be a very powerful strategy for improved future outcomes.

Service providers suggested that young children with ASD should receive services either at their homes or an interim place should be provided for them to receive developmentally appropriate stimulation. There is growing evidence internationally on the effectiveness of caregiver mediated interventions (37–39). In a study from the Western Cape province of South Africa, Guler et al. (40) suggested that contextually relevant and sustainable caregiver-led interventions could bridge the service gap in low-income countries where intensive early intervention programmes are not financially accessible to the majority of the population. Furthermore de Vries (14) argued that caregiver-led interventions such as naturalistic developmental behavioral interventions (NDBIs) would be ideal for LMIC contexts where the need is far greater than the number of "expert" service providers available to deliver interventions. Therefore foregrounding policy initiatives and service efforts that target early intervention will contribute to bridging the current education service gap in the long term.

Apart from the need for early action, the ASD diagnosis also influences access to development opportunities throughout the lifespan. Critical to these opportunities is the mainstream participation and inclusion of people with disability in learning environments. Hehir et al. (41) reported on the short- and long-term benefits of inclusive education for people with disabilities: improved social and cognitive development as well as better opportunities for further education and employment. In the study by Pillay, Duncan and de Vries (25) 89% of the school-going population of children with ASD in the Western Cape attended schools for children with special educational needs and only 10% were in ordinary mainstream schools. An important perspective held by the service providers that a significant proportion of children with ASD in special schools could potentially be better placed in mainstream education supports the need for action to identify and shift children with ASD in special schools to mainstream schools. This however would require advocacy and training to facilitate successful inclusion of these children in mainstream schools.

Service providers felt that a label of ASD was associated with "fear" in educators and poor inclusion of children with ASD in mainstream schools. Nah and Tan (42) reported that caregivers seeking mainstream education services for their children with ASD in Singapore were hesitant to disclose the diagnosis of ASD due to fear of stigma and perceived negative attitudes of educators. Studies have shown that mainstream educators tend to have negative perceptions of teaching children with ASD due to lack of competence in managing social, communication and behavioral issues associated with ASD (43). According to Simpson et al. (44) a diagnosis of ASD warrants highly qualified educators with sound knowledge of evidence-based practices. They suggested that higher education institutions like universities and educator training colleges should work collaboratively with schools to improve the scope of training programmes to prepare educators for working with children with ASD (44).

A diagnosis of ASD also warrants curriculum content that is shaped by knowledge of the educational needs of this population (44, 45). Service providers in the current study expressed concerns that the skills taught to children with ASD in special schools were "mundane" and not optimal for development of their full potential. ASD special schools in the Western Cape do not follow a prescribed curriculum, only some of the schools work with individualized education and development plans (IEDPs) and children leave school at the age of eighteen without a national certificate and with limited vocational opportunities. In South Africa there are no policies specific to what children with ASD should learn, how they are taught and where they should be educated. Service providers felt that those responsible for developing special needs education policies and curricula had little or no knowledge of ASD and that the people with the necessary educational expertise and experience were not consulted. In Canada, after decades of conflict with policy makers, parent groups, with the help of researchers were able to influence policy, improve service delivery including customized programmes for their children with ASD (8). The participants in this study advocated for differentiation of the curriculum taking into account the heterogeneity of ASD. The development of a customized and national qualification aligned curriculum for learners with ASD would go a long way toward addressing the current service gaps identified in this study.

Taking into consideration the many contextual constraints that service providers described, participants acknowledged the need for an appropriate range of infrastructure and resources to provide educational opportunities for every child with ASD according to their profile of needs. At the time of this research, the Western Cape was the only province in South Africa that had a list of children with ASD waiting for a place to become available in a special school. Although the waiting list has been contentious as outlined in comments from participants, others remarked that the waiting list itself was an important resource for identifying infrastructure and service gaps. Information about the number of children waiting, their ages and other socio-cultural factors could serve to inform future planning.

Service providers felt that the lack human and infrastructural resources, compounded by the lack of competence in decision makers could negatively impact educator wellbeing. With special schools under pressure from decision-makers to admit more children with ASD in a class as a means to alleviate the waiting list, service providers expressed concerns that educators would experience "burn out". Brunsting et al. (46) raised similar concerns about the wellbeing demands placed on ASD educators in specialized education settings. They argue that having more children with ASD in a class can lead to educator burn out despite educator's having the knowledge and skills to work with children with ASD. Mrstik et al. (47) reported that retention of educators of children with ASD in the special education sector was a major problem in the United States as well as other countries around the world resulting in a shortage of special educators. In a phenomenological study on the lived experience of educating students with ASD, educators reported that high workloads coupled with poor support from administrators was a major source of stress in educators (47).

While some service providers felt that more special schools should be provided for children with ASD others felt that greater efforts should be made to ensure successful inclusion of children with ASD in existing mainstream schools. In a study that explored the challenges and facilitators of mainstreaming children with ASD, Lindsay et al. (48) adopted the Lipsky and Gartner (49) model to inform their analysis. The model identified both human and financial resources as essential elements for inclusion (49). Similarly, Simpson et al. (44) argued for the benefits of allocating resources to address the increasing prevalence of ASD in contemporary societies, pointing out that policymakers should make financial resources available for professional development programmes and other resources that would allow mainstream classrooms to become more conducive to learning for children with ASD. The Western Cape is relatively well-resourced in comparison to other provinces (with two of the five ASD specific special schools in South Africa and several satellite ASD unit classes attached to special schools across the province), however some of the service providers felt that taking into consideration the increasing number of children being identified with ASD and the significant backlog of children already waiting for ASD educational services in the province, a range of appropriate educational opportunities for children with ASD should be explored and supported in order to bridge the service gap.

Service providers also commented on the need to learn from international good practice in order to develop contextually relevant local innovations to "build the bridges" between the current and future service landscape for children with ASD in the province. Service providers felt that greater efforts should be made for long-term planning rather than "knee-jerk" or reactionary decision making. Lessons from policymakers in Canada has shown that rushed decision-making due to political pressures resulted in poor outcomes for children with ASD and a more proactive rather than reactive approach was prescribed (8). Stronger intersectoral and inter-professional collaboration was advocated. Service providers felt that where collaboration between the different stakeholder groups was happening there was progress therefore calling for larger scale collaborative efforts. According to Cloet et al. (50) collaboration is key to meeting the diverse needs of children with ASD. Finally, service providers suggested that aligning with international ASD best practice frameworks and drawing on key concepts could support the development of contextually relevant policies and practices for the South African context.

Limitations and future research

We acknowledge a range of limitations of this qualitative study. First, this study was conducted in one better resourced province in South Africa, and in one sub-Saharan African country. Caution should therefore be taken to generalize findings to other provinces and other LMIC. However, the high level of concern expressed about ASD education in this South African province suggests that even greater challenges may present in other South African provinces and in other LMIC. Second, although the interviews were conducted primarily by the first author, all the authors were involved in developing the interview schedule and analysing the data. Third, results reflected the perspectives of a small group of ASD service providers and findings may therefore not be easily generalisable. However, the participants represented a broad range of perspectives, and many years of collective expertise and experience of the educational system in the province. Findings are therefore likely to be a fair representation of the state of education systems in the Western Cape. Fourth, data were collected in 2018, and there may have been some "hardware" and "software" improvements since the study. However, no major changes have been observed by the authors since the data collection for this study. More research would be necessary to determine the extent to which these improvements have impacted service delivery for children with ASD and their families. Fifth, our study focused on perspectives of service providers. It will also be important to seek the views of caregivers of children with ASD, and of government stakeholders in order to ensure a multi-level evaluation of the ASD educational landscape in the province.

Conclusion

Despite the many complex challenges of delivering educational services to children with ASD and their families in the Western Cape province of South Africa, the overarching message from participants was that everyone was doing the best they could. Service providers felt that services could improve if collaborative efforts were made by different stakeholder groups to understand and strengthen education systems. Educator training to facilitate inclusive education for children with ASD in the greater education system, parent-mediated early intervention, and intersectoral and inter-professional collaboration were identified as areas that could bridge the service gap. Drawing on international frameworks to develop contextually appropriate ASD policies and best practice for South Africa were recommended.

Data availability statement

Enquiries about the primary data can be directed to the corresponding author.

Ethics statement

Ethical approval was granted by the University of Cape Town Human Research Ethics Committee (HREC reference: 072/2016) as well as the Western Cape Department of Education (reference: 20150727-1712).

Author contributions

SP, MD, and PJDV participated in the conception and design of the study. SP collected the data. MD and SP were involved in the first level of data analysis, consensus on the final categories, and themes were reached through discussions with all the authors. All authors approved the final version of the manuscript.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

References

1. American Psychiatric Association. *Diagnostic and Statistical Manual of Mental Disorders*. 5th edition. Washington, DC: American Psychiatric Association. (2013). doi: 10.1176/appi.books.9780890425596

2. Schlebusch L, Dada S, Samuels A. Family quality of life of South African families raising children with autism spectrum disorder. *JADD.* (2017) 47:1966–77. doi: 10.1007/s10803-017-3102-8

3. Jones S, Bremer E, Lloyd M. Autism spectrum disorder: family quality of life while waiting for intervention services. *Qual Life Res.* (2017) 26:331–42. doi: 10.1007/s11136-016-1382-7

4. Vasilopoulou E, Nisbet J. The quality of life of parents of children with autism spectrum disorder: a systematic review. *Res Autism Spectr Disord.* (2016) 23:36–49. doi: 10.1016/j.rasd.2015.11.008

5. Cidav Z, Lawer L, Marcus S, Mandell, D. Age-related variation in health service use and associated expenditures among children with autism. *JADD.* (2013) 43:924–931. doi: 10.1007/s10803-012-1637-2

6. Turcotte P, Mathew M, Shea L, Brusilovskiy E, Nonnemacher S. Service needs across the lifespan for individuals with autism. *JADD.* (2016) 46:2480–9. doi: 10.1007/s10803-016-2787-4

7. Kanne S, Bishop S. Editorial Perspective: the autism waitlist crisis and remembering what families need. *J Child Psychol Psychiatry.* (2021) 62:140–2. doi: 10.1111/jcpp.13254

8. Shepherd C, Waddell C. A qualitative study of autism policy in Canada: seeking consensus on children's services. *JADD.* (2015) 45:3550–2564. doi: 10.1007/s10803-015-2502-x

9. McCall B, UK failing to meet the needs of people with autism. *Lancet.* (2017) 389:351–351. doi: 10.1016/S0140-6736(17)30197-6

10. Olusanya B, Davis A, Wertlieb D, Boo N, Nair M, Halpern R, et al. Developmental disabilities among children younger than 5 years in 195 countries and territories, 1990–2016: a systematic analysis for the Global Burden of Disease Study 2016. *Lancet Global Health.* (2018) 6:e1100–e1121. doi: 10.1016/S2214-109X(18)30309-7

11. Olusanya BO, Halpern R, Cheung VG, Nair MK, Boo NY, Hadders-Algra M, et al. Disability in children: a global problem needing a well-coordinated global action. *BMJ Paediatrics Open.* (2022) 6:e001397. doi: 10.1136/bmjpo-2021-001397

12. Tekola B, Baheretibeb Y, Roth I, Tilahun D, Fekadu A, Hanlon C, et al. Challenges and opportunities to improve autism services in low-income countries: lessons from a situational analysis in Ethiopia. *Global Mental Health.* (2016) 3:e21–e21. doi: 10.1017/gmh.2016.17

13. Abubakar A, Ssewanyana D, Newton C. A systematic review of research on autism spectrum disorders in sub-saharan Africa. *Behav. Neurol.* (2016) 2016:3501910–3501914. doi: 10.1155/2016/3501910

14. de Vries P J. Thinking globally to meet local needs: autism spectrum disorders in Africa and other low-resource environments. *Curr Opin Neurol.* (2016) 29:130–6. doi: 10.1097/WCO.0000000000000297

15. Franz L, Chambers N, von Isenburg M, de Vries PJ. Autism spectrum disorder in sub-Saharan Africa: a comprehensive scoping review. *Autism Res.* (2017) 10:723–49. doi: 10.1002/aur.1766

16. Statistics South Africa. Statistical release P0302 mid-year population estimates. (2019). Available online at: https://www.statssa.gov.za/publications/P0302/P03022019.pdf (accessed March 27, 2022).

17. Sulla V, Zikhali P. Overcoming Poverty and Inequality in South Africa: An Assessment of Drivers, Constraints and Opportunities. The World Bank Group. Washington, DC. (2018). Available online at: http://documents.worldbank.org/curated/en/530481521735906534/Overcoming-Poverty-and-Inequality-in-South-Africa-An-Assessment-of-Drivers-Constraints-and-Opportunities (accessed March 27, 2022).

18. World Population Review. Gini Coefficient by country (2022). Available online at: https://worldpopulationreview.com/country-rankings/gini-coefficient-by-country (accessed March 27, 2022).

19. Omotoso K, Koch S. Assessing changes in social determinants of health inequalities in South Africa: a decomposition analysis. *Int J Equity Health.* (2018) 17:181–13. doi: 10.1186/s12939-018-0885-y

20. Veriava F. *Realising the Right to Basic Education in South Africa: The Role of the Courts and Civil Society*. Claremont, Cape Town: Juta and Company. (2019).

21. Mayosi B, Benatar S. Health and health care in South Africa — 20 years after Mandela. *N Engl J Med.* (2014) 371:1344–53. doi: 10.1056/NEJMsr1405012

22. Department of Education. Education White Paper 6: Special needs education – Building an inclusive education and training system. 2001. Available online at: https://wcedonline.westerncape.gov.za/Specialised-ed/documents/WP6.pdf (accessed March 27, 2022).

23. Department of Basic Education. Education Statistics in South Africa 2016. Department of Basic Education. (2018). Available online at: https://www.education.gov.za/Portals/0/Documents/Publications/Education%20Statistic%20SA%202016.pdf?ver=2018-11-01-095102-947 (accessed March 27, 2022).

24. Hodgson TF. The right to inclusive education in South Africa : recreating disability apartheid through failed inclusion policies. *South African Law J.* (2018) 135:461–501. Available online at: https://hdl.handle.net/10520/EJC-11538df385

25. Pillay S, Duncan M, de Vries P. Autism in the Western Cape province of South Africa: Rates, socio-demographics, disability and educational characteristics in one million school children. *Autism*. (2021) 25:1076–89. doi: 10.1177/1362361320978042

26. Pillay S, Duncan M, de Vries PJ. Who*'s* waiting for a school? Rates, socio-demographics, disability and referral profile of children with autism spectrum disorder awaiting school placement in the Western Cape Province of South Africa. *Autism*. (2022) 13623613211067324–13623613211067324. doi: 10.1177/13623613211067324 [Epub ahead of print].

27. Sheikh K, Gilson L, Agyepong I, Hanson K, Ssengooba F, Bennett S. Building the field of health policy and systems research: framing the questions. *Public Library Sci.* (2011) 8:e1001073. doi: 10.1371/journal.pmed.1001073

28. Major CH, Savin Baden M. *Qualitative Research: The Essential Guide to Theory and Practice*. Abingdon and New York: Routledge. (2013). ISBN: 9780415674782

29. Babbie E, Mouton J. *The Practice of Social Research*. Cape Town, South Africa: Oxford University Press. (2014).

30. Thomas J, Harden A. Methods for the thematic synthesis of qualitative research in systematic reviews. *BMC Med Res Methodol.* (2008) 8:1–10. doi: 10.1186/1471-2288-8-45

31. Lincoln Y, Guba E. Establishing trustworthiness. In: Bryman A, Burgess RG. (Eds), *Qualitative Research.* London: SAGE. (1999).

32. Creswell JW. *Research Design. Qualitative, Quantitative and Mixed Methods Approaches.* 4th edition Thousand Oaks, CA: Sage. (2014).

33. Vohra R, Madhavan S, Sambamoorthi U, St Peter C. Access to services, quality of care, and family impact for children with autism, other developmental disabilities, and other mental health conditions. *Autism*. (2014) 18:815–26. doi: 10.1177/1362361313512902

34. Lai J, Weiss JA. Priority service needs and receipt across the lifespan for individuals with autism spectrum disorder. *Autism.* (2017) 10:1436–47. doi: 10.1002/aur.1786

35. Vivanti G, Dissanayake C. Outcome for children receiving the early start Denver model before and after 48 months. *JADD*. (2016) 46:2441–2449. doi: 10.1007/s10803-016-2777-6

36. Dawson, G. Early behavioral intervention, brain plasticity, and the prevention of autism spectrum disorder. *Develop Psychopathol*. (2008) 20:775–803. doi: 10.1017/S0954579408000370

37. Kasari C, Gulsrud A, Wong C, Kwon S, Locke J. Randomized controlled caregiver mediated joint engagement intervention for toddlers with autism. *JADD.* (2010) 40:1045–56. doi: 10.1007/s10803-010-0955-5

38. Wetherby A, Woods J, Guthrie W, Delehanty A, Brown J, Morgan L, et al. Changing developmental trajectories of toddlers with autism spectrum disorder: strategies for bridging research to community practice. *J Speech, Lang Hear Res.* (2018) 61:2615–28. doi: 10.1044/2018_JSLHR-L-RSAUT-18-0028

39. Naveed S, Waqas A, Amray AN, Memon RI, Javed N, Tahir MA, et al. Implementation and effectiveness of non-specialist mediated interventions for children with Autism Spectrum Disorder: a systematic review and meta-analysis. *PLoS ONE.* (2019) 14:e0224362. doi: 10.1371/journal.pone.0224362

40. Guler J, de Vries PJ, Seris N, Shabalala N, Franz L. The importance of context in early autism intervention: a qualitative South African study. *Autism.* (2018) 22:1005–17. doi: 10.1177/1362361317716604

41. Hehir T, Grindal T, Freeman B, Lamoreau R, Borquaye Y, Burke S. A summary of the evidence on inclusive education. (2016). Available online at: http://alana.org.br/wp-content/uploads/2016/12/A_Summary_of_the_evidence_on_inclusive_education.pdf (accessed March 27, 2022).

42. Nah Y, Tan JW. The effect of diagnostic labels on teachers' perceptions of behaviours of students with autism spectrum disorder. *Br J Educ Psychol.* (2021) 91:315–327. doi: 10.1111/bjep.12368

43. Hayes D. *Inclusion and Autism: General Education Teachers' Perceptions.* ProQuest Dissertations Publishing. (2014).

44. Simpson RL, Mundschenk NA, Heflin LJ. Issues, policies, and recommendations for improving the education of learners with autism spectrum disorders. *J Disab Policy Stud.* (2011) 22:3–17. doi: 10.1177/1044207310394850

45. Sanahuja Gavaldà Q. Improving the process of inclusive education in children with ASD (autism spectrum disorder) in mainstream schools. *Procedia.* (2012) 46:4072–6. doi: 10.1016/j.sbspro.2012.06.200

46. Brunsting NC, Sreckovic MA, Lane KL. Special education teacher burnout: a synthesis of research from 1979 to 2013. *Educ Treat Children.* (2014) 37:681–711. doi: 10.1353/etc.2014.0032

47. Mrstik S, Pearl C, Hopkins R, Vasquez III E, Marino MT. Combating Special Educator Attrition: Mentor Teachers' Perceptions of Job Satisfaction, Resiliency, and Retention. *Australasian J Special Inclus Educ.* (2019) 43:27–40. doi: 10.1017/jsi.2018.20

48. Lindsay S, Proulx M, Thomson N, Scott H. Educators' challenges of including children with autism spectrum disorder in mainstream classrooms. *Int J Disab Develop Educ.* (2013) 60:347–362. doi: 10.1080/1034912X.2013.846470

49. Lipsky D, Gartner A. *Inclusion and School Reform: Transforming America's Class-Room*. Baltimore, MD: Paul H Brookes. (1997).

50. Cloet E, Vissers S, Leys M. Interorganizational and multidisciplinary collaboration for persons with Autism Spectrum Disorder: towards more integration of care in Flanders. *Int J Integr Care.* (2019) 19:181. doi: 10.5334/ijic.s3481

frontiers | Frontiers in Psychiatry

TYPE Original Research
PUBLISHED 29 July 2022
DOI 10.3389/fpsyt.2022.903341

OPEN ACCESS

EDITED BY
Diana Schendel,
Drexel University, United States

REVIEWED BY
Mamta Nebhinani,
All India Institute of Medical Sciences
Jodhpur, India
Amanda Bakian,
The University of Utah, United States

*CORRESPONDENCE
Gauri Divan
gauri.divan@sangath.in

SPECIALTY SECTION
This article was submitted to
Autism,
a section of the journal
Frontiers in Psychiatry

RECEIVED 24 March 2022
ACCEPTED 01 July 2022
PUBLISHED 29 July 2022

CITATION
Naithani L, Sangwan P, Roy SG, Menon S, Azar Z, Lakhera S, Kumar D, Abhilashi MK, Roy R, Vajaratkar V, Taylor C, Patel V, Green J and Divan G (2022) From engagement to competency: The pathway to making disability naïve frontline workers competent in the delivery of an evidence-based autism intervention in New Delhi, India.
Front. Psychiatry 13:903341.
doi: 10.3389/fpsyt.2022.903341

From engagement to competency: The pathway to making disability naïve frontline workers competent in the delivery of an evidence-based autism intervention in New Delhi, India

Lavangi Naithani[1], Priya Sangwan[1], Sanjana Guha Roy[1], Sreepriya Menon[1], Zakiya Azar[1], Shweta Lakhera[1], Divya Kumar[1], Minal Kakra Abhilashi[1], Reetabrata Roy[1], Vivek Vajaratkar[1], Carol Taylor[2], Vikram Patel[3], Jonathan Green[2] and Gauri Divan[1]*

[1]Sangath, New Delhi, India, [2]Division of Neuroscience and Experimental Psychology, University of Manchester, Manchester, United Kingdom, [3]Harvard Medical School, Boston, MA, United States

Background: As countries like India improve access to maternal and infant care, the health systems need to develop services that enable all children to thrive. A key demographic which needs to be supported are children with disabilities, such as autism. With an estimated prevalence of one percent, there are over five million young children who need services to support their needs. However, the paucity of specialist care makes access to interventions difficult. In this context a public health research not-for-profit is evaluating the effectiveness of the task-sharing approach to support the delivery of an evidenced social communication intervention for young children with autism. This paper describes the process of engaging and training the non-specialist frontline Accredited Social Health Activists (ASHAs), who are embedded within the Ministry of Health and Family Welfare under the Delhi State Health Mission, to deliver a complex intervention for autism to inform the future scalability of services for neurodevelopmental disorders.

Methods: The present study describes the process which included (i) engagement meetings, (ii) recruitment, (iii) training, (iv) internship, and (v) competency evaluation. The shortlisted ASHAs received a 7-day classroom training followed by an internship period with practice cases. Finally, competency assessments, comprising of a test of knowledge and skills through role-plays, was administered.

Results: Twenty three Primary Urban Health Centers across seven districts of Delhi were approached and 408 ASHAs were engaged in initial meetings. Telephonic screening with 127 ASHAs resulted in 72 ASHAs being selected for in-person interviews. Of the 45 ASHAs who attended, 33 were shortlisted for training and 18 completed it. Fifteen ASHAs entered the internship of which 7 ASHAs achieved competency.

Discussion and conclusion: There was significant attrition along the pathway to having a competent non-specialist worker deliver a complex autism intervention. The lessons learnt from this process can inform the possibility of developing a cadre of disability specific frontline health workers who can deliver evidence-based interventions for neurodevelopmental disorders under supervision.

KEYWORDS

Accredited Social Health Activist (ASHA), autism spectrum disorders (ASD), evidence-based intervention, engagement, recruitment, competency, non-specialist health workers

Background

India has unique challenges in providing universal health and rehabilitation coverage to its population of 1.3 billion. Though improvements in the provision of health care over the last 75 years has had a positive impact on life expectancy, non-communicable diseases (which include mental health and neurological disorders) contribute a significant burden to death and disabilities (1). Individuals with Autism Spectrum Disorders or autism [hereafter] have a core social communication impairment, restricted and repetitive sensory—motor behaviors. It is a pervasive lifelong condition, where families and individuals require long term support which varies as the individuals' needs change over time (2). However, evidence-based early interventions can transform the trajectory and support increased inclusion and participation of an individual (3, 4). A population-based study of neurodevelopmental disorders in children aged 2–9 years in India, revealed that one in eight children have a neurodevelopmental disability including hearing impairment and intellectual disability (5). Amongst these, autism has an estimated prevalence of 1.12 (0.74–1.68) per 100 children (5). Unfortunately, the majority of these children in India do not receive evidence-based interventions and families struggle to access appropriate care (6).

Even in 2017, the country's doctor to patient ratio was less than the WHO's minimum recommendation of 1 doctor for every 1,000 people of the population (7). This acute lack of human resources was addressed by the Ministry of Health and Family Welfare (MoHFW) by establishing the National Rural Health Mission in 2005 and introducing the Accredited Social Health Activist (ASHA). This cadre of frontline health workers was designated with the task of promoting health awareness at a community level with an aim to increasing the utilization of primary health services by mobilizing populations (8, 9), and providing services for maternal and child healthcare (10).

The mission now deploys ASHAs across 32 states and Union territories in India, and an individual ASHA on an average caters to a population of 1,000 individuals (11). Their required characteristics include being women between the ages of 20–45 years, residents in their communities of work, who are or were married (12) with a minimum eight grade education (13). On recruitment, they receive foundational modular training of up to 5 weeks (14) on a range of topics which includes community health and rights, maternal health, new-born care, infant and young child nutrition, reproductive health and infections (15). The ASHA's role is incentivized against monthly targets which are contextually set, based on the needs of their communities. Studies have observed that the health outcomes of communities are reflected more by the nature of training (content and duration) provided to ASHAs, as against their educational qualifications (10). The importance of regular refreshers (13, 16) and competency-based trainings along with mentorship for monitoring knowledge levels while maintaining the skills and motivation of ASHAs (10) has also been observed.

The establishing of the Health Mission along with other policy moves have resulted in improvements in health statistics for example, a decline in infant mortality from 74.4 deaths per 1,000 live births in 2005 to 32 deaths per 1,000 in 2018 (17), and a shift in focus from child survival to supporting children to thrive. With this in mind the MoHFW launched the Rashtriya Bal Swasthya Karyakram (RBSK) in 2013, with an aim of providing early identification and interventions for children (0–18 years). The RBSK program covers 30 conditions including developmental delays and disabilities. Its services are delivered through the District Early Intervention Centres (DEIC) which are designated for interventions and follow ups, including the provision of specialized therapies for autism (11). The ASHAs role in it is to support home-based screening and monitoring of children. Though in theory, the RBSK aims to provide comprehensive diagnostic and rehabilitative services, studies have pointed out the manpower shortage in the DEICs (18–20), a high turnover of staff (20) which impacts the care being delivered and an uneven roll out across the states of India. As a result, most services for children with disabilities including autism are only available in metropolitan centers which require long commutes to specialized facilities, with associated intervention and transportation cost and loss of daily wages for many families (21, 22). Within this context, a not-for-profit research organization, Sangath adapted an evidence-based intervention for autism to be delivered in the community using the process of task-sharing (22). Task-sharing is a successful strategy in addressing the paucity of specialists in low resource settings by allowing specialists to transfer certain skilled roles to non-specialists under supervision (23).

PASS, a social communication intervention for children with autism; has been adapted and expanded with a "Plus" component for task-sharing with non-specialists from the UK developed PACT intervention (24, 25). The non-specialists are supported with training and supervision to deliver 12 sessions to each parent—child dyad every fortnight over a period of 8 months. The intervention uses a 6–8-min parent-child play session recorded live on a smartphone. This is used by the counselor to guide the parent's learning of strategies which support social communication through reflective video feedback on segments of the recorded play. The parents are encouraged to practice the strategies for half an hour every day with their child to help reinforce new ways of interacting with their child. The Plus component supports the family with strategies for commonly co-occurring problems. Two pilot randomized controlled trials in South Asia set in two states of India, Goa and Kolhapur, have evaluated the acceptability and feasibility of the non-specialist health worker delivery of this intervention for autism (24, 26). Both trials used project-based non-specialists. However, to answer the question on scalability and sustainability, the aim of this current study is to determine whether an existing frontline worker; the ASHA could be trained to competently deliver this intervention.

This paper describes the process from engagement to competency of ASHAs as part of a larger trial, the Communication-centered Parent-mediated treatment for Autism Spectrum disorder in South Asia (COMPASS) project which is evaluating the effectiveness and cost-effectiveness of the PASS Plus intervention in New Delhi, India (ISRCTN ID: 21454676) (http://www.sangath.in/compass/). The project has received ethical clearance from the review boards at Sangath, University of Manchester and the Health Ministries Screening Committee in India. The aim of the COMPASS trial is to support the gaps in the RBSK and support the widened mandate of the ASHA's role beyond promoting child survival and providing standardized packages of care through DEICs. This paper delineates the steps in identification, recruitment, training and supervision of non-specialist frontline workers to support the scaling up of complex interventions, an important step in achieving universal health coverage of evidence-based interventions for children with disabilities.

Methods

This work carried out in Delhi had the following steps in the process of getting frontline workers to competency:

1) Mapping of families of children with autism

The COMPASS trial was recruiting families from two tertiary centres in New Delhi: Maulana Azad Medical College and associated Lok Nayak Hospital (MAMC-LNH) and All India Institute of Medical Sciences (AIIMS). The team reviewed their databases of children with autism and using this information mapped districts from the 11 within New Delhi, which had the highest concentration of children with autism. These identified districts were shared with the State Program Officer (SPO) and ASHA Nodal Officer, who issued letters of instruction to the Chief Medical Officer (CMO) of the identified districts. The aim was to support allocation of trial cases within districts to resident ASHAs thus minimizing their travel time. Steps 1 and 2 were carried out simultaneously.

2) Engagement with the health system

Along with mapping, the team *engaged with senior personnel in the health system*, the Senior Program Officer of the Department of Health, Delhi State Health Mission (DSHM); this involved discussions on the method of remuneration that was acceptable to the health system while allowing access to the ASHA workers.

The *engagement with the ASHAs* was planned as a two-step process. The first step were in the form of group meetings in which a standard presentation comprising a video on autism was shown at the primary urban health centres (PUHC), a nodal centre for ASHA workers. This was followed by a discussion and distribution of brochures which described the project, the intervention, its aims and their possible roles in it. ASHAs attending the meetings were informed that working with this project, would be an additional responsibility that they would have to shoulder. Remuneration or incentives the project would support for the work were discussed these were approved by the Program Officer and were linked to training and supervisions attended, sessions delivered, along with a bonus for completion of predetermined sessions per case. The ASHA supervisors, the Auxiliary Nurse Midwifes (ANMs) and Medical Officer In-charges (MOICs) were also invited to attend these engagement meetings. Key demographic data on all ASHA attendees was documented including their willingness to be contacted by the team again. The second step was a follow-up call within a week with those ASHAs who had expressed an interest during the meetings. The names of ASHAs who exhibited interest were then discussed with their respective MOICs, in order to gauge whether the ASHAs had a record of commitment and capability for additional work during the project period.

3) Recruitment of frontline workers

The initial *telephonic interview* was conducted with ASHAs who had shown an interest. They were given more details including the tentative project workload, required travel and their need for digital fluency with smartphones was enquired upon. They were then invited to an *in-person interview*, comprising five components: (i) An introductory presentation on the project (ii) small group reflective discussions around possible challenges for the work along with a case-based field scenario to allow the research team to gauge individual problem-solving abilities, communication, listening and collaborative skills; (iii) multiple-choice quiz to evaluate sensitivity toward disabilities and attitudes around teamwork and responsibility; (iv) digital skills task to assess proficiency with smartphones to support digital learning, data entry and navigating the basics of a smartphone interface, and finally (v) interviews to help evaluate an individual's understanding of the project activities and their commitment for long-term engagement with the project. A key inquiry was to understand the ASHA's ability to self-reflect on their personal strengths and weaknesses, to gauge the level of support from their family for this extra work and their willingness to travel out of their catchment area, if required (see Supplementary materials, Section I).

4) Training

Shortlisted candidates received a *seven-day training* of the PASS Plus intervention which included an overview of social communication, typical developmental parent—child interactions and foundational counseling skills. The trainings were scheduled on the weekends to facilitate ease of attendance and each training day ran for ∼6 h. Training was conducted by a team of intervention coordinators (ICs) with Masters' degree in the field of psychology, early childhood education, special education, and social work (LN, PS, SL, SGR, ZA, SM); who had achieved competency in the delivery of the PASS Plus intervention under the guidance of regional master trainers (VV and GD) (26). The training focused on autism, key skills and strategies of Stage 1 of the PASS communication intervention. The training schedule with topic areas covered and average time spent on each, is included as Supplementary materials, Section II. The training aimed at supporting digital literacy, an essential component for the video –feedback methodology used in the intervention. It included a blended approach with audio-visual content on a learning management system as building blocks for discussions and role plays. Focused observation of video clips of parent child interactions; a key skill for the non-specialists was a significant part of the training. Training was participatory to accommodate the needs of different learners and the content, methods and pace was continuously adapted as per the daily feedback given by the trainees, who had no previous training in developmental disorders. Each trainee was evaluated in several ways; a pre- and post-training knowledge test, module-specific quizzes and a trainer's checklist to evaluate participation and skills (on elements such as attention, oral participation, self-reflection, punctuality, role-play performance, particularly for their ability to describe video clips chosen) during the training. The cumulative scores on these various measures, helped identify high performing ASHAs in the training. See Supplementary materials, Section III, for details on some of the assessment measures used in training.

ASHAs who performed well across these multiple indicators were selected to enter an *internship* period, in which they were assigned two families of children with autism as practice cases. An Initial Home Visit facilitated by their ICs, served to introduce the "ASHA counselor" to the family. Each ASHA was initially paired with a project-based salaried non-specialist worker, who supported the session delivery by video recording the complete session. During this period, a supportive supervision structure was established, which included individual supervision of a minimum of three independently delivered sessions to practice cases along with attendance at four group supervision sessions of peer counselors. A critical aspect of all supervision sessions was the use of a quality rating measure (see Supplementary materials, Section IV). This measure evaluates the generic counseling skills and intervention-specific skills demonstrated within a session and is adapted and refined from the quality rating measure used in previous trials (26). In individual supervisions, ASHAs received one-on-one feedback from their assigned IC and in group supervision the peers were also encouraged to give feedback under guidance, so as to collectively develop peer supervision skills. Based on the skill or knowledge deficit mapped on the quality rating measures, group refresher trainings were held to build skills and team cohesion. ASHAs were encouraged to additionally fill a short self-reflection questionnaire which supported them to critically evaluate sessions they themselves had delivered.

5) Competency assessment

After delivering a minimum of three sessions across two practice families and if considered prepared on supervisory feedback, the counselor undertook a competency assessment. The PASS competency assessment is an objective review of both the knowledge and skills of a non-specialist. The former is assessed via multiple choice questions (e.g., on autism, communication and on case-based scenarios) and the latter was designed to be evaluated through an objective role-play of standardized session scenarios. The competency measure scores both general counseling skills (for e.g., the use of open-ended questions, paraphrasing and validating) as well as intervention-specific skills (such as identifying relevant video clips, choosing the correct strategy and addressing parent concerns). All trainees were unfamiliar with role-plays as an evaluation measure which resulted in some of them under performing during this component. To support a more realistic assessment of their skills an additional element was added to the competency measure, where an independent IC rated a recorded practice session using the same rating scale. ASHA counselor who were unable to achieve competency continued within their internship sessions before they were allowed to attempt the assessment again.

Results

We describe the results of the steps followed to support a disability naïve frontline worker from the health system in New Delhi, India, called an ASHA, to gain skills to become competent in the delivery of a complex social communication intervention for autism; PASS Plus.

1) Mapping of families of children with autism

Forty three PUHCs in eight districts were identified at the time of mapping, and the project was given permission to access 26, however only 23 of these across seven districts, were approached, based on health systems recommendation and the concentration of mapped families.

2) Engagement with the health system

Besides access to engage with 26 PUHCs in seven of the 11 districts of Delhi, the project received permission to incentivize the ASHAs for the training, supervision and sessions delivered for the project. The amount was agreed upon by the State Program Officer DSHM which reflected a pragmatic amount that the health system could support for scale up.

The *engagement meetings with frontline workers* took place in two tranches; from February 2019 to April 2019 and from April 2019 to May 2019. In some PUHCs, the MOICs and ANMs also attended the meetings. In the first tranche, the team visited 14 PUHCs across the South, Southeast, East, and Central districts. Two PUHC's had a high percentage of newly recruited ASHAs and were not approached. In the second tranche, nine PUHCs across Northwest, West, East and Southwest districts were visited. Renovations in one PUHC of the Southwest district prevented a visit. MOICs of most PUHCs ensured maximum attendance at these engagement meetings. A total 408 of registered 466 ASHAs attended these group meetings. These ASHAs (n = 408) represented districts which are tabulated in Table 1.

TABLE 1 Summary of PUHCs in Delhi and those approached by COMPASS.

S.No.	District name	No. of PUHCs permitted to access	No. of PUHCs approached	Total no. of ASHAs attending/ capacity*
1.	SOUTH	3	3	34 (36)
2.	SOUTH EAST	2	2	51 (55)
3.	CENTRAL	7	5	62 (70)
4.	EAST	5	5	91 (106)
5.	NORTH WEST	3	3	72 (84)
6.	SOUTH WEST	3	2	66 (74)
7.	WEST	3	3	32 (41)
	Total	**26**	**23**	**408 (466)**

*Total ASHAs registered at the given Primary Urban Health Centers.

Of the 408 ASHAs at the engagement meetings, 188 ASHAs showed an interest in the project on the follow-up call; 218 ASHAs declined the invitation to join; while two left immediately after the presentation without sharing their contact information. A few ASHAs asked for more time to support discussions with family members. Reasons given by ASHAs (n = 218) who declined to join the project included the inability to take on additional responsibility for a multiplicity of reasons which included; their current ASHA or household workloads, having dependent young or old family members, personal health concerns, a reluctance to travel and disinterest in engaging with new digital skills. A number of them reported a lack of family support in taking on this extra work, while in some PUHCs a lack of peers engaging with the project dissuaded a few from joining. To help support the last concern, the project strategized to recruit at least two ASHAs from one PUHC, as peer support.

3) Recruitment of frontline workers

ASHAs who received a positive recommendation from their MOICs were prioritized for the first tranche (n = 125). In the second tranche we widened the circle for recruitment to include ASHAs (n = 47) who showed motivation on the initial telephonic outreach. We included these recommended ASHAs along with those who had shown interest at the group meetings for *telephonic interviews*. Of 172 ASHAs, 127 engaged with the telephonic interview; 41 ASHAs who declined the telephonic interview cited similar reasons as above. Some additionally stated a lack of confidence in their line managers extending

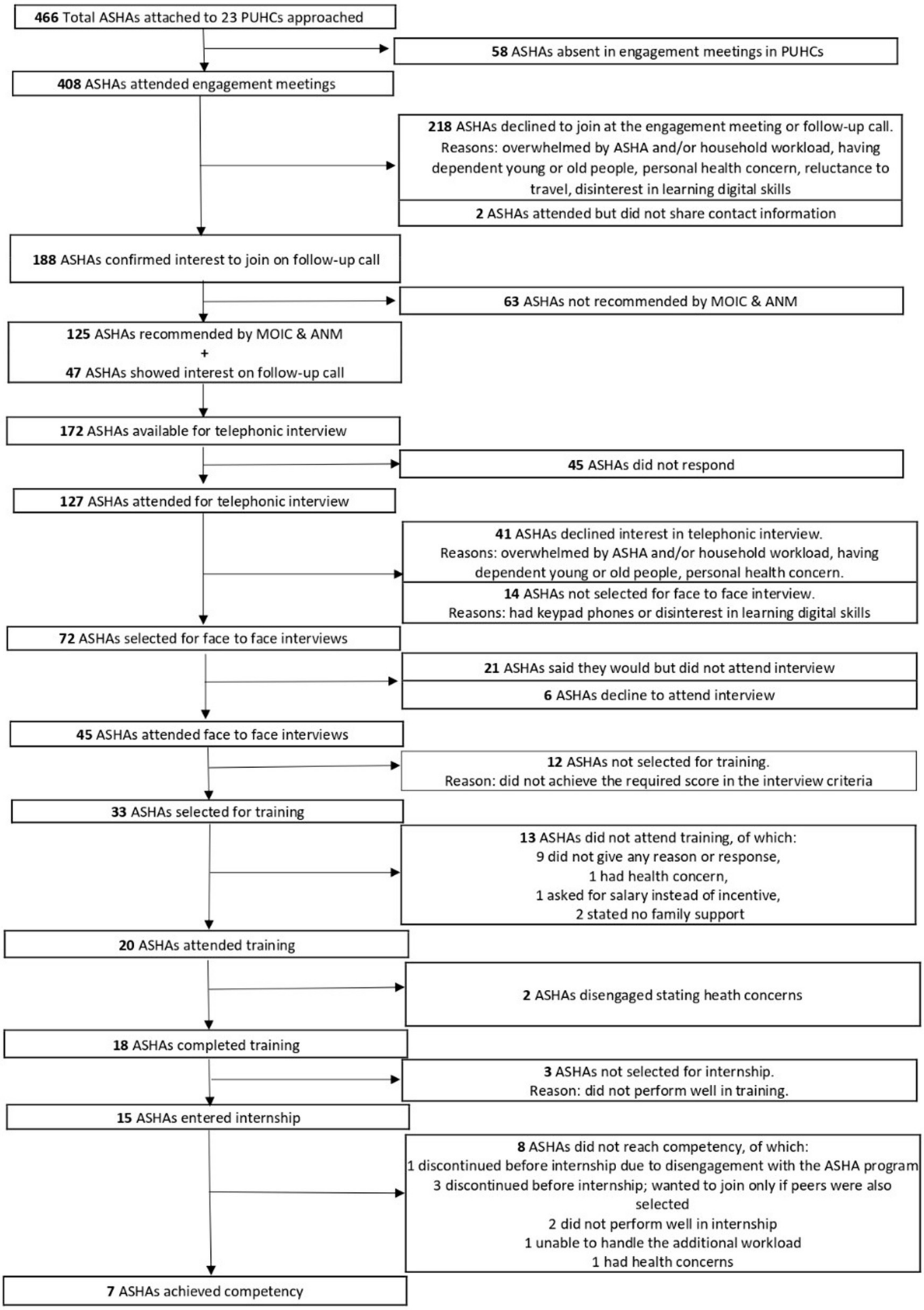


FIGURE 1
Flowchart depicting number of ASHAs involved at various stages of the project.

them support for this extra work, others revealed that they were already engaged in other private employment to supplement their incomes. ASHAs who owned smartphones and were familiar with digital applications were given a preference ($n = 14$). This latter criterion was eased for the second tranche and instead a digital literacy module was included within the training.

Seventy two ASHAs were identified for *in-person interviews*, of which only 45 attended. We were unable to reconnect with the majority of the ASHAs ($n = 21$) who did not attend the interview, though the six who responded stated that the geographic distance from the training centre was a barrier or that they were unavailable on the day of the interview. The age range of ASHAs attending the training represented women from 31 to 55 years; their work experience ranged from 6 months to 11 years (mean of 7 years) and their educational qualifications varied from 10th grade completion ($n = 25$); high school completion ($n = 15$), one with a diploma and seven with graduate education.

Thirty three ASHAs were selected for training based on their total interview scores. Of these, 20 ASHAs attended training and the majority ($n = 9$) of those declining to attend did not provide a reason. Of the other non-participating ASHAs, two were not supported by their families to take up this additional work, one ASHA shared concerns around her health which prevented her from travel associated with this work, and one ASHA clearly stated her dissatisfaction with the value of the incentives being offered and requested a salary.

4) Training

Eighteen of the 20 ASHAs who attended the first day of the training, completed it. Two ASHAs didn't attend all the days of training stating health concerns. For ASHAs who missed training days due to work at the PUHC or some personal obligations over the weekends, catch-up training was organized on weekdays. Based on the knowledge and skills gained during the training, 15 ASHAs were selected to proceed into the *internship* to deliver practice sessions under supervision.

The COMPASS team also obtained the functionality scores for the previous 6 months for the ASHAs. These are scores are tracked by the DSHM against the core activities that ASHAs need to conduct every month (e.g., immunization coverage, registering and supporting pregnancies, percentage institutional deliveries). ASHAs need to achieve a minimum of 50% on these core activities which allows their individual performance to be incentivized. An ASHA is deemed "functional" if they achieve a minimum of fifty percent every month. All ASHAs shortlisted for COMPASS had achieved this.

Six of the 15 ASHAs disengaged with the project during the internship period. One resigned from her duties in the health system, three ASHAs refused to join since their peers were not shortlisted, one ASHA found it difficult to balance both the project and ASHA workloads and another started experiencing significant health issues.

5) Competency assessment

Seven of the nine ASHAs passed the objective competency assessment. Collins et al. presented 24 shared "core" competencies for all mental, neurological, and substance use disorder providers that included empathic communication (27). Of these 24 competencies, there are six around screening and identification, two around formal diagnosis and referral and 16 around treatment and care. Of these, the training process was able to support the development of two within screening and identification (including the ability to describe the signs and symptoms of autism, and demonstrate cultural competence by an increased understanding of diverse families beliefs regarding autism); one supporting the recognition of the thresholds for referrals; and seven from the treatment domain (including supporting a community-based intervention for young children with autism, supporting the mental health of parents, developing and establishing a therapeutic alliance with the family). These competencies were measured during the assessment conducted after each counselor had delivered a minimum of three sessions across two practice families.

The 1998 National Community Health Advisor Study, conducted in the United States, identified a list of 18 qualities that are essential to have as a successful community health worker (28). Of these "being connected to the community" is a key characteristic of the ASHA defined by her being a resident in the community that she works in. As a trusted member of the local community who already conducts home visits, this made her the ideal choice for our intervention delivery. Other qualities such as being open-minded, non-judgmental, motivated and capable of self-directed work, and empathy (28), were evaluated through our interviews and training process, wherein we found that those ASHAs who across the interview phase and during their training and internship, could openly talk about their challenges and were observed to be motivated, committed, and showed openness to learn novel things managed to make their way to competency (see Supplementary materials, Section I).

Even after 9 months of supportive supervision and multiple refresher trainings, two ASHAs were unable to gain optimal skills during their internship and were discontinued from the project. The mean time to competency was just over 8 months, but was impacted by a number of the rate limiting issues which included the delay in assigning practice cases, and personal reasons which caused certain trainees to take a temporary break from the project.

The mean age and work experience of seven ASHAs who achieved competency is similar to that of all ASHAs ($n = 45$) interviewed. However, the overall gain in knowledge from the training of 7 competent ASHAs averaged (mean percentage scores 64% pre-training and 81% post-training scores) higher than that of 13 ASHAs who did not achieve competency (mean percentage scores 47% pre-training and 65% post-training). ASHAs who were above 50 years of age and had been in service for 10 or more years were minimally represented in the group chosen for training, and those that did, did not achieve competency. Figure 1 illustrates the attrition of ASHAs across the various stages of involvement with the project.

Discussion

We set out to evaluate whether health system frontline workers, the Accredited Social Health Activist, could be supported to learn and deliver a complex social communication intervention for autism in New Delhi, India. The process described here is nested in a larger ongoing effectiveness and cost-effectiveness trial and the impact of the ASHA training on intervention delivery to families with an autistic child will be published when the trial is complete. There is now substantial evidence for the potential of non-specialist frontline workers to effectively deliver a wide range of preventive, promotive and curative services (29). It has been shown that they can play a role in meeting the needs of poor and marginalized populations in contexts of weak health systems, resource constraints, and vast inequalities. In urban populations, frontline workers perform important roles that other providers are not well-positioned to deliver, including roles related to outreach, behavioral change, psychosocial support and managing chronic diseases (30). With this background knowledge, PASS Plus has been designed to address challenges faced by families of children with autism and is a community-based non-specialist delivered intervention. In the context of India, ASHAs are mandated to conduct home visits for improving the health within communities (31) and their core counseling skills were identified as a suitable choice for delivering a psychosocial intervention for autism, with a view to the potential for scaling up an evidence-based intervention.

In the current study, though we aimed to train a larger number of ASHAs, we were only able to get a fraction of the initial attendees interested in the work. There are a number of reasons for this. The initial high attrition can be attributed to the hierarchical system that ASHAs work in; this meant that many who may have been disinterested in the work were expected to attend the engagement meetings. Since the majority of ASHAs were naïve to disabilities, we wanted the process to support the identification of those individuals who were able to commit to additional work beyond their mandated activities. Our process revealed some obvious but also interesting characteristics. We found that years of work experience was not an accurate marker for engagement with this project, instead it was an inclination to learn something novel and to engage in new tasks which were critical to success. We noted that ASHAs who were in the position for many years seemed to lack the motivation to take on new challenges. On the other hand, new recruits to the health system, were still learning their core curriculum and were unable to take on the additional workload. In addition, the functionality scores we had obtained from the DSHM were neither indicative nor predictive of an ASHA's ability to deliver this complex intervention.

An ASHA's work is a part-time voluntary activity for which she receives an honorarium, incentivized for activities delivered. As a result, we found that an ASHA whose own personal commitments were significant [e.g., those with young children, or elderly dependents], were unable to make the time for this project. Over the period of internship, it was the ASHAs who had agency, reflected in their ability to make informed independent decisions without consulting other household members and who demonstrated the confidence to travel out of their catchment area, were able to engage with this work.

The PASS Plus intervention uses the technique of video feedback, and hence requires a level of digital literacy for its delivery. This influenced an individual's ability to be trained and succeed in delivering this intervention. Our finding that even without access to a smartphone, individuals who were keen to learn were able to enhance their personal digital literacy under supervision. It was the ASHAs who asked questions, repeatedly tried to master practical tasks, and who engaged with the topics being discussed, that achieved proficiency in the reflective approach used during session delivery for PASS Plus. Our training and supervision processes supported the development of ten of the 24 competencies suggested by Collins et al. (27), however, further research should aim to track quality of these competencies over time when such a program is implemented at scale.

A critical aspect of the project which has allowed the ASHAs we trained to deliver a quality intervention has been the building of a framework of supportive supervision and developing a methodology to encourage peer-to-peer support during the internship period. This helped create a skilled set of workers who have been able to stay motivated, engaged and keen to improve their skills. An objective quality rating measure and self-reflection questionnaire used consistently across sessions, allowed trainees to gain skills in evaluating sessions effectively. We observed that those who demonstrated the capacity of accurate self-reflection for their own sessions and had the ability to receive constructive criticism from peers and supervisors were able to quickly move to competency. The involvement of peers in programmatic training and supervision will be critical to ensuring the scalability of task-sharing for complex interventions in the future. However, these observations need to be studied systematically through further research.

A key limitation of our study is that we only worked in an urban setting of Delhi and hence the workers themselves may not be representative of non-specialist frontline workers across the country. Additionally, we were only able to reach out to a small number of ASHAs since this work was conducted within the timelines of the research project. It is possible that from a larger group we may have been able to identify a higher percentage of individuals who could be supported to gain competency.

Future empirical studies should also objectively measure the various personal attributes of community health workers such as the 18 qualities described by Brown et al. (28) along with those revealed through our processes, which may support the achievement of competency in delivering such complex intervention, and could be compared across intervention studies. The WHO Global Recommendation and Guidelines on task-shifting (32) states that having clear recruitment criteria that states behavioral and technical skills needed for the job makes identification of desired qualities in a candidate easier. Within India itself, there are other frontline workers such as an Anganwadi Teacher (AWT) or ANMs who may also be appropriate to deliver this intervention, depending on the context of the community, but this exploration was beyond the scope of this study.

With the prevalence of autism at one percent (5); it is clear that we do not need to train every ASHA in the intervention, however there is a need to be able to identify individuals who could potentially support families for any program that aims to scale up intervention services for children with autism. Through the methodology we used, we were able to identify key characteristics that would help the health system identify ASHAs who would be able to deliver complex interventions for autism.

Through this systematic process of recruitment, training and supportive supervision we have demonstrated a feasible methodology of training non-specialist frontline workers with no exposure to autism, to achieve competency in delivering a complex intervention using digital technology. However, we recognize that the process of identification of the characteristics of workers who could take on these roles needs refinement to optimize the effort made to achieve this at scale. While we await effectiveness data, this process illustrates a possible approach of scaling up an evidence-based intervention delivered within a supervisory framework. From expensive, difficult to reach city-based centers to community-based delivery in the homes of families of children with disabilities. This is an early indication that we can envision a system of support for vulnerable families of children with complex neurodevelopmental disorders. This support entails high quality services within their communities by a disability-specific worker, who can be trained across a number of disability modules and could then be integrated within the existing health system frameworks such as the RBSK in India.

Data availability statement

The original contributions presented in the study are included in the article/Supplementary material, further inquiries can be directed to the corresponding author/s.

Ethics statement

The studies involving human participants were reviewed and approved by Sangath IRB, Indian Council of Medical Research and the University of Manchester's Ethics Committee (UREC 2; ref no.: 2019-5223-11996). Written informed consent for participation was not required for this study in accordance with the national legislation and the institutional requirements.

Author contributions

GD, VV, CT, RR, VP, and JG conceived the idea for the research paper. LN, PS, SM, SR, SL, and ZA conducted literature review, with support from GD. RR, DK, and MA were involved in the formative work and engagement with the health system. ZA, SR, SM, PS, and LN were involved in mobilizing engagement meetings. LN, PS, SM, SR, SL, and ZA were involved in processes of recruitment, training, internship, supervision, and competency assessment of ASHAs. LN, PS, SM, ZA, SR, SL, and GD drafted the first version of the manuscript. LN, PS, SR, RR, VV, CT, VP, JG, and GD contributed to subsequent versions. All authors have read and approved the final manuscript.

Funding

This work has been conducted with the support of a Medical Research Council, UK grant. Grant Reference MR/R006164/1.

Acknowledgments

We would like to acknowledge the support of Sonakshi Pandey, Gitanjali Lall, Sanchita Johri, and Saani Shakeel in mobilizing the engagement meetings. Our biggest thanks is for the amazing frontline ASHA workers who have stood by this work in the face of personal challenges but also the devastating pandemic in India. We are also grateful to our funding agency Medical Research Council, UK that supported all research activities through the grant MR/R006164/1.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2022.903341/full#supplementary-material

References

1. Patel V, Chatterji S, Chisholm D, Ebrahim S, Gopalakrishna G, Mathers C, et al. Chronic diseases and injuries in India. *Lancet.* (2011) 377:413–28. doi: 10.1016/S0140-6736(10)61188-9

2. Green J. Editorial perspective: delivering autism intervention through development. *J Child Psychol Psychiatry.* (2019) 60:1353–6. doi: 10.1111/jcpp.13110

3. Gulsrud AC, Hellemann GS, Freeman SFN, Kasari C. Two to ten years: developmental trajectories of joint attention in children with ASD who received targeted social communication interventions. *Autism Res.* (2014) 7:207–15. doi: 10.1002/aur.1360

4. Pickles A, Le Couteur A, Leadbitter K, Salomone E, Cole-Fletcher R, Tobin H, et al. Parent-mediated social communication therapy for young children with autism (PACT): long-term follow-up of a randomised controlled trial. *Lancet.* (2016) 388:2501–9. doi: 10.1016/S0140-6736(16)31229-6

5. Arora NK, Nair MKC, Gulati S, Deshmukh V, Mohapatra A, Mishra D, et al. Neurodevelopmental disorders in children aged 2-9 years: population-based burden estimates across five regions in India. *PloS Med.* (2018) 15:e1002615. doi: 10.1371/journal.pmed.1002615

6. Divan G, Vajaratkar V, Desai MU, Strik-Lievers L, Patel V. Challenges, coping strategies, and unmet needs of families with a child with autism spectrum disorder in Goa, India. *Autism Res.* (2012) 5:190–200. doi: 10.1002/aur.1225

7. Kumar R, Pal R. India achieves WHO recommended doctor population ratio: a call for paradigm shift in public health discourse! *J Fam Med Prim Care.* (2018) 7:841–4. doi: 10.4103/jfmpc.jfmpc_218_18

8. Kohli C, Kishore J, Sharma S, Nayak H. Knowledge and practice of Accredited Social Health Activists for maternal healthcare delivery in Delhi. *J Fam Med Prim Care.* (2015) 4:359. doi: 10.4103/2249-4863.161317

9. Mane AB, Khandekar SV. Strengthening primary health care through asha workers: a novel approach in India. *Prim Heal Care.* (2014) 4:1–4. doi: 10.4172/2167-1079.1000149

10. Sundararaman T, Ved R, Gupta G, Samatha M. Determinants of functionality and effectiveness of community health workers: results from evaluation of ASHA program in eight Indian states. *BMC Proc.* (2012) 6:O30. doi: 10.1186/1753-6561-6-S5-O30

11. Jodhpur MS, Unicef MK, Sciences M. *Operational Guidelines Rashtriya Bal Swasthya Rashtriya Bal Swasthya Karyakram (RBSK) Child Health Screening and Early Intervention Services under NRHM Ministry of Health & Family Welfare.* Ministry of Health & Family Welfare Government of India (2012).

12. Panda M, Nanda S, Giri RC. A study on the work profile of ASHA workers in a district of Odisha in eastern India. *Int J Community Med Public Heal.* (2019) 6:675. doi: 10.18203/2394-6040.ijcmph20190190

13. Agarwal S, Curtis SL, Angeles G, Speizer IS, Singh K, Thomas JC. The impact of India's Accredited Social Health Activist (ASHA) program on the utilization of maternity services: a nationally representative longitudinal modelling study. *Hum Resour Health.* (2019) 17:68. doi: 10.1186/s12960-019-0402-4

14. Scott K, George AS, Ved RR. Taking stock of 10 years of published research on the ASHA programme: examining India's national community health worker programme from a health systems perspective. *Heal Res Policy Syst.* (2019) 17:1–17. doi: 10.1186/s12961-019-0427-0

15. National Health Mission (2013). *Induction Training Module for ASHAs in Urban Areas.* Available online at: http://nhm.gov.in/images/pdf/communitisation/asha/ASHA_Induction_Module_English.pdf (accessd March 12, 2022).

16. Shrivastava A, Srivastava A. Measuring communication competence and effectiveness of ASHAs (accredited social health activist) in their leadership role at rural settings of Uttar Pradesh (India). *Leadersh Heal Serv.* (2016) 29:69–81. doi: 10.1108/LHS-12-2014-0079

17. Office of The Registrar General of India. *SRS Bulletin, Sample Registration System, Office of the registrar general, India.* Vol. 52, Government of India (2019).

18. Panigrahy BK, Swain A. A Cross-sectional study to evaluate the functioning and infrastructure of mobile health teams and deic (district early intervention centre) at Koraput District of Odisha Under Rastriya Bal Swasthya Karyakram (RbSK). *World J Pharm Med Res.* (2019) 5:165–72.

19. Parmar DS, Raghunath DD, Dixit DS, Bansal DSB, Patidar DA. A cross-sectional study to evaluate the functioning and infrastructure of DEIC, and client satisfaction Ujjain and Indore districts established under RBSK. *IOSR J Dent Med Sci.* (2016) 15:92–4. doi: 10.9790/0853-1509119294

20. Rameshbabu B, Kumaravel K, Balaji J, Sathya P, Shobia N. Health Conditions screened by the 4D's Approach in a District Early Intervention Centre (DEIC) under Rashtriya Bal Swasthya Karyakram (RBSK) Program. *Pediatr Oncall.* (2019) 16:73–8. doi: 10.7199/ped.oncall.2019.48

21. Singh AK, Kumar R, Mishra CK, Khera A, Srivastava A. Moving from survival to healthy survival through child health screening and early intervention services under Rashtriya Bal Swasthya Karyakram (RBSK). *Indian J Pediatr.* (2015) 82:1012–8. doi: 10.1007/s12098-015-1823-2

22. Divan G, Hamdani SU, Vajartkar V, Minhas A, Taylor C, Aldred C, et al. Adapting an evidence-based intervention for autism spectrum disorder for scaling up in resource-constrained settings: the development of the PASS intervention in South Asia. *Glob Health Action.* (2015) 8:27278. doi: 10.3402/gha.v8.27278

23. Jeemon P, Anand TN, Maria Joseph L, Geetha A V, Prabhakaran D. Task sharing with non-physician health-care workers for management of blood pressure in low-income and middle-income countries: a systematic review and meta-analysis. *Lancet Global Health.* (2019) 7:e761–71. doi: 10.1016/S2214-109X(19)30077-4

24. Divan G, Vajaratkar V, Cardozo P, Huzurbazar S, Verma M, Howarth E, et al. The feasibility and effectiveness of PASS plus, a lay health worker delivered comprehensive intervention for autism spectrum disorders: pilot RCT in a rural low and middle income country setting. *Autism Res.* (2019) 12:328–39. doi: 10.1002/aur.1978

25. Green J, Charman T, McConachie H, Aldred C, Slonims V, Howlin P, et al. Parent-mediated communication-focused treatment in children with autism (PACT): a randomised controlled trial. *Lancet.* (2010) 375:2152–60. doi: 10.1016/S0140-6736(10)60587-9

26. Rahman A, Divan G, Hamdani SU, Vajaratkar V, Taylor C, Leadbitter K, et al. Effectiveness of the parent-mediated intervention for children with autism spectrum disorder in south Asia in India and Pakistan (PASS): a randomised controlled trial. *Lancet Psychiatry.* (2016) 3:128–36. doi: 10.1016/S2215-0366(15)00388-0

27. Collins PY, Musisi S, Frehywot S, Patel V. The core competencies for mental, neurological, and substance use disorder care in sub-Saharan Africa. *Glob Health Action.* (2015) 8:1–6. doi: 10.3402/gha.v8.26682

28. Brown O, Kangovi S, Wiggins N, Alvarado CS. Supervision strategies and community health worker effectiveness in health care settings. *NAM Perspect.* (2020) 2020:10.31478/202003c. doi: 10.31478/202003c

29. Cometto G, Ford N, Pfaffman-Zambruni J, Akl EA, Lehmann U, McPake B, et al. Health policy and system support to optimise community health worker programmes: an abridged WHO guideline. *Lancet Glob Heal.* (2018) 6:e1397–404. doi: 10.1016/S2214-109X(18)30482-0

30. Ludwick T, Morgan A, Kane S, Kelaher M, McPake B. The distinctive roles of urban community health workers in low-and middle-income countries: a scoping review of the literature. *Health Policy Plan.* (2020) 35:1039–52. doi: 10.1093/heapol/czaa049

31. NHSRC. *Comprehensive Primary Health Care through Health and Wellness Centers Operational Guidelines* (2018).

32. WHO. *Task Shifting. Global Recomendations and Guidelines.* World Health Organization (2008). p. 1–88.

frontiers | Frontiers in Psychiatry

TYPE Original Research
PUBLISHED 16 August 2022
DOI 10.3389/fpsyt.2022.915380

Age of diagnosis, service access, and rights of autistic individuals in Argentina: Caregivers reports of changes and similarities across time

Maria Cecilia Montenegro[1]*, Estefani Bernal[1], Sebastian Cukier[2], Daniel Valdez[3], Alexia Rattazzi[2], Gabriela Garrido[4], Analia Rosoli[5], Cristiane Silvestre Paula[6], Ricardo Garcia[7] and Cecilia Montiel-Nava[1]

[1]Department of Psychological Sciences, University of Texas Rio Grande Valley, Edinburg, TX, United States, [2]Research Department, Programa Argentino para Niños, Adolescentes y Adultos con Condiciones del Espectro Autista (PANAACEA), Buenos Aires, Argentina, [3]Research Department, Facultad Latinoamericana de Ciencias Sociales (FLACSO), Buenos Aires, Argentina, [4]Department of Child and Adolescent Psychiatry, Universidad de la República, Montevideo, Uruguay, [5]Research Department, Organización Estados Iberoamericanos para la Educación, la Ciencia y la Cultura (OEI), Santo Domingo, Dominican Republic, [6]Research Department, Programa de Pós-Graduação em Distúrbios do Desenvolvimento Universidade Presbiteriana Mackenzie (UPM), São Paulo, SP, Brazil, [7]Research Department, Universidad de Chile, Santiago, Chile

OPEN ACCESS

EDITED BY
Diana Schendel,
Drexel University, United States

REVIEWED BY
Benjamin Black,
University of Missouri, United States
Hemant Bhargav,
National Institute of Mental Health and Neurosciences (NIMHANS), India

*CORRESPONDENCE
Maria Cecilia Montenegro
maria.montenegro01@utrgv.edu

SPECIALTY SECTION
This article was submitted to Autism,
a section of the journal Frontiers in Psychiatry

RECEIVED 07 April 2022
ACCEPTED 22 July 2022
PUBLISHED 16 August 2022

CITATION
Montenegro MC, Bernal E, Cukier S, Valdez D, Rattazzi A, Garrido G, Rosoli A, Silvestre Paula C, Garcia R and Montiel-Nava C (2022) Age of diagnosis, service access, and rights of autistic individuals in Argentina: Caregivers reports of changes and similarities across time. *Front. Psychiatry* 13:915380. doi: 10.3389/fpsyt.2022.915380

Background: Many countries have developed health initiatives to protect those with disabilities and developmental concerns in the past few years. However, the needs of autistic individuals are still short of being fulfilled. Partially due to limited research expenditure, which would allow bridging the gap between evidence and practice, the long time it takes to implement passed laws, and the limited operationalization of inclusive policies.

Objective: To quantitatively examine changes in the child's age at the time of caregiver's first developmental concerns and age of diagnosis of their autistic child across 5 years. Also, to address challenges experienced by caregivers (e.g., reported service barriers) and the work still needing to be done in Argentina based on caregivers' reports of their priorities (e.g., ensuring their child receives better services).

Methods: Two independent samples of caregivers of autistic individuals were surveyed by the Red Espectro Autista Latinoamerica (REAL) in 2015 ($n = 763$) and the World Health Organization (WHO) in 2020 ($n = 422$). Similar items in both surveys were compared through descriptive inferential analysis and chi-square tests for categorical variables.

Results: Compared to the 2015 sample, for the 2020 sample, more caregivers reported an earlier age of first concerns and an earlier age of a professional diagnosis. In the 2020 sample, more children diagnosed before the age of three had a doctor or a teacher noticing the first developmental concern. Also, in this sample, fewer caregivers reported service barriers (e.g., limited availability, waitlist, costs, etc.) and a need for better social support and better health services. However, rates of caregivers indicating a need for more rights for autistic individuals and greater protection of existing rights increased. There was no change in the reported rate of family members who stopped working to care for the autistic individual. For both samples, there was statistically significant differences in individual (physician, teacher, caregiver) noticing first developmental concern and the age of diagnosis, with the majority having a caregiver noticing the first concern.

Conclusion: The 5 years that separate both samples show an improvement in developmental concerns being noticed, a decrease in age of diagnosis, and an improvement in several service areas such as community awareness. Also, caregivers reported fewer barriers to service accessibility, thus suggesting a positive impact stemming from changes in public policies, non-profit organizations' work through awareness campaigns, and advocates' strives toward greater awareness. Nonetheless, a similar proportion of family members reported ceasing working to care for autistic individuals and perceived that the fundamental rights of their autistic children needed to be protected. These results imply that despite better care pathways in Argentina, there are still gaps when attempting to meet the needs of autistic individuals and their families. The present study provides a meaningful understanding of existing gaps and help exemplify the perceived improvements when non-profit agencies and advocates promote increased rights and community awareness in addition to the established laws focusing on ASD.

KEYWORDS

caregivers, ASD, Argentina, age of diagnosis, first concern, service barriers, priorities

Introduction

The global prevalence of Autism Spectrum Disorder (ASD) is estimated to be 1 in 100 (1) and is in stark contrast to estimates in the United States showing an increased prevalence of ASD to 1 in 44 (2). Low global prevalence estimates suggest barriers to identification in other parts of the world. Although countries like the United States have minimized barriers to accessing appropriate evaluation and intervention services for children with ASD, other countries have historically lagged in this regard due to socioeconomic and cultural factors that further deepen the treatment gap and limit ASD community awareness (3–6). One of these countries, Argentina, is plagued by economic disparity throughout its regions (7), and information regarding Autism prevalence is limited. One study found the prevalence rate of disability in children to be 3.2%, with 40% of these children being identified with ASD (8). This may be an underestimation, as this study only included children who had obtained a Unique Certificate of Disability (UCD), and it has historically been identified that a significantly larger proportion of children with disability do not apply for a UCD (9). Despite its status as an upper-middle income country, Argentina experiences socioeconomic, political, and environmental inequities, all factors that have been shown to contribute to health disparities (10). The most recent economic data indicates that 40.6% of the population of Argentina lives in poverty, with an additional 10.7% living in extreme poverty (7). Although Argentina's health care system is on the path to being one of universal health coverage, discrepancies in the access to and the provision of health services exist among the population. Most recent financial information indicates 36% of the country's population has no insurance and relies on the public health sector for health treatment (7). Moreover, due to the structural framework and fragmented funding of the healthcare system, disparities in screening, time to diagnosis, and treatment of serious conditions have been found to vary among lower and higher-income districts and regions (11). These disparities could have a significant impact on the identification and treatment of conditions such as ASD, especially when considering the limited availability of trained professionals or specialists trained to identify ASD (5, 12, 13) and provide the required subsequent treatment (4). Furthermore, even when receiving treatment or intervention services such as speech therapy, occupational therapy, or behavior therapy, individuals are receiving services at a frequency below recommended therapeutic standards (14). Paula et al. (5) addressed barriers to care as reported by caregivers of autistic individuals in multiple Latin American countries, including Argentina. In said study, a high percentage of caregivers reported barriers to services reflected through long waiting lists, high treatment costs, and limited specialized services (5).

In addition to healthcare disparities and structural barriers to early ASD diagnosis and intervention, limited community awareness combined with unique cultural factors have been proposed to impact help-seeking behaviors (15). Latino parents within the United States report experiencing limited knowledge of ASD and resources available as well as cultural views that appear to be instrumental in delaying ASD diagnosis (16). Although there is evidence to suggest the experiences of parents in Latin American countries might be similar (5, 17) research exploring parent experiences regarding their understanding of ASD as well as information available to them and the community has been deficient (5, 18). This has begun to change considerably within the past decade, primarily through the self-advocacy movement and the establishment of parent support groups and autism associations. The establishment of organizations such as Red Espectro Autista (RedEA), which consists of representatives from various autism support groups, serves to increase ASD awareness, empower autistic individuals and family members, promote educational inclusion as well as political and social changes in Argentina (19, 20). In the area of research, networks such as Red Espectro Autista Latinoamericana (REAL) have been established to allow opportunities for Argentinian advocates and researchers to collaborate with other Latin American countries in research to promote increased awareness of ASD in Latin America (5). This movement toward advocacy and awareness has led to increased research opportunities focusing on interventions for parents (21, 22), as well as the validation of popular screening and diagnostic tools for Latin American populations (23–31).

Argentina has also passed laws, focusing on individuals with disabilities and in some cases focusing solely on ASD. For example, Act 27043 (the Comprehensive and Interdisciplinary Approach to persons with Autistic Spectrum Disorders) passed in 2014 with the aim to "ensure clinical and epidemiological investigation, early detection, diagnosis and treatment, dissemination and access to intervention services to autistic spectrum disorders" and to integrate early screening and diagnostic procedures along with required services into the Mandatory Medical Program (9). Yet, despite its passing, autistic Argentinians had to wait more than 5 years for this law to finally be legally implemented (32). This is unfortunate when one considers that early identification and diagnosis of ASD can lead to better outcomes for autistic individuals through the early access and utilization of intervention services (33). However, despite its benefits, for many autistic individuals, access to early intervention does not occur due to delayed diagnosis (17, 34). Although evidence indicates ASD can be diagnosed by 18 months of age, the average age of diagnosis for children in the United States is 4 years of age, and for some minorities, diagnosis occurs later (35–37). At present, there is limited information regarding the age of diagnosis for ASD in Latin America; however, some research estimates have identified a mean age of diagnosis at approximately 4.5 years of age with initial concerns having been noted at 2 years of age (14, 17, 38). This delay in diagnosis may profoundly impact the life trajectory of autistic individuals and their families.

Despite the limited understanding concerning the ASD experience for autistic individuals and their families in Argentina, this paper responds to the call made by the ASD community to represent better ASD knowledge outside the United States and European Countries (18) by exploring the changes in patterns of diagnosis and caregiver perceptions across a five-year span in Argentina. Also, it seeks to address the possible changes that have occurred since the previously mentioned non-profits launched and laws were implemented.

Aims

The present study's aims are two-fold. First, quantitatively examine changes in the age of developmental concerns of autistic children, age of diagnosis, and diagnostic awareness across 5 years. For this purpose, we examined changes in the age and type of first concerns observed by caregivers, differences in age and type of diagnosis received by autistic children, and the association between the person who noticed the first concern (e.g., parents, physician or teacher) and the eventual age of diagnosis. The second aim is to identify changes in the challenges experienced by caregivers (e.g., reported service barriers) and the priorities and needs of Argentinian caregivers of autistic children (e.g., ensuring their child receives better services). This last aim would offer some information on the possible impact of implemented awareness campaigns and laws aimed at protecting individuals with disabilities on the reduction of systemic barriers in Argentina.

Method

Sample characteristics

Caregivers of autistic individuals completed an anonymous online questionnaire in Argentina at two different times, 2015 and 2020. Respondents were different each year the questionnaire was distributed. The 2015 survey consisted of 763 caregivers. Table 1 shows the demographic characteristics of both samples. For the 2015 sample, the majority of caregivers had some college or a higher educational degree ($n = 534$, 70.7%), and the most common diagnosis in their child was ASD or an autism diagnosis 34.1% ($n = 260$). In the 2020 sample, most of the sample had some college education or a higher degree as well ($n = 341$, 80.8%). Of the 422 respondents, the majority reported their child as autistic or ASD ($n = 305$, 72.3%). While children attending a private school was the top chosen alternative ($n = 344$, 49.4%) in the 2015 sample, in the 2020 sample, almost half of caregivers indicated the *other* alternative ($n = 170$, 47.9%). From this last group (2020), 31.5% ($n = 54$) of caregivers who endorsed the *other* category indicated that their child was attending a special school for children with disabilities. The age of autistic children was similar across samples, with most caregivers endorsing having a child

TABLE 1 Demographic characteristics of the sample.

	2015 $N = 763$		2020 $N = 422$		x^2	p
	N	%	N	%		
Caregiver education					27.98	***
Primary	15	(2)	9	(2.1)		
Secondary	206	(27.3)	72	(17.1)		
Some college	257	(34)	130	(30.8)		
College degree	216	(28.6)	173	(41)		
Graduate degree	61	(8.1)	38	(9)		
Age of child					1.29	
Younger than 6	273	(35.8)	117	(39.5)		
Older than 6	490	(64.2)	179	(60.5)		
Diagnosis					182.101	***
Autism/ASD	260	(34.1)	305	(72.3)		
PDD-NOS	257	(33.7)	60	(14.2)		
PDD	128	(16.8)	47	(11.1)		
Asperger	118	(15.5)	10	(2.4)		
Type of School					37.13	***
Public school	276	(39.6)	78	(21.8)		
Private school	344	(49.4)	85	(23.8)		
Other	37	(5.3)	171	(47.9)		
No longer goes to school because of age	40	(5.7)	23	(6.4)		
Available school assistance	**355**	**(47.1)**	**145**	**(42.6)**	1.86	

Includes valid responses only, may not total sample.
$^*p < 0.05$; $^{**}p < 0.01$; $^{***}p < 0.001$.

older than 6 (Table 1). For both samples, inclusion criteria specified participants being at least 18 years old and the caregiver of an autistic child. The exclusion criteria were caregivers of individuals without an ASD reported diagnosis or those residing outside Argentina. To better allow for comparisons, both samples were drafted through similar channels. Both in 2015 and 2020, questionnaires were distributed through civil organizations like PANAACEA and RedEA, the largest autism non-profit agencies in Argentina. This allowed for samples to be comparable regarding autism awareness since participants in both were connected to similar organizations.

Procedure and instrument

Both surveys were adapted from the Caregivers Needs Survey (CNS) developed by Autism Speaks to better understand autistic individuals' needs (39). This is a retrospective study in which caregivers of autistic individuals were surveyed about their clinical and social history. The questionnaire inquired about: demographic information of caregivers and autistic individuals (e.g., age, educational attainment), first developmental concern (type and age it occurred), information on ASD diagnosis (e.g., age, provider who diagnosed, diagnostic label), service utilization, educational services, caregivers' perceptions, perceived impact of ASD, stigma, quality of life, and challenges and priorities. For both adapted versions of the survey, the CNS Spanish translation was reviewed for appropriateness and adapted by REAL clinicians who work with children with developmental disorders and their families. Following this adaptation, caregivers of autistic individuals examined the instrument, and wording was modified better to reflect autistic individuals and their families' experiences. The final version of each survey differs in some items, yet similar items across both samples were compared for this study's purpose. These comparisons were on items regarding demographic characteristics of autistic children and caregivers, service barriers, diagnostic and first concern information, and caregivers' perceptions regarding rights and needs. Montenegro et al. (40), Montiel-Nava et al. (14), and Paula et al. (5) describe the adaptation process for the 2015-version of the survey. This is the first publication of the 2020 version. Both versions were distributed online. After entering the website and prior to start completing the questionnaire, participants needed to provide their consent by typing their initials. Approval to conduct this study was obtained through a local ethics board.

Statistical analysis

Data was filled out in excel files and merged in SPSS for the statistical analysis. Comparisons between items in the two surveys were conducted through descriptive inferential analysis and chi-square tests of independence At least 80% of expected cell frequencies were greater than five. All statistical analyses were performed using IBM SPSS, version 26.

Variables

Respondents characteristics

Both surveys inquired about demographic information by asking about caregiver education (categorical), child diagnosis (categorical), and the type of schooling the child attended (categorical). In addition, the samples included autistic adults that could have been diagnosed while the PDD category was the current one.

First concerns

To better understand caregivers concerns regarding their child's development, type of first observed concern and the age at which it occurred were queried. For the observed first concern, caregivers could choose among six types (categorical;

TABLE 2 First developmental concern and age of diagnosis.

	2015 $N = 763$		2020 $N = 422$		x^2	p
Age of first concern					113.27	***
0–12 months	108	(14.4)	82	(19.6)		
12–18 months	85	(11.4)	121	(28.9)		
18–24 months	142	(19.0)	101	(24.2)		
24-36 months	249	(33.3)	79	(18.9)		
36–72 months	155	(20.3)	30	(7.2)		
72+ months	9	(1.2)	4	(1.0)		
Observed first concern*						
Medical problems	135	(19)	64	(15.2)	2.64	
Behavior difficulties	460	(61.8)	117	(27.7)	125.28	***
No response to sounds/name	473	(63.9)	267	(63.3)	0.37	
Sameness/difficulty with changes	466	(63)	179	(42.4)	45.98	***
Gross motor problems	178	(24.5)	97	(23)	0.329	
Age of diagnosis					15.136	**
0–3 years old	500	(66.4)	325	(77.2)		
4–8 years old	201	(26.7)	77	(18.3)		
9–12 years old	34	(4.5)	12	(2.9)		
13–17 years old	13	(1.7)	2	(0.5)		
18+ years old	5	(0.7)	5	(1.2)		

Includes valid responses only, may not total sample.
*$p < 0.05$; **$p < 0.01$; ***$p < 0.001$.

TABLE 3 Sample year and individual who noticed first developmental concern.

	2015 $N = 747$		2020 $N = 392$		x^2	p
	N	%	*N*	%		
First person noticed concern					47.32	***
Family member	612	(81.9)	253	(64.5)		
Physician	54	(7.2)	72	(18.6)		
Teacher	81	(10.8)	130	(16.8)		

Includes valid responses only, may not total sample.
*$p < 0.05$; **$p < 0.01$; ***$p < 0.001$.

medical problems, behavioral difficulties, no response to sounds or names, insistence on sameness or difficulty with changes, and gross motor problems). Caregivers could pick one or multiple concerns that applied to their child. Age of first observed concern was categorized in ranges (0–12 months, 12–18 months, 18–24 months, 24–36 months, 36–72 months and 72 months and up). For the person noticing first concern, alternatives to choose from were family members, doctor, and teacher (categorical, see Tables 2, 3).

Service barriers and challenges

Multiple alternatives were provided for types of barriers when accessing services for their autistic child (e.g., not qualifying for services, services not available in the area, among others, Table 5). Caregivers could indicate more than one service barrier (binary, yes and no). To assess financial impact of ASD, participants were asked whether a family member ceased work to care for autistic child (binary, yes and no). Binary options were provided concerning challenging characteristics observed in autistic children, and caregivers could endorse more than one (e.g., problematic behaviors, daily living skills, health problems, and so on). These challenging behaviors were summed together to assess number endorsed by caregivers (Table 6).

Age of diagnosis

Age of diagnosis was categorized into ranges (0–3, 4–8, 9–12, 13–17, and 18+ years old) (Table 4).

Priorities

Caregivers' perceived priorities in terms of support, community awareness, and autistic individuals' rights were measured using binary alternatives (yes and no). Caregivers could choose more than one item (Table 5).

Results

Age of first concerns

In terms of the age of first concerns, there were significant differences between the two samples ($x^2(9) = 113.274$, $p < 0.001$). Caregivers noticed developmental first concerns earlier in the 2020 sample. The category of concern noticed at 0–12 months increased from 14.4 % ($n = 108$) in 2015 to 19.6% ($n = 82$) in 2020. This also occurred with the age range of 12–18 months with 11.4% ($n = 85$) in 2015 and 28.9% ($n = 121$) in 2020, and 18–24 months which increased from 19% ($n = 142$) in 2015 to 24.2% ($n = 101$) in 2020. Overall, in the 2015 sample, 44.8% ($n = 335$) of caregivers noticed a first developmental concern before their child's 24 months, whereas 72.7% ($n = 304$) did so in the 2020 one.

Type of first concerns

There was no statistically significant difference ($x^2(1) = 0.37$, $p > 0.05$) when comparing the most common observed first concern, *not responding to name,* which remained the highest choice with ~64% of caregivers in both samples endorsing it. Both samples' least common first concern was the presence of medical problems ($x^2(1) = 2.34$, $p > 0.05$), remaining below 20% (Table 2).

TABLE 4 Age of diagnosis and person who first noticed developmental concern.

	Family member		Physician		Teacher		Total			
Age of diagnosis	***N***	**%**	***N***	**%**	***N***	**%**	***N***	**%**	x^2	p
2015									39.95	***
0–3 years	427	(86.6)	37	(7.5)	29	(5.9)	493	(100)		
4–8 years	140	(71.4)	15	(7.6)	41	(21)	196	(100)		
9 years and older	40	(87)	2	(4.3)	4	(8.7)	46	(100)		
2020									45.09	***
0–3 years	215	(71.4)	57	(19)	29	(9.6)	301	(100)		
4–8 years	33	(37.5)	14	(16)	41	(46.5)	88	(100)		
9 years and older	5	(33.3)	1	(6.7)	9	(60)	15	(100)		

Includes valid responses only, may not total sample.
*$p > 0.05$; **$p < 0.01$; ***$p < 0.001$.

TABLE 5 Differences between family member ceasing work and challenging behaviors.

	Ceased work (*N* = 1,176)		x^2	p
Number of challenging behaviors			22.14	***
0–1	25	(6.2)		
2–3	336	(83.6)		
More than 4	41	(10.2)		

Includes valid responses only, may not total sample.
*$p < 0.05$; **$p < 0.01$; ***$p < 0.001$.

Differences across samples were identified ($p < 0.001$) for behavior difficulties ($x^2(1) = 125.28$) and insistence on sameness ($x^2(1) = 45.98$). Frequency of caregiver reporting of behavior difficulties decreased from 61.8% ($n = 460$) in the 2015 sample to 27.7% ($n = 117$) in 2020, whereas insistence on sameness decreased from 63 ($n = 466$) to 42.4% ($n = 179$).

Age of diagnosis and type of diagnosis

When comparing age of diagnosis ($x^2(2) = 15.14$, $p = 0.002$) and type of diagnosis ($x^2(4) = 182.10$, $p < 0.001$) there was a statistical significance differences between both samples. For the 2015 sample, 34.1% of caregivers indicated their child having an ASD or autism diagnosis. This was closely followed by PDD-NOS diagnosis ($n = 257$, 33.7%). For age of diagnosis, most caregivers ($n = 500$, 66.4%) indicated their child was diagnosed before the age of three. Yet almost 30% ($n = 201$) were diagnosed between 4 and 8 years old. On the other hand, in the 2020 sample, autism or ASD diagnosis increased to 77.3% ($n = 305$), whereas PDD-NOS decreased to 14.2% ($n = 60$). Diagnosis before the age of 3 years old rose to 77.2% ($n = 325$), and diagnosis between ages 4 and 8 decreased to 18.3% ($n = 77$).

Age of diagnosis and individual who noticed first concern

There was a statistically significant difference ($x^2(2) = 47.32$, $p < 0.05$) between individual (family member, physician, teacher) who noticed first concerns about child's development and sample year. As exhibited in Table 3, family members are the most frequent individuals who notice developmental concerns in both samples. In contrast, teachers had second place in the 2015 sample (10.7%, $n = 81$), while physicians had it for the 2020 sample (18.6%, $n = 16.8$). A chi-square test of independence was also conducted to assess frequency differences in individual who noticed first concern and the age of diagnosis of the autistic child in each sample year. This analysis showed statistically significant differences between the person who noticed the first concern and the age of diagnosis for both the 2015 sample ($x^2(6) = 39.95$, $p < 0.001$) and 2020 one ($x^2(6) = 45.09$, $p < 0.001$). For the 2015 survey, almost 90% ($n = 427$) of participants who indicated a family member noticing first developmental concern had their child diagnosed on or before 3 years of age. Among those diagnosed before the age of three, only 7.5% had their first concern noticed by a physician ($n = 37$), and 5.9% by a teacher ($n = 29$). In the 2020 sample, 19% ($n = 57$) had a physician and 9.6% a teacher ($n = 57$) noticing the first developmental concern. Among those children diagnosed between the ages of 4 and 8, in the 2015 sample, 21% of teachers noticed first concern ($n = 41$), while in the 2020 sample, 46.5% had their teacher noticing ($n = 215$); showing an increase of teacher's awareness.

Service barriers and caregivers challenges

Caregiver reports of service barriers decreased in all categories. Experienced delays due to waitlist significantly decreased from 43.5% in 2015 to 16.4% in 2020 ($x^2(1) = 81.97$, $p < 0.001$), treatment cost from 33.5% to 12.6% ($x^2(1) = 60.99$,

TABLE 6 Service barriers, challenges, and priorities.

	2015 $N = 763$		2020 $N = 422$		x^2	p
Service barriers						
Not qualifying for services	131	(18.2)	39	(9.2)	16.76	***
Not available in their area	191	(26.5)	76	(18)	10.78	**0.001
Waiting list	245	(43.5)	69	(16.4)	81.97	***
Treatment costs	243	(33.5)	53	(12.6)	60.99	***
Limited Information	127	(17.9)	18	(4.3)	43.98	***
Other	122	(17.8)	57	(13.5)	3.5	***
Family member ceased working	257	(34.1)	145	(34.3)	0.57	
Challenges						
Challenging behaviors	276	(36.2)	133	(31.5)	2.61	
Daily living skills difficulties	260	(34.1)	166	(39.3)	3.23	
Health problems	48	(6.3)	23	(5.5)	0.34	
Sleep problems	76	(10)	95	(22.5)	34.67	***
Eating/feeding difficulties	109	(14.3)	122	(28.9)	37.03	***
Social interaction difficulties	413	(54.1)	224	(53.1)	0.12	
Restricted/repetitive behaviors	155	(20.3)	129	(30.6)	15.68	***
Communication difficulties	332	(43.5)	232	(55)	14.18	***
Impaired safety/notion of danger	157	(20.6)	58	(13.7)	8.54	0.003
Sensory	88	(11.5)	131	(31)	68.65	***
Priorities						
Receiving social support	285	(37.4)	105	(24.9)	19.14	***
Basic rights protected	480	(19.0)	266	(18.7)	0.02	
Better health services	363	(47.6)	113	(26.8)	48.91	***
Better education services	444	(58.2)	222	(52.6)	3.44	
More rights for autistic individuals	198	(26)	132	(31.3)	3.84	*
Improved implementation existing rights	169	(22.1)	187	(44.3)	63.51	***
More information about ASD	176	(23.1)	137	(32.5)	12.35	***
Home support	97	(12.7)	68	(16.1)	2.62	
Community awareness	310	(40.6)	194	(46)	3.17	

Includes valid responses only, may not total sample. Percentages are within sample year. Since caregivers could choose more than one option percentage total does not equal 100%. $^*p < 0.05$; $^{**}p < 0.01$; $^{***}p < 0.001$.

$p < 0.001$), limited information from 17.9 to 4.3% ($x^2(1) = 43.98$, $p < 0.001$), not qualifying for services from 18.2 to 9.2% ($x^2(1) = 16.76$, $p < 0.001$), and service not available in their area decreased from 26.5 to 18% ($x^2(1) = 10.78$, $p = 0.001$). When exploring the financial impact of caring for an autistic individual, caregivers' reports of a family member having to stop working to care for their child showed no statistical significance ($p > 0.05$) when comparing both samples, with more than 34% of caregivers indicating agreement ($x^2(2) = 0.57$, $p > 0.05$). There was a statistically significant difference between the amount of challenging behaviors and the number of those reporting that a family member had ceased working ($x^2(2) = 22.14$, $p < 0.05$), showing an increased rate of family member ceasing work as number of challenging behaviors increased (Table 5).

Concerning autistic characteristics that were challenging for caregivers, there were no statistically significant differences in social interaction difficulties which showed the highest rate in both samples ($x^2(1) = 0.12$, $p > 0.05$) with an almost 55% endorsement. Differences were identified, with an increased number of participants from the 2020 sample reporting greater concerns in several areas. Communication difficulties were endorsed at a significantly higher rate ($x^2(1) = 14.18$, $p < 0.001$) in 2020 (55%) than in 2015 (43.5%). Moreover, sensory issues ($x^2(1) = 68.65$, $p < 0.001$) saw an increase with 11.5% in 2015 vs. 31% in 2020, restricted and repetitive behaviors ($x^2(1) = 15.68$, $p < 0.001$) with 20.3% in 2015 and 30.6% in 2020, sleep problems ($x^2(1) = 34.67$, $p < 0.001$) with more than double the rate in 2020 (22.5%), and eating or feeding difficulties also with little more than double the percentage of caregivers within the sample (28.9%) indicating endorsement in 2020. Significant decreases in challenges were also identified with impaired sense of safety and notion of danger ($x^2(1) = 8.54$, $p = 0.003$) decreased from 20.6 to 13.7% (Table 6).

Caregiver's priorities

Priority being placed on better educational services continued to be the most frequently chosen item among caregivers, with more than 50% doing so; however, there was no statistically significant difference ($x^2(1) = 3.44$, $p = 0.003$) between the two groups. Significant differences among both samples were for caregivers wanting: more rights for autistic individuals ($x^2(1) = 3.84$, $p = 0.5$), improved implementation of existing rights ($x^2(1) = 63.51$, $p < 0.001$), and more information about ASD ($x^2(1) = 12.35$, $p < 0.001$). All of these had higher endorsement by caregivers in the 2020 sample (see Table 2). Whereas, rates of priorities being that autistic child receives social support ($x^2(1) = 19.14$, $p < 0.001$) and better health services ($x^2(1) = 48.91$, $p < 0.001$) decreased in the 2020 sample. On the contrary, basic rights being protected ($x^2(1) = 0.02$, $p > 0.05$), home support ($x^2(1) = 2.62$, $p > 0.05$), and community awareness ($x^2(1) = 3.17$, $p > 0.05$) showed no statistically significant difference between the two samples.

Discussion

Our first goal was to observe quantitative differences in the age of first concerns, individuals who first noticed developmental concerns, and changes in the age of diagnosis.

In our latest sample, more caregivers reported first concerns before their child was 24 months. In line with research from other countries (41), our results showed an increased caregivers' awareness in the 2020 sample, as evidenced by more caregivers reporting their child's first developmental concern before the age of 24 months. In addition, it highlights the importance of caregivers as an essential element for identifying ASD early signs (37). In our study, caregivers were the most likely to notice early concerns instead of physicians and teachers. This rate increase of earlier diagnosis noticed by caregivers may reflect a better understanding of ASD in Argentina which has been promoted by advocacy groups that have advocated for the enforcement of policies, laws, and adherence to the Convention of the Rights of Persons with Disabilities. This convention, which Argentina and other countries adhered to, promotes attitude changes to improve the quality of life of disabled individuals and decrease barriers toward the inclusion and protection of those with disabilities (14). Previous literature has indicated how better awareness could help with earlier recognition and stigma reduction (42). It is thus imperative to continue advocacy work and the implementation of strategies that promote increased community awareness and global health response toward an increased community capacity (1). This is particularly relevant when considering that a lack of ASD knowledge is associated with misconceptions and a deficit view of the condition, which conceptualizes ASD as an illness (43). Additionally, an increased attention and appropriate information could permit improved accessibility to better quality intervention and treatment services (34) and alleviation of symptom severity (33).

Regarding types of first observed concerns, lack of response to name and insistence on sameness remained consistently high in the latest sample. These two characteristics are part of the diagnostic criteria identified in the DSM-5 and ICD-10 (44, 45). Not responding to their name is encompassed in deficits in social-emotional reciprocity (44). On the other hand, caregivers from the 2020 sample were least likely to endorse behavior difficulties and medical and gross motor concerns as first developmental concerns. This could be explained by ASD diagnostic criteria since, despite aggression, medical conditions, and gross motor delays being commonly co-occurring conditions (46), they are not required to meet an ASD diagnosis (47, 48). Our results seem to imply an attunement in identifying ASD core characteristics. However, more than 20% of caregivers continue to report common co-occurring conditions as the first noticed developmental concerns. These results highlight the need for service providers and clinicians to hear caregivers' reports of co-occurring conditions due to their increased prevalence among autistic individuals compared to typically developing children (48). Also, as previously indicated, caregivers continue to notice developmental concerns earlier.

In both samples, of those children diagnosed before the age of three, most caregivers (>70%) had noticed the first developmental concern. In other words, when caregivers noticed a developmental difference in their children, those children were more likely to be diagnosed earlier. Yet, interestingly, in the 2020 sample, the frequency of children diagnosed before the age of three who had a physician or teacher noticing the first developmental concern increased. These results imply two things: caregivers seem to be heard by providers who diagnose their children before preschool age, and physicians and teachers seem to have more awareness and knowledge. This possible increased awareness might reflect the proactive work being conducted in Argentina, which has taken an active and continuous stance in promoting awareness and inclusion of those individuals with disabilities. For instance, non-profit organizations, such as RedEA, work toward greater community awareness and advocacy for quality-of-life improvement (19, 20, 49). Moreover, Argentina has established laws that protect individuals with disabilities, particularly autism. In 2014 the National Autism Law (previously mentioned as Act 27043) passed and was implemented legally by 2019. This law is complemented by the National Disability Law (20). These laws mandate better accessibility to diagnosis and healthcare and emphasize a comprehensive and interdisciplinary approach, the training of healthcare professionals, and increased research efforts (20). Therefore, changes in decreased age of first concerns, age of diagnosis, and the individual who noticed developmental differences might reflect changes in community awareness and increased knowledge brought upon by the work of non-profits, advocates, and the implementation of policies which focus on the protection of those with ASD. This information is relevant to other countries, which might still be behind in disability laws and advocacy work. For example, in Latin America, Argentina is the only country that has a legal framework focusing on those with ASD (14).

Our second goal was to identify challenges experienced by caregivers and address possible gaps in policies and practices concerning services, rights, and support. In the 2020 sample, fewer parents indicated accessibility issues when observing service barriers. Despite these results pointing to a positive trend in service provision in Argentina, a large percentage of families reported still facing barriers. One of those challenges that remained steadfast across both samples was family members continue quitting their work to care for an autistic individual. This is problematic when considering the high cost of caring for somebody with ASD (50). For families already struggling with financial challenges, eliminating one source of income could translate to an even more distressing situation, particularly for those whose children have more severe symptomatology (51, 52). Horlin et al. (51) indicated that 90% of the family cost related to ASD is due to loss of income. Additionally, increased cost was associated with the number of ASD symptoms (51). In our sample, reports of challenging characteristics such as social interaction, sensory issues, and restrictive, repetitive behaviors remained high or increased among caregivers. Also, in both samples, most caregivers reported their child having more than two challenging behaviors, among which more than 90% had a family member ceasing to work to care for their autistic child. It is thus expected that for these caregivers, the cost of caring for an autistic individual means added financial impact. The added cost of ASD and the loss of income for Argentinian caregivers could be exceptionally burdensome given the precarious economic affairs currently plaguing the country. From 2015 to 2020, many of the country's financial indicators have shown a negative trend, with its gross domestic product (GDP) decreasing by more than 200 billion dollars (7). To understand how significant this sum is, one can compare it to Argentina's current GDP of 389 billion (7). In 2015 1 in every 3 Argentinians was living below the poverty line, and this economic situation did not improve in the following years, resulting in austerity measures and a long-lasting recession lasting until this day (53). In our sample, the added number of problematic behaviors combined with family members ceasing to work in an environment struggling with recession and poverty implies more challenges requiring caregivers of autistic individuals to reorganize their priorities to meet their child's needs better (54).

Although there have been similarities and differences observed in the challenges expressed by caregivers, caregiver priorities have remained relatively consistent across both samples. Child education and receiving better educational services remained the highest endorsed priority. The second most endorsed priority was to improve the implementation of existing rights (increased endorsement in the 2020 sample) and increased community awareness (similar rates in both samples). Reporting of educational services as a continued priority is understandable. ASD cases in the mainstream school system have seen a 25-fold increase in recent years (55). The rise in cases meant an increased need for more school personnel that understands developmental concerns, and this need translated to the use of support teachers that form part of the specialized support in schools (55). Also, despite Argentina establishing laws outlining policies for the educational inclusion of children with ASD, how these policies and laws are operationalized has yet to be determined. Therefore, there is little accountability when it comes to enforcing such legislature, and concerns regarding the quality of education for ASD children have been raised (55). For example, teacher training programs do not provide up-to-date ASD information in curricular programs which could result in teachers not being adequately trained to implement inclusive practices in school settings (55). Based on our identified results, it is imperative to have more efficient teachers and support staff training on ASD and, include a more precise operationalization on the needs of an inclusive school system.

Although Argentina has a legal framework aiming to to protect the rights of those with ASD, there are certain limitations to its implementation. For example, some of those laws refer to disabled people as "people that suffer" from said disability, promoting stereotypes and prejudice. Such language across legal documents perpetuates the categorization of autistic individuals as handicapped and thus silencing the heterogeneity and neurodiversity within the spectrum (56). In addition, complex bureaucracy, extensive and unclear paperwork limit laws implementation. Argentina disability laws clearly outline policies to establish the rights of children with ASD, including educational inclusion and feasible access to appropriate diagnosis and treatment; however, there is not a mechanism explaining how these policies are operationalized (56). Currently, caregivers of autistic individuals and self-advocates have organized several activities to protest for which they consider a failure to comply with legally established laws resulting in multiple children not receiving an ASD diagnosis, which in turn delays access to timely treatment (57). Despite laws aiming at protecting the rights of those with disabilities in Argentina, many families do not know how to navigate a somewhat cumbersome system. For example, in Argentina, the previously mentioned *Certificado Único De Discapacidad* (Unique Certificate of Disability, UCD) is a public document that enables individuals with disabilities to exercise their rights and access social benefits as described by national laws (58). Yet only 14.6% of individuals with disabilities have access to the UCD (9). Families of individuals with disabilities confront an uncharted territory when trying to find diagnosis and treatment while navigating "bureaucratic obstacles, originating from its health system and society" [(9), p. 355]. These obstacles in addition to the limited knowledge of existing rights, increases worry, uncertainty, and exhaustion in families, which further hinders autistic individuals' full inclusion in society (9).

Community awareness continues to be reported by caregivers as a priority. It is important to mention that Argentina has made strides toward increased community awareness. As previously mentioned, non-profit organizations' main goal is to increase awareness and empower autistic individuals. Additionally, in 2015, an important ASD awareness campaign titled *Mirame* won the first prize for a recognized national competition (59). This campaign had a significant public impact and increased its website visitors, which included information on autism screening, early signs recognition, and available local services (5). Due to the continued endorsement of community awareness as a priority, future research could further explore caregiver input on increasing awareness.

Our study provides a window into the lives of Argentinian caregivers. Its ability to assess their experiences within a 5-year gap also allows for the observation of differences across time on multiple aspects such as first developmental concerns, age of diagnosis, challenges and priorities of caregivers of autistic individuals. In Argentina, GDP expenditure dedicated to research is only 0.49%, whereas in countries like the United States estimated expenditure is at 2.20% (7). Elsabbagh et al. (18) indicated that health research funding in multiple countries is limited with only 10% of the global healthcare expenditure going to 90% of the world's population (18). Yet, through the present study, researchers and specialists in Argentina have responded to the call made by previous publications to fill the ASD knowledge gap in other parts of the world outside the United States and Europe (18, 60).

Despite its contributions, the present study presents some limitations such as the lack of confirmatory diagnosis of the children in the study. Also, most respondents in both samples had some form of higher education and thus did not represent those with lower educational attainment. Lastly, there is limited background information to further make comparisons between both populations from which the two samples were drawn. As such, despite both samples being derived from similar channels to enhance similarities, one cannot discard baseline differences which could be contributing to the results presented. Yet, knowledge gathered from this study could help elucidate possible progress in Argentina in terms of age of diagnosis, caregiver increased awareness of developmental concerns, and type of concerns caregivers continue to notice in their autistic children. Through caregivers' challenges and perceived priorities, policy and lawmakers can gain insight into the work still needed to be done for better educational inclusion and implementation of protective rights.

Taken together, this study's results imply an improvement in the notice of developmental concerns, a decreased age of diagnosis, and an improvement in several services in caregivers of autistic individuals in Argentina. Nonetheless, many caregivers reported barriers or rights still needing to be protected or improved. Our findings help illustrate not only Argentina's reality in terms of their ASD experience but also help inform of the possible steps toward greater community engagement and implementation of changes in public policies and practice in other Latin American countries.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation upon request.

Ethics statement

The studies involving human participants were reviewed and approved by Universidad Catolica Argentina (UCA). The patients/participants provided their written informed consent to participate in this study.

Author contributions

MM, EB, SC, DV, and CM-N contributed to conception and design of the study. SC, DV, and ARa collected the data. MM organized the database and performed the statistical analysis. MM and EB wrote the first draft of the manuscript. CM-N wrote sections of the manuscript. All authors contributed to manuscript revision, read, and approved the submitted version.

Acknowledgments

We thank all caregivers who responded to the questionnaire's utilized in this study. Also, we express gratitude to PANAACEA and RedEA for distributing said surveys.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

References

1. Zeidan J, Fombonne E, Scorah J, Ibrahim A, Durkin MS, Saxena S, et al. Global prevalence of autism: a systematic review update. *Autism Res.* (2022) 15:778–90. doi: 10.1002/aur.2696

2. Center for Disease Control and Prevention. *Data and Statistics on Autism Spectrum Disorder*. Available online at: https://www.cdc.gov/ncbddd/autism/data.html (accessed March 18, 2022).

3. Baxter AJ, Brugha TS, Erskine HE, Scheurer RW, Vos T, Scott JG. The epidemiology and global burden of autism spectrum disorders. *Psychol Med.* (2015) 45:601–13. doi: 10.1017/S003329171400172X

4. Kohn R, Ali A, Puac-Polanco V, Figueroa C, López-Soto V, Morgan K, et al. Mental health in the Americas: an overview of the treatment gap. *Rev Panam Salud Publica.* (2018) 42:e165. doi: 10.26633/RPSP.2018.165

5. Paula CS, Cukier S, Cunha GR, Irarrazaval M, Montiel-Nava C, Garcia R, et al. Challenges, priorities, barriers to care, and stigma in families of people with autism: Similarities and differences among six Latin American countries. *Autism.* (2020) 24:2228–42. doi: 10.1177/1362361320940073

6. Shaw KA, McArthur D, Hughes MM, Bakian AV, Lee L, Pettygrove S, et al. Progress and disparities in early identification of autism spectrum disorder: autism and developmental disabilities monitoring network, 2002-2016. *J Am Acad Child Adolesc Psychiatry.* (2022) 61:905–14. doi: 10.1016/j.jaac.2021.11.019

7. The World Bank. *Research and Development Expenditure (% of GDP).* Available online at: https://data.worldbank.org/indicator/GB.XPD.RSDV.GD.ZS (accessed March 18, 2022).

8. Puga C, Pagotto V, Giunta D, Vicens J, Leist M, Vaucheret Paz E, et al. Prevalence and incidence of disability based on the unique certificate of disability at a teaching hospital in the metropolitan area of Buenos Aires. *Arch Argent Pediatr.* (2019) 117:183–7. doi: 10.5546/aap.2019.eng.18

9. Moya G. Rights of persons with conditions associated with disability: current legal framework. [Derechos de los pacientes con enfermedades que se asocien con discapacidad: marco legal actual]. *Arch Argent Pediatr.* (2016) 114:355–60. doi: 10.5546/aap.2016.eng.355

10. Rubin L, Geller R, Nodvin J, Marcus M, Howett M, Merrick J. Break the cycle of environmental health disparities in vulnerable children. *Rev Environ Health.* (2011) 26:135–7. doi: 10.1515/reveh.2011.019

11. Rubinstein A, Zerbino MC, Cejas C, López A. Making universal health care effective in argentina: a blueprint for reform. *Health Syst Reform.* (2018) 4:203–13. doi: 10.1080/23288604.2018.1477537

12. Brookman-Frazee LI, Drahota A, Stadnick N. Training community mental health therapists to deliver a package of evidence-based practice strategies for school-age children with autism spectrum disorders: a pilot study. *J Autism Dev Disord.* (2012) 42:1651–61. doi: 10.1007/s10803-011-1406-7

13. Carbone PS. Moving from research to practice in the primary care of children with autism spectrum disorders. *Acad Pediatr.* (2013) 13:390–9. doi: 10.1016/j.acap.2013.04.003

14. Montiel-Nava C, Cukier S, Garrido G, Valdez D, Paula C, García R, et al. Service encounters across the lifespan in individuals with autism spectrum disorders: results from a multisite study in Latin America. *Res Autism Spectr Disord.* (2020) 79:1–13. doi: 10.1016/j.rasd.2020.101670

15. de Leeuw A, Happé F, Hoekstra RA. A conceptual framework for understanding the cultural and contextual factors on autism across the globe. *Autism Res.* (2020) 13:1029–50. doi: 10.1002/aur.2276

16. Zuckerman KE, Sinche B, Mejia A, Cobian M, Becker T, Nicolaidis C. Latino parents' perspectives on barriers to autism diagnosis. *Acad Pediatr.* (2014) 14:301–8. doi: 10.1016/j.acap.2013.12.004

17. Montiel-Nava C, Chacín JA, González-Ávila Z. Age of diagnosis of autism spectrum disorder in Latino children: the case of Venezuelan children. *Autism.* (2017) 21:573–80. doi: 10.1177/1362361317701267

18. Elsabbagh M, Divan G, Koh YJ, Kim YS, Kauchali S, Marcín C, et al. Global prevalence of autism and other pervasive developmental disorders. *Autism Res.* (2012) 5:160–79. doi: 10.1002/aur.239

19. Red Espectro Autista. *Quiénes Somos*. Available online at: from http://redea.org.ar/quienes-somos/ (accessed March 20, 2022).

20. Lord C, Charman T, Havdahl A, Carbone P, Anagnostou E, Boyd B. The lancet commission on the future of care and clinical research in autism. *Lancet.* (2020) 399:271–334. doi: 10.1016/S0140-6736(21)01541-5

21. Bagaiolo LF, Mari JJ, Bordini D, Ribeiro TC, Martone MCC, Caetano SC, et al. Procedures and compliance of a video modeling applied behavior analysis intervention for Brazilian parents of children with autism spectrum disorders. *Autism.* (2017) 21:603–10. doi: 10.1177/1362361316677718

22. Bordini D, Lowenthal R, Gadelha A, Araujo Filho GM, Mari JJ, Paula CS. Impact of training in autism for primary care providers: a pilot study. *Revista Brasileira de Psiquiatria.* (2015) 37:63–6. doi: 10.1590/1516-4446-2014-1367

23. Backes B, Mônego BG, Bosa CA, Bandeira DR. Psychometric properties of assessment instruments for autism spectrum disorder: a systematic review of Brazilian studies. *J Bras Psiquiatr.* (2014) 63:154–64. doi: 10.1590/0047-2085000000020

24. Becker MM, Wágner MB, Bosa CA, Schmidt C, Longo D, Papaleo C, et al. Translation and validation of Autism Diagnostic Interview-Revised (ADI-R) for autism diagnosis in Brazil. *Arquivos de Neuro-Psiquiatria.* (2012) 70:185–90.doi: 10.1590/s0004-282x2012000300006

25. Coelho-Medeiros ME, Bronstein J, Aedo K, Pereira JA, Arraño V, Perez CA, et al. M-CHAT-R/F Validation as a screening tool for early detection in children with autism spectrum disorder. [Validación del M-CHAT-R/F como instrumento de tamizaje para detección precoz en niños con trastorno del espectro autista]. *Revista Chilena de Pediatria.* (2019) 90:492–9. doi: 10.32641/rchped.v90i5.703

26. Cuesta-Gómez JL, Andrea Manzone L, Posada-De-La-Paz M. Modified checklist for autism in toddlers cross-cultural adaptation for Argentina. *Int J Dev Disabil.* (2016) 62:117–23. doi: 10.1179/2047387715Y.0000000006

27. Galdino MP, Pegoraro LFL, Saad LO, Grodberg D, Celeri E. Evidence of validity of the autism mental status examination (AMSE) in a Brazilian sample. *J Autism Dev Disord.* (2020) 50:2320–5. doi: 10.1007/s10803-018-3530-0

28. Manzone L. Adaptación y validación del modified checklist for autism in toddler para población urbana argentina. *Psicodebate.* (2013) 13:79. doi: 10.18682/pd.v13i0.363

29. Paula CS, Cunha GR, Bordini D, Brunoni D, Moya AC, Bosa CA, et al. Identifying autism with a brief and low-cost screening instrument—OERA: construct validity, invariance testing, and agreement between judges. *J Autism Dev Disord.* (2018) 48:1780–91. doi: 10.1007/s10803-017-3440-6

30. Soto S, Linas K, Jacobstein D, Biel M, Migdal T, Anthony B, et al. Review of cultural adaptations of screening tools for autism spectrum disorders. *Autism.* (2015) 19:646–61. doi: 10.1177/1362361314541012

31. Stewart LA, Lee LC. Screening for autism spectrum disorder in low- and middle-income countries: A systematic review. *Autism.* (2017) 21:527–39.

32. Ayuso M. *Tras cinco años de espera se reglamentó la "ley de autismo": ¿qué cambios plantea*? Available online at: https://www.lanacion.com.ar/comunidad/tras-cinco-anos-espera-se-reglamento-ley-nid2308093/ (accessed March 18, 2022).

33. Fein D, Barton M, Dumont-Mathieu T. Optimizing outcome in autism spectrum disorders. *Policy Insight Behav Brain Sci.* (2017) 4:71–8. doi: 10.1177/2372732216685098

34. Zuckerman KE, Lindly OJ, Reyes NM, Chavez AE, Macias K, Smith KN. Disparities in diagnosis and treatment of autism in Latino and Non-Latino White families. *Pediatrics.* (2017) 139:1–10. doi: 10.1542/peds.2016-3010

35. Maenner MJ, Shaw KA, Baio J, Washington A, Patrick M, DiRienzo M, et al. *Prevalence of Autism Spectrum Disorder Among Children Aged 8 Years - Autism and Developmental Disabilities Monitoring Network, 11 Sites, United States, 2016.* Morbidity and mortality weekly report. Surveillance Summaries. Washington, DC (2002).

36. Magaña S, Lopez K, Aguinaga A, Morton H. Access to diagnosis and treatment services among Latino children with autism spectrum disorders. *J Intellect Dev Disabil.* (2013) 51:141–53.

37. Tanner A, Dounavi K. The emergence of autism symptoms prior to 18 months of age: a systematic literature review. *J Autism Dev Disord.* (2021) 51:973–93. doi: 10.1007/s10803-020-04618-w

38. Ribeiro SH, Paula CS, Bordini D, Mari JJ, Caetano SC. Barriers to early identification of autism in Brazil. *Revista Brasileira de Psiquiatria.* (2017) 39:352–4. doi: 10.1590/1516-4446-2016-2141

39. Daniels AM, Como A, Herguner S, Kostadinova K, Stosic J, Shih A. Autism in Southeast Europe: a survey of caregivers of children with autism spectrum disorders. *J Autism Dev Disord.* (2017) 47:2314–25. doi: 10.1007/s10803-017-3145-x

40. Montenegro, MC, Abdul-Chani M, Valdez, D, Rosoli A, Garrido G, Cukier S. Perceived stigma and barriers to accessing services: experience of caregivers of autistic children residing in Latin America. *Res Dev Disabil.* (2022) 120:104123. doi: 10.1016/j.ridd.2021.104123

41. Parmeggiani A, Corinaldesi A, Posar A. Early features of autism spectrum disorder: a cross-sectional study. *Ital J Pediatr.* (2019) 45:144. doi: 10.1186/s13052-019-0733-8

42. Alsehemi MA, Abousaadah MM, Sairafi RA, Jan MM. Public awareness of autism spectrum disorder. *Neurosciences.* (2017) 22:213–5. doi: 10.17712/nsj.2017.3.20160525

43. Papoudi D, Jørgensen C, Guldberg K, Meadan H. Perceptions, experiences, and needs of parents of culturally and linguistically diverse children with autism: a scoping review. *J Autism Dev Disord.* (2021) 8:195–212. doi: 10.1007/s40489-020-00210-1

44. American Psychiatric Association. *Diagnostic and Statistical Manual of Mental Disorders* (5th ed.) Arlington, VA: American Psychiatric Association (2013).

45. World Health Organization. *International Statistical Classification of Diseases and Related Health Problems* (10th ed.). Available online at: https://icd.who.int/browse10/2016/en.2016 (accessed March 18, 2022).

46. Fitzpatrick SE, Srivorakiat L, Wink LK, Pedapati EV, Erickson CA. Aggression in autism spectrum disorder: presentation and treatment options. *Neuropsychiatr Dis Treat.* (2016) 12:1525–38. doi: 10.2147/NDT.S84585

47. Hodges H, Fealko C, Soares N. Autism spectrum disorder: definition, epidemiology, causes, and clinical evaluation. *Transl Pediatr.* (2020) 9:S55–65. doi: 10.21037/tp.2019.09.09

48. Mohd Nordin A, Ismail J, Kamal Nor N. Motor development in children with autism spectrum disorder. *Front Pediatr.* (2012) 9:598276. doi: 10.3389/fped.2021.598276

49. Panaacea. *Construyamos una Sociedad más Amable y Respetuosa de la Diversidad*. Available online at: https://www.panaacea.org/?p=quienes (accessed March 18, 2022).

50. Pickard KE, Ingersoll BR. Quality versus quantity: the role of socioeconomic status on parent-reported service knowledge, service use, unmet service needs,

and barriers to service use. *Autism*. (2016) 20:106–15. doi: 10.1177/1362361315569745

51. Horlin C, Falkmer M, Parsons R, Albrecht MA, Falkmer T. The cost of autism spectrum disorders. *PLoS ONE*. (2014) 9:e106552. doi: 10.1371/journal.pone.0106552

52. Kogan MD, Strickland BB, Blumberg SJ, Singh GK, Perrin JM, van Dyck PC, et al. national profile of the health care experiences and family impact of autism spectrum disorder among children in the United States, 2005-2006. *Pediatrics*. (2008) 122:e1149–1158.

53. Congressional Research Service. *Argentina's Economic Crisis and Default*. (2020). Available online at: https://sgp.fas.org/crs/row/IF10991.pdf (accessed March 18, 2022).

54. Hoefman R, Payakachat N, van Exel J, Kuhlthau K, Kovacs E, Pyne J, et al. Caring for a child with autism spectrum disorder and parents' quality of life: application of the CarerQol. *J Autism Dev Disord*. (2014) 44:1933–45. doi: 10.1007/s10803-014-2066-1

55. Reichow BBA, Barton EE, Odom SL. *Handbook of Early Childhood Special Education*. Cham: Springer International Publishing AG (2016).

56. Goycolea WG. Políticas y tratamientos del autismo. *Revista de Psicología*. (2021) 20:130–49. doi: 10.24215/2422572Xe046

57. Telam. *Padres de niños con autismo reclamaron coberturas integrales de salud*. Available online at: https://www.telam.com.ar/notas/202202/583312-padres-ninos-autismo-burocracia-cumplimiento-ley.html (accessed March 18, 2022).

58. Gobierno.com.ar. *Cómo obtener el Certificado Único de Discapacidad (CUD)*. Available online at: https://www.argentina.gob.ar/servicio/como-obtener-el-certificado-unico-de-discapacidad-cud (accessed April 4, 2022).

59. RedEA. *Mirame*. Available online at: http://redea.org.ar/mirame/ (accessed March 20, 2022).

60. Durkin MS, Elsabbagh M, Barbaro J, Gladstone M, Happe F, Hoekstra RA, et al. Autism screening and diagnosis in low resource settings: challenges and opportunities to enhance research and services worldwide. *Autism Res*. (2015) 8:473–6. doi: 10.1002/aur.1575

frontiers | Frontiers in Psychiatry

TYPE Review
PUBLISHED 25 August 2022
DOI 10.3389/fpsyt.2022.915999

OPEN ACCESS

EDITED BY
Diana Schendel,
Drexel University, United States

REVIEWED BY
Valentina Riva,
Eugenio Medea (IRCCS), Italy
Fred Genesee,
McGill University, Canada

*CORRESPONDENCE
Myriam L. H. Beauchamp
myriam.beauchamp@mail.mcgill.ca;
mryiam.beauchamp@mcgill.ca

SPECIALTY SECTION
This article was submitted to Autism,
a section of the journal
Frontiers in Psychiatry

RECEIVED 08 April 2022
ACCEPTED 11 July 2022
PUBLISHED 25 August 2022

CITATION
Beauchamp MLH, Amorim K, Wunderlich SN, Lai J, Scorah J and Elsabbagh M (2022) Barriers to access and utilization of healthcare services for minority-language speakers with neurodevelopmental disorders: A scoping review.
Front. Psychiatry 13:915999.
doi: 10.3389/fpsyt.2022.915999

Barriers to access and utilization of healthcare services for minority-language speakers with neurodevelopmental disorders: A scoping review

Myriam L. H. Beauchamp[1]*, Kaela Amorim[1], Samantha N. Wunderlich[1], Jonathan Lai[2], Julie Scorah[1] and Mayada Elsabbagh[1]

[1]Montreal Neurological Institute, McGill University, Montreal, QC, Canada, [2]Autism Alliance of Canada and Institute of Health Policy, Management and Evaluation, Dalla Lana School of Public Health, University of Toronto, Toronto, ON, Canada

Introduction: Minority-language speakers in the general population face barriers to accessing healthcare services. This scoping review aims to examine the barriers to healthcare access for minority-language speakers who have a neurodevelopmental disorder. Our goal is to inform healthcare practitioners and policy makers thus improving healthcare services for this population.

Inclusion criteria: Information was collected from studies whose participants include individuals with a neurodevelopmental disorder (NDD) who are minority-language speakers, their family members, and healthcare professionals who work with them. We examined access to healthcare services across both medical and para-medical services.

Method: Searches were completed using several databases. We included all types of experimental, quasi-experimental, observational and descriptive studies, as well as studies using qualitative methodologies. Evidence selection and data extraction was completed by two independent reviewers and compared. Data extraction focused on the barriers to accessing and to utilizing healthcare for minority-language speakers with NDDs. The search process and ensuing results were fully reported using a diagram from the *Preferred Reporting Items for Systematic Reviews and Meta-analyses extension for scoping review*.

Results: Following the database search, a total of 28 articles met our final selection criteria and two articles were hand-picked based on our knowledge of the literature, for a total of 30 articles. These studies revealed that minority-language speakers with NDDs and their families experience several barriers to accessing and utilizing healthcare services. These barriers, identified at the Systems, Provider and Family Experience levels, have important consequences on children's outcomes and families' well-being.

Discussion: While our review outlined several barriers to access and utilization of healthcare services for minority-language speakers with NDDs and their families, our findings give rise to concrete solutions. These solutions have the potential to mitigate the identified barriers, including development and implementation of policies and guidelines that support minority-language speakers, practitioner training, availability of referral pathways to appropriate services, access to tools and other resources such as interpretation services, and partnership with caregivers. Further research needs to shift from describing barriers to examining the efficacy of the proposed solutions in mitigating and eliminating identified barriers, and ensuring equity in healthcare for minority-language speakers with NDDs.

KEYWORDS

healthcare access, bilingual, neurodevelopmental disorders, minority-language speakers, multilingual, healthcare disparities

Introduction

Worldwide, there are more bilingual speakers than monolingual speakers (1) and many of these individuals live in environments where their "mother tongue" is a minority language [i.e., a language not spoken by most people in a given environment; (2)]. These minority-language speakers often face barriers to access or utilization of healthcare services (3). This is true regardless of whether they are speakers of an official minority language or a non-official minority language (4, 5). A key barrier encountered by minority language speakers is discordance between them and their healthcare practitioner's preferred or known language, which in turn can lead to communication breakdowns that can have important consequences for patients' health and well-being (6). Other barriers include the use of ad hoc interpreters (7), a reduced number of available services (8, 9), and barriers linked to negative attitudes held by healthcare workers regarding minority-language speakers (7, 10). Additionally, in the context of pediatric healthcare, parents of children from minority-language families also face barriers. Specifically, parents with limited proficiency in the majority language are reported to have more difficulty accessing healthcare services for their child, face challenges in communicating with healthcare practitioners, and are also more likely to misunderstand their child's diagnosis and treatment plan (11).

Barriers facing minority language speakers are amplified in the presence of neurodevelopmental disorders (NDD). Such conditions appear at birth or early in life and have an impact on development across the lifespan (12). This class of conditions includes autism spectrum disorder, intellectual disability, communication disorders, learning disabilities and motor disorders (12). A recent report indicates that 17% of children have been diagnosed with an NDD (13). For these children and their families, barriers to equal access and utilization of healthcare services have been linked to social determinants such as ethnicity, race and immigration status. These children and their families experience important disparities in accessing diagnostic and intervention services (14). Ethnicity has been linked to delays in accessing diagnostic services (15), which then delays children's ability to access intervention services. It has also been linked to reduced number of services, higher levels of unmet needs, particularly with regards to intensive intervention services, respite care, and psychological services (15). Similarly, children from immigrant families tend to access autism services at a later age than non-immigrant children (16) and their parents often report feelings of isolation and loneliness (17). In addition, ethnically diverse families report having access to fewer sources of information, a lack of resource material, and fewer social supports (15, 18). Professionals also receive little or no training in cultural sensitivity, even when they frequently work with culturally diverse populations (18). Moreover, the lack of access to interpreters is also a barrier to offering healthcare services that are culturally sensitive and appropriate (18). Finally, limited proficiency in the majority language makes it much more challenging for immigrant families to navigate the service delivery systems (14).

While studies clearly show that social determinants such as immigration status, ethnicity and race are linked to disparities in services for individuals with NDDs and their families, relatively few studies have focused specifically on the link between being a minority-language speaker with an NDD and barriers to healthcare access and utilization. Indeed, much of the research on minority-language speakers with NDD has instead focused on whether children with NDDs should learn more than one language. Traditionally, the belief has been that for children who already have challenges acquiring one language, the presence of a second language would likely increase language delays or that it would somehow confuse the child (19). Such erroneous beliefs have led to parents from minority-language backgrounds to avoid the use of their minority language with their child, which can have negative consequences on child-parent communication and can limit language-learning opportunities [(20), see (19) for discussion]. Moreover, many healthcare practitioners appear to share these beliefs and recommend against bilingualism for these children (20). In contrast, research over the past 20 years indicates that children with various NDDs can become bilinguals and that they can develop language abilities similar to those of their monolingual peers with similar developmental profiles, in at least one of their languages. This is true for children on the autism spectrum (21, 22), with a developmental language disorder [DLD, formally Specific Language Impairment; (23)], Down's Syndrome (24), and other NDDs. However, bilinguals do not always have monolingual-like abilities in both of their languages. Indeed, bilinguals' abilities in each of their languages are seldom equal (1) and are influenced by several factors, most notably the amount of exposure that children receive to each of their languages (25–28). Therefore, in children who are exposed to more than one language, language abilities in one of their languages that differ from the monolingual norm could indicate a language disorder, but could equally indicate a lack of exposure to that language.

The aim of the current scoping review is to identify the barriers to accessing and utilizing healthcare services for individuals with NDDs who are minority-language speakers. Examining barriers specific to minority language speakers is important for several reasons. First, this subgroup is not always captured in the social determinants of race, ethnicity, or immigration status. Additionally, race, ethnicity, and immigration status can be linked to being a language minority but are not necessarily so. Thus, examining the influence of being a minority-language speaker as a stand-alone social determinant will shed light on barriers specific to this minority group.

Method

A preliminary search of Embase, Google Scholar and PsychINFO was completed on December 3rd, 2021, and there were no published scoping or systematic reviews on this subject. A search of the Open Science Framework (OSF) on December 6th, 2021, also revealed no registered ongoing studies or publications in preprint examining our research aim. Our research protocol was published on the OSF platform on March 1st, 2022, prior to data extraction commencing.

Types of sources

For this review, we considered all types of experimental and quasi-experimental study design types. We also included observational studies and descriptive studies. In addition, qualitative studies including phenomenological interviews were included. Reviews were excluded, as were conference posters and conference talks, and unpublished (gray) literature. Moreover, for the purposes of this review, no language or date restriction criteria was imposed.

Eligibility criteria

To be considered for this review, studies were required to meet the following criteria:

A) Population: Studies were considered if they include individuals with NDDs who were minority-language speakers of any age, or caregivers of people with NDDs who are minority-language speakers. Additionally, study participants were required to be defined as minority-language speakers, bilinguals, multilinguals or as having limited English proficiency. A full list of the population criteria can be found in Table 1.

B) Condition: Following the DSM-5 (12), we defined NDDs as follows: Autism Spectrum Disorder [ASD; we also included in this category Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS) and Asperger's since these terms would have been used in studies prior to changes in the DSM-5 published in 2013], Attention Deficit Disorder (with and without hyperactivity; ADD and ADHD), Intellectual Disability, Communication Disorder [including a Developmental Language Disorder, Speech Sounds Disorders, Social Communication (Pragmatic) Disorder], and Learning Disorders. A full list of the conditions can be found in Table 1. Additionally, we defined minority-language speakers as individuals who speak a language other than the majority language (i.e., the language spoken by most individuals in a given environment.) using the following terms: bilingual, multilingual, minority language, and limited English proficiency. We included the terms "bilingual" and "multilingual" since we consider that, in most cases minority-language speakers will be (or need to become) minimally bilingual.

C) Context: Any context in which formal healthcare services are delivered by doctors, nurses, speech-language pathologists (SLPs), psychologists, social workers, occupational therapists, physiotherapists, or early interventionists, including within medical and academic contexts, and within the home. In addition, we defined healthcare access to services as the ability to obtain services from any medical or paramedical professional including doctors, nurses, speech-language pathologists, psychologists, social workers, occupational therapists, physiotherapists. We also included the terms *therapists, intervention, interventionist, healthcare, health services* and *healthcare services.* Given that some healthcare services are offered within the school system, we included access to such services in schools by combining the above terms

TABLE 1 Inclusion and exclusion criteria for studies.

Category	Inclusion	Exclusion
Population	• Bilingual, minority-language speakers, minoritized-language speakers AND • Disorders (Neurodevelopmental, ASD, Asperger's, PDD-NOS, ADD, ADHD, Down's Syndrome, Global developmental delay, Intellectual Disability, Language Disorder, Language Impairment, Communication Disorder (Developmental Language Disorder, Speech Sounds Disorders, Social Communication (Pragmatic) Disorder), and Learning Disorders)	• Other NDDs beyond our inclusion criteria
Context	• Clinical services in healthcare or educational settings, e.g., medical doctors, SLPs, SLTs, psychologists, social workers, doctors, nurses, occupational therapists, physiotherapists, and administrators • Early educators (ABA therapists, psycho-educator etc.) • Parents and caregivers of individuals with NDDs • Individuals with NDDs	• Other school workers (E.g., teachers and educational assistants)
Types of evidence	• Original research	• Reviews • Conference abstracts or posters • Government documents • Dissertations • Other "gray" literature
Concept	• Actual or perceived barriers to healthcare or educational services, e.g., beliefs about second language access or ability in children with NDDs • Knowledge of bilingualism in individuals with NDDs • Access or utilization of healthcare for minoritized language speakers with NDD	

with the term *school*. A full list of the contexts can be found in Table 1.

Search strategy

Based on prior knowledge of the literature, and following a MeSH term search, we developed an initial list of terms to designate NDDs, minority-language speakers, as well as a list of contexts (such as healthcare) where barriers to accessing and utilizing healthcare services could occur. Next, we completed an initial search using PubMed to identify articles examining healthcare service access for minority-language speakers. We validated and adjusted our initial list by comparing our search terms to (a) the keywords from abstracts and titles, (b) the keywords from the indexing list and (c) the MeSH terms linked to 80 existing studies that dealt with our three themes (NDDs, minority-language speakers, healthcare access). Our final list of search terms can be found in Table 2. Searches were completed using the following databases: PubMed, Embase, PsychINFO, Scopus, and CINAHL.

Source of evidence selection

A pilot test of our inclusion/exclusion criteria was completed to validate our list of inclusion and exclusion criteria. Two reviewers (Amorim and Wunderlich) independently downloaded the first 100 articles from the PubMed database to the web application, Rayyan (29), and applied the inclusion/exclusion criteria to these articles. Discrepancies were discussed by the first author (Beauchamp) and the second and third authors (Amorim and Wunderlich). Adjustments to the list were not required.

Full database searches were completed by the two reviewers (Amorim and Wunderlich) on February 21, 2022, and articles were uploaded to Rayyan. After deleting all duplicates, 1,171 articles were retained. Next, titles and abstracts were independently screened against the inclusion/exclusion criteria. For articles that were rejected, reviewers indicated the reason for the rejection (i.e., different population, different concept, different context, or different evidence). In cases where the abstract or title did not permit us to ascertain whether the article met our criteria, the article in question underwent a full text review. The two reviewers' final lists of articles were compared. Disagreements regarding article selection were discussed amongst the first, second and third authors (Beauchamp, Amorim, and Wunderlich). A total of 28 articles met our selection criteria. In addition, two articles that had not been flagged by our search were added based on the first author's knowledge of the literature. The search process and ensuing results are fully reported in Figure 1 of this scoping review using a diagram from the *Preferred Reporting Items*

TABLE 2 Example of SCOPUS search terms.

Search line	Search string	Concept
1	TITLE-ABS-KEY("neurodevelopmental disorders" OR "neurod#v*" OR "NDD" OR "autism" OR "ASD" OR "Aspergers" OR "PDD-NOS" OR "pervasive developmental disorder-not otherwise specified" OR "ADHD" OR "attention deficit hyperactivity disorder" OR "GDD" OR "global developmental delay" OR "intellectual disabilit*" OR "intellectual disability" OR "LD" OR "language disorder*" OR "language impairment" OR "communication disorder*" OR "learning disorder")	Neurodevelopmental Disorder
2	TITLE-ABS-KEY("bilingual*" OR "minority language*" OR "limited English" OR "multilingual*" OR "minoritized language")	Language
3	TITLE-ABS-KEY("healthcare" OR "school*" OR "doctor*" OR "Social worker*" OR "Medical*" OR "Clinician*" OR "SLP" OR "Speech language patholo*" OR "Nurse*" OR "psycholog*" OR "Therap*" OR "Treat*" OR "Health service*" OR "Health care" OR "Intervent*" OR "Practitioner*" OR "Professional*" OR "parent*" OR "caregiver*" OR "clinic" OR "hospital" OR "ward" OR "individuals" OR "people" OR "adolescents" OR "children" OR "adults") AND ("health services accessibility" OR "healthcare disparities" OR "access" OR "Health Care Quality, Access, and Evaluation")	Healthcare
4	1 and 2 and 3	Combining concepts

for Systematic Reviews and Meta-analyses extension for scoping review [PRISMA-ScR; (31)].

Data extraction

Following protocol registration, the two reviewers (Amorim and Wunderlich) independently extracted data from the articles included in our scoping review using data extraction forms. The use of these data extraction forms ensured that data extraction was completed in the same way across the two reviewers. To ensure that our data extraction documents were optimal for this project, the reviewers completed a data extraction pilot: Prior to registering the study protocol, the reviewers independently reviewed the same five articles (one quantitative, three qualitative, and one mixed design) using the data extraction forms, and the data extracted from each article was compared. No changes were required following the data extraction pilot. For the full data extraction process, data were extracted by the second and third authors separately (Amorim and Wunderlich). For each article information about the participants, contexts, and concepts of each article, the methodology used and the findings as they relate to our research question were extracted. Next, the extracted data were compared. Discrepancies were discussed with the first author and resolved. Finally, the extracted data were combined into a single working document to facilitate writing.

Results

As Table 3 shows, a total of 30 articles were selected: 28 found *via* database search and two through hand search. These include quantitative ($n = 20$), qualitative ($n = 7$), and mixed-model ($n = 3$) studies. Studies were conducted in and across a variety of countries: Australia ($n = 3$), Belgium ($n = 1$), Bulgaria ($n = 1$), Canada ($n = 6$), Denmark ($n = 1$), Egypt ($n = 1$), France ($n = 1$), Germany ($n = 1$), Greece ($n = 1$), Iceland ($n = 1$), India ($n = 1$), Israel ($n = 1$), Malaysia ($n = 1$), Malta ($n = 1$), the Netherlands ($n = 3$), Republic of Ireland ($n = 1$), Singapore ($n = 1$), South Africa ($n = 2$), Sweden ($n = 3$), the United Kingdom ($n = 5$), and the United States of America ($n = 19$) and examined a wide range of issues linked to access and utilization of healthcare services for minority-language speakers with NDDs. These include practitioners' thoughts, feelings and confidence related to serving minority-language speakers with NDDs, their understanding of and adherence to clinical guidelines, and language or geographical concordance with minority-language speakers. Other studies included minority-language parents' and individuals' perceptions on accessing healthcare, factors that influence their ability to access and utilize healthcare, their thoughts, and their beliefs and choices regarding bilingualism as it relates to healthcare access. Participants included: (1) caregivers who were minority-language speakers and who had children diagnosed with an NDD, and (2) service practitioners (mostly doctors, nurses and SLPs) and administrators. Data included in these studies were collected through questionnaires ($n = 15$), interviews ($n = 7$), retrospective record review ($n = 5$), cross-sectional or mixed-mode survey ($n = 3$), focus group ($n = 2$), census data ($n = 1$), and mystery shopper experimental approach ($n = 1$).

Data analysis and presentation

Data included in the analysis reflects our research question. Based on the data extraction, we identified barriers to healthcare

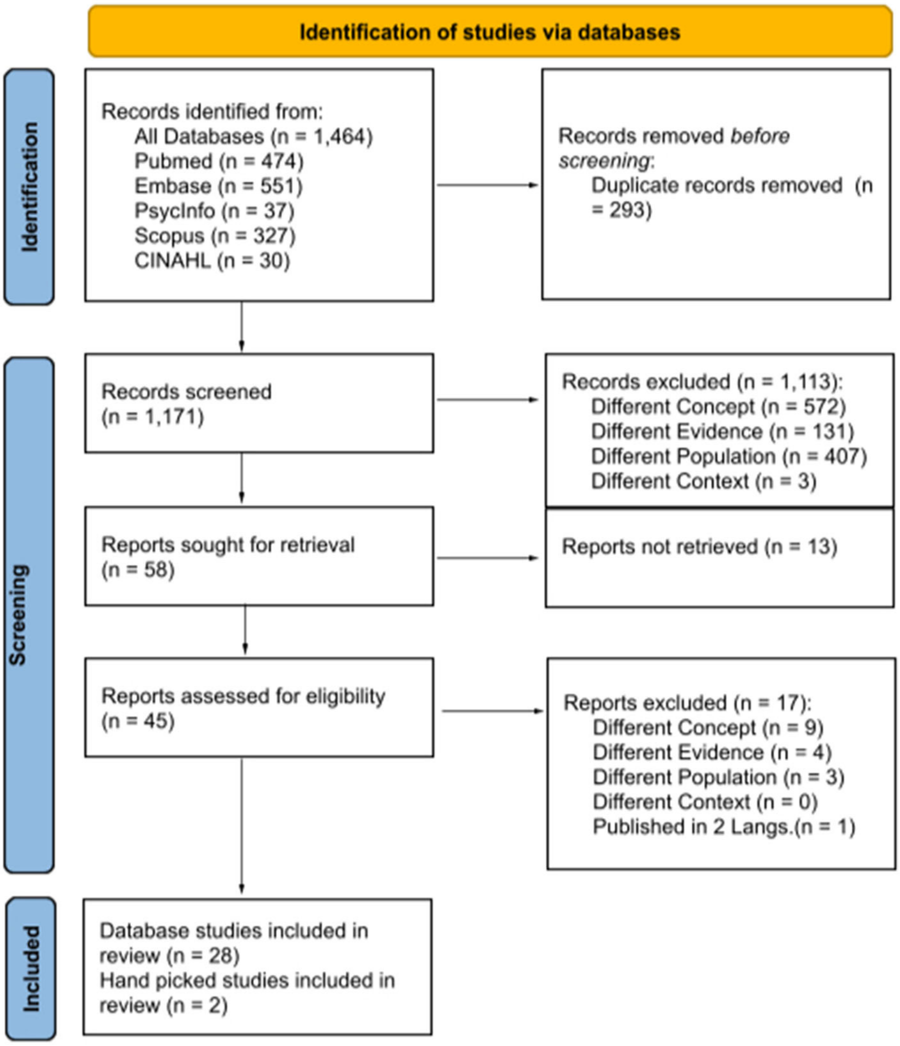


FIGURE 1
PRISMA 2020 flow diagram of included studies identified via database search and hand pick. From Page et al. (30) https://prisma-statement.org/PRISMAStatement/CitingAndUsingPRISMA.

access and utilization for individuals with an NDD and their families who are minority-language speakers. For each barrier we also identified by which participants it was reported (e.g., individuals, clinicians, parents), and the context (e.g., psychologists in schools).

The reader should note that throughout, we use the term "minority-language speakers" to refer to (a) individuals who speak only a minority language and who have received little to no exposure to the majority language and (b) to bilinguals since for them, one language is generally a minority language. We also use the term "bilingual" to refer to children who are exposed to two (or more) languages, even if they are not fluent in these languages or are in the process of acquiring them.

Barriers

As Table 4 shows, our review revealed several barriers to accessing and utilizing healthcare services. We classified them into three main domains: Systems, Practitioners, and Family Experience. In the following, we discuss barriers identified within each of the three domains and note the number of articles in which the target barrier was identified.

Systems

This domain refers to policies, procedures, or practices that tend to have a negative impact on access or utilization of healthcare services (36). Within this domain, five main themes

TABLE 3 Characteristics of included studies.

Authors	Study objectives	Participants	*N* of participants	Location	Data collection methods	Domain		
						System	Provider	Family experience
Bergeron et al. (32)	Understand culturally and linguistically diverse parents' perceptions of and practices around their child's language disorder	Parents born outside of Canada who have lived in Canada for less than 20 years	6	Canada	Phenomenological interviews	✓		
Bird et al. (33)	Investigate bilingualism in families with a child with ASD	Parents in a bilingual family who have one or more children with ASD	49	Canada, USA, Greece, France, Egypt, and Singapore	Questionnaire	✓	✓	
Caesar et al. (34)	Investigate the frequency SLPs used recommended practices when assessing bilingual students	Public school clinicians in Michigan	130	USA	Questionnaire	✓	✓	
de Valenzuela et al. (35)	Examine issues related to the inclusion and exclusion of students with developmental disabilities in and from bilingual opportunities	Policy makers, professionals, and practitioners in special needs and/or language education	79	Canada, USA, UK, the Netherlands	Semi-structured, one-on-one interviews	✓		
Fong et al. (36)	Document the barriers and facilitators Korean immigrant families encounter when accessing autism-related services	Korean parents of children with ASD	20	Canada	Individual semi-structured interview	✓	✓	✓
Hammer et al. (37)	Determine the level of training and confidence of SLPs in serving Spanish–English bilingual children	SLPs	213	USA	Questionnaire	✓		
Jimenez et al. (38)	Determine the national average wait time for developmental pediatric evaluations, and to understand differences in access	Developmental, neurodevelopmental, developmental-behavioral, or developmental disability clinics	90	USA	Mystery shopper study *via* phone calls	✓		
Jordaan (39)	Establish caseload characteristics, language profiles and proficiencies, and practices of SLPs regarding bilingual clients	SLPs providing intervention to bilingual children	99	Israel, Malta, Belgium, India, Canada, USA, UK, Sweden, Malaysia, Bulgaria, Denmark, Iceland, and South Africa	Questionnaire	✓	✓	

(Continued)

TABLE 3 (Continued)

Authors	Study objectives	Participants	*N* of participants	Location	Data collection methods	Domain		
						System	Provider	Family experience
Kritikos (40)	Determine SLPs' beliefs about language assessment of bilingual/bicultural individuals	SLPs	811	USA	Questionnaire	✓	✓	
Kuhn et al. (41)	Examine child and family factors that predict receiving a diagnostic evaluation and a confirmed ASD diagnosis	Medical Records of racial/ethnic minority children who screened positive for ASD	309	USA	Retrospective medical record review	✓		
Marinova-Todd et al. (42)	Gather information from professionals about their practices and opinions pertaining to the provision of bilingual supports to students with developmental disabilities	SLPs, teachers, language specialists, early childhood educator, administrators, and other professionals	361	Canada, USA, UK, and the Netherlands	Questionnaire	✓	✓	
Mcleod and Baker (43)	Describe practices regarding assessment and service delivery for children with speech sound disorders	SLPs	231	Australia	Questionnaire	✓	✓	
Mennen and Stansfield (44)	Identify the level to which SLP services meet recommendations; to examine caseloads; and to determine whether services are in place to meet the needs of those bilingual clients	SLPs and their managers	21	UK	Questionnaire and interviews	✓		
Mulgrew et al. (45)	Investigate the perceptions and practices of SLPs in the assessment of bilingual English–Irish-speaking children	Community-based practicing pediatric SLPs	181	Republic of Ireland and UK	Online cross-sectional survey	✓	✓	
Nayeb et al. (46)	Investigate nurses' perceptions of language screening and their practice for bilingual children	Nurses who perform language screening of bilingual children.	863	Sweden	Online questionnaire	✓	✓	
Pascoe et al. (47)	Investigate assessments and interventions used by SLPs in the Western Cape when working with children with speech difficulties	SLPs working with children	28	South Africa	Questionnaire	✓	✓	

(Continued)

TABLE 3 (Continued)

Authors	Study objectives	Participants	*N* of participants	Location	Data collection methods	Domain		
						System	Provider	Family experience
Rethfeldt (48)	Examine current provision of speech-language intervention services for multilingual children	SLP practices	28 SLP practices	Germany	Cross-sectional survey	✓	✓	✓
Rodrigues et al. (49)	Compare the receipt of developmental surveillance and screening among children	Medical records of children who attended wellness child visits	450	USA	Retrospective medical record review	✓	✓	
St Amant et al. (50)	Examine the influence of current ethnic and acculturation differences, with an emphasis on parental primary language, on child involvement in ASD-specific services	Medical records of children receiving services with a confirmed individualized education plan	152	USA	Retrospective medical record review	✓		
Vanegas (51)	Examine child, maternal, and family-level factors on the age of first autism spectrum disorder diagnosis among a diverse, clinical sample	Medical records of children with ASD	221	USA	Retrospective medical record review			
Verdon et al. (52)	Make a geographical comparison between multilingual children and multilingual SLP services in Australia	SLPs and children aged 4–5 years old	2849 SLPs; 4386 children	Australia	Questionnaire and census data	✓		
Wiefferink et al. (53)	Understand factors that may influence early identification by providing a detailed description of caseload characteristics	Medical records of children with language difficulties	9932	Netherlands	Retrospective medical record review	✓	✓	
Williams and McLeod (54)	Examine Australian SLPs' perspectives and experiences of multilingualism, including their assessment and intervention practices	SLPs working with multilingual children in Australia in 2010	128	Australia	Questionnaire	✓	✓	
Yu (20)	Explore the influences for and the effects of the language choices made by the mothers in relationship to their children with ASD	Parents who spoke Mandarin Chinese and had a child with ASD	10	USA	Phenomenological interviews	✓	✓	

(Continued)

TABLE 3 (Continued)

Authors	Study objectives	Participants	*N* of participants	Location	Data collection methods	Domain		
						System	Provider	Family experience
Yu and Hsia (55)	Examine the constraints and affordances of heritage language maintenance efforts in three families of children on the autism spectrum	Parents who have children with ASD and identify Chinese as a heritage language	3	USA	Interview	✓	✓	
Zuckerman et al. (56)	Develop and test a brief, English/Spanish bilingual parent-reported scale of perceived community ASD stigma	Parents of children with ASD aged 2 to 10 years old	380	USA	Questionnaire	✓		✓
Zuckerman et al. (57)	Compare barriers to ASD diagnosis and current ASD-related service use among families with English proficiency or limited English proficiency	Parents of children with ASD.	352	USA	Mixed-mode survey	✓		✓
Zuckerman et al. (58)	To assess ASD and developmental screening practices, attitudes toward ASD identification in Latino children, and barriers to ASD identification for Latino children	California pediatricians	267	USA	Questionnaire	✓	✓	
Zuckerman et al. (59)	To assess barriers to ASD diagnosis in the Latino community	Parents of children with ASD	33	USA	Focus group	✓		✓
Zuckerman et al. (60)	To qualitatively assess the potential barriers of ASD in the Latino community	Latino parents of typically developing children	30	USA	Focus group or individual semi-structured interview	✓		

TABLE 4 Thematic barriers by domain found in included studies.

Barriers	Domain			Citations
	System	Provider	Family experience	
1. Disconnect between the needs of users and services offered	✓			(20, 32–35, 38, 40, 42, 48, 52, 55)
2. Quality of treatment	✓			(34, 37, 40, 41, 46, 48–50, 53, 54, 56–58)
3. Lack of training for healthcare professionals	✓			(20, 33, 34, 37, 39, 40, 44, 46, 47, 54, 55)
4. Difficulties accessing interpreters	✓			(34, 37, 39–41, 43, 45, 48, 58–60)
5. Lack of available information in minority languages	✓			(32, 36, 48, 57, 58, 60)
6. Personal characteristics of healthcare practitioners		✓		(40, 58)
7. Practitioners' often erroneous beliefs regarding language development		✓		(20, 33, 39, 40, 46–48, 55)
8. Practitioner's lack of using evidence-based practices		✓		(34, 39, 42, 43, 45, 47, 48, 53, 54, 58)
9. Challenges in offering family-centered services		✓		(36, 39, 54)
10. Lack of resources		✓		(34, 39, 40, 45, 46, 49)
11. Feelings of distrust toward language discordant healthcare providers			✓	(36, 57, 59)
12. Feelings of stigma			✓	(48, 56)

emerged: (1) disconnect between the needs of users and services offered, (2) treatment quality, (3) lack or training for healthcare professionals, (4) challenges with interpretive services, and (5) lack of quality information in the minority language.

The first barrier within this domain is the *disconnect between the needs of users and services offered* (11 studies). This barrier is evidenced first and foremost by the lack of services offered in languages other than the majority language, and by the lack of interventionists who speak a language other than the majority language (20, 33, 34, 40, 52). Indeed, while caregivers report wanting services in their minority-language, they also report challenges in finding intervention support in that language (20, 32, 33, 55). What is striking is that the need for services in minority languages may not be recognized by those who work within the healthcare system. For example, in a study of SLPs in Germany, only 40% of practitioners considered that service options for minority-language speakers were inadequate (48), which seem to conflict with the challenges in finding services in the minority-language that parents experience. Sadly, challenges in finding services in the minority language have led some caregivers to make the difficult choice to only speak the majority language with their child with an NDD, believing that their child would otherwise "lose out" on receiving intervention (20). This barrier is also linked to findings that access to services for minority-language speakers with NDDs are seldom prioritized (35, 42) and that access to a language other than the majority language is viewed as being separate from, rather than integrated into, special needs services (35). What is particularly concerning is that frequently, programmes that offer services for children with NDDs, including those that are publicly funded, do not make accommodations for minority-language speakers despite regulations and guidelines that call for such accommodations [such as the Title VI of the Civil Rights Act and National Standard for Culturally and Linguistically Appropriate Services in the USA; (38)]. Such barriers lead to children with NDDs from minority-language families experiencing important challenges when trying to access and use healthcare.

Even when minority-language speakers with NDDs are able to access healthcare services (be it in the majority or the minority language), they often experience barriers with regards to the *quality of the treatment* that they receive (13 studies). Indeed, children from minority-language households are less likely than their majority-language peers to receive a developmental screener (41, 49, 58), or a referral for a language or developmental assessment from their pediatrician (46, 48, 49, 53). Consequently, children from minority-language households tend to receive an NDD diagnosis later than their majority-language peers [(48), although see (47) for contradicting findings]. Additionally, minority-language children may be offered fewer hours of intervention services (50, 57) when compared to majority-language speakers, and their intervention plans tend to include fewer social-skills goals (50, 56–58). Of note, these findings were maintained even after controlling for various demographic variables such as socio-economic status. Interestingly, language-based disparities may differ based on the social standing of the speaker's language. For example, in a study completed in Denver, Colorado, USA, Rodrigues and colleagues found that children who spoke neither English nor Spanish were less likely to receive developmental screening than Spanish-speaking or English-speaking children [while there was no difference between these two groups; (49)]. These disparities likely reflect differences in the social standing of the Spanish language in the study's geographic context compared to the standing of other minority languages. Indeed, according to the Census Bureau, Hispanics, and Latinos make-up ∼29% of the population in Denver (61), and of those, 57% report speaking Spanish at home (62). Because of Spanish's relatively high representation in the community, practitioners were able to access more materials and tools in Spanish compared to other minority languages, which limited the disparities between Spanish and English-speaking children (49). Taken together, minority-language speakers with NDDs face important challenges in access and utilization of services. However, other factors such a lack of professionals trained to work with minority-language children with NDDs (34, 37, 40, 46, 54) also create important barriers for these children.

While studies may suggest that it is preferable for minority-language speakers to work with interventionists who speak their minority language (37, 44), it is not always possible for them to do so. However, even when practitioners do not speak their client's minority language, they can work with them and support the development of this minority-language. Nonetheless, many interventionists express feeling unprepared to work with minority-language speakers with NDDs and their families (34, 37, 46, 54). This feeling is likely the result of the next barrier, the *lack of training for healthcare professionals to better support minority-language speakers* (11 studies). Practitioners frequently report receiving no instruction regarding the administration of language screening tools and the interpretation of ensuing results when assessing bilingual children, while the vast majority also report receiving such training to assess majority-language monolingual children (46). Although these findings may not be surprising when thinking about healthcare practitioners such as pediatricians and nurses, it is surprising to find similar trends for SLPs. Indeed, SLPs frequently report not receiving adequate training to work with minority-language families during their graduate studies (34, 37, 54) and beyond their university training (37, 54). For example, Pascoe and colleagues found that SLPs frequently rate their level of confidence to work with children from bilingual households as being rather low, even when they have had many years of experience (47). What is particularly troubling is that SLPs report a lack of mandatory training, even in contexts where their caseloads include several minority-language children (44). This lack of training for healthcare practitioners has led to challenges in correctly identifying language disorders in children from minority-language families (39, 40, 47). It has also resulted in caregivers receiving inconsistent or erroneous advice regarding whether they should raise their child in a bilingual context (20, 33, 39, 55). It is therefore not surprising that many families have made the heartbreaking choice to avoid speaking their minority-language with their child with an NDD which has important repercussions for language, communication, and psychosocial development [see (18) and (19) for discussion].

Beyond access to training, *difficulties in accessing interpreters* (11 studies) is also frequently cited as a barrier to accessing healthcare services (34, 39–41, 43, 48, 58–60). Caregivers report that interpreters are often unavailable and when they are available, it may only be by telephone, which caregivers regard as inappropriate and inadequate given the sensitivity of the subjects discussed with the practitioner (41, 60). The lack of interpreters hinders effective communication between caregivers and practitioners. Most notably, caregivers report that the lack of access to interpreters leads to challenges in scheduling appointments and in navigating the healthcare system (59). Caregivers also report that interpreters are often poorly trained (59, 60), which creates additional barriers to their ability to express their concerns regarding their child's development. Even when interpreters are professionally trained, they seldom have received training to support healthcare workers in their work [for example supporting SLPs during speech and language assessments (39, 45)]. Finally, practitioners who infrequently work with interpreters (trained or otherwise) may find it challenging to do so, particularly when they've not received training as to how to work effectively with interpreters (40). Unfortunately, findings suggest that few practitioners receive this type of training (37, 40).

In addition to the lack of interpreters, caregivers who are minority-language speakers also report a *lack of available information in minority languages* (six articles). Parents often have difficulty accessing printed information in their minority language about their child's disorder (60) and guidance or information regarding the various services and resources available to their family (36). And, even when materials are available, their quality is often poor (60). Moreover, because materials offered in the majority language often include medical jargon, they are difficult for minority-language caregivers to understand [particularly for those caregivers who have limited levels of proficiency in the majority language; (60)]. Consequently, caregivers who have limited proficiency in the majority language are less likely to seek out the information about their child's disorder and have more challenges understanding the information obtained (36) which leads them to have lower levels of knowledge about NDDs (32, 48, 57, 58, 60) than majority-language caregivers. This lack of information regarding child development and NDDs may lead to further delays in children with NDDs being identified (60).

Overall, the barriers identified in this domain reflect that sad truth that the needs of minority-language speakers with NDDs and their families are less likely to be met compared to those of majority-language peers and their families (56, 57). These unmet needs include the lack of intervention services or other care services, and the lack of adequate information (56, 57, 59). Unfortunately, the barriers within the System's domain are not the only barriers that minority-language families face in accessing and utilizing healthcare. They also face barriers from practitioners themselves.

Practitioners

The second domain refers to barriers related to the characteristics and beliefs of practitioners. Such barriers emerged in 17 studies and include: (1) characteristics of the practitioners, (2) practitioners' (erroneous) beliefs regarding language development in minority-language children with NDDs, (3) the lack of evidenced-based practices (4) challenges in offering family-centered services and (5) the lack of resources required to work with minority-language children with NDDs.

The first barrier within the Practitioners domain is defined as the *characteristics of the practitioners* (two articles). One such characteristic is the lack of linguistic diversity. In their study, Zuckerman and colleagues found that 56% of primary care practitioners who worked with minority-language speakers listed language discordance (i.e., practitioners speaking a language that is different from their patient's language) as a barrier to working effectively with their patients (58). A second characteristic identified as a barrier is a practitioner's caseload and the linguistic background of the other practitioners within their practice, which has an influence on their ability to work effectively with minority-language speakers. For example, pediatricians who have few minority-language patients (<25% of their caseload) are less likely to feel confident in identifying signs of autism in minority-language children (58). And those whose practice does not include colleagues who were minority-language speakers were less likely to offer developmental or autism screenings to children from minority-language backgrounds (58). Kritikos and colleagues also found that SLPs who identified as monolinguals reported feeling less proficient when working with an interpreter than bilingual SLPs (40). Thus, a practitioner's personal characteristics can influence the services rendered.

A second barrier to emerge was *practitioners' often erroneous beliefs regarding language development in minority-language in children with NDDs* (eight articles). Findings indicate that healthcare practitioners are often weary of recommending that children with NDDs be exposed to two languages, even when these children are from minority-language households (20, 33, 39, 55). This weariness seems to stem from the belief that in children with NDDs, particularly those that affect language and cognition, exposure to two languages will cause additional delay in language development (46). Thus, practitioners often recommend that families refrain from exposing children with NDDs to a second language. Given language constraints related to the language of schooling and of intervention, minority-language families are therefore generally told to avoid using their minority/heritage-language with their child in favor of the majority-language (20, 33). As previously discussed, this practice is not evidence-based and may even be detrimental to children's development [(19), also see (18) for discussion]. In addition, practitioners are also less likely to refer children from bilingual households for SLP services either because they are cautious of over-referring and/or due to their lack of knowledge regarding language development in minority-language children (40, 46, 47). Such practices could have important consequences for children's outcomes and families' well-being. The lack of understanding regarding language development in minority-language (either who are single or dual/multi-language learners) children also leads to challenges in assessing their language abilities. When conducting a language assessment with minority-language speakers, it is important to differentiate between children who have not yet completely acquired the majority language because they have not been sufficiently exposed to this language, from children who have a true language disorder (39, 40, 47). When practitioners do not have a solid understanding of bilingual development in children, they are more likely to identify a language disorder in children who are simply in the process of acquiring the language in which they were tested. For example, Rethfeldt (48) found that 57% of minority-language children who were diagnosed as having a DLD by a pediatrician were judged by SLPs as simply having a delay in the acquisition of the majority language rather than a true DLD, indicating overdiagnosis of DLDs on the part of pediatricians. In contrast, likely because of minority-language children's incomplete acquisition of the majority-language, practitioners may be less prone to identify the early signs of NDDs in children these children and therefore may not refer them to services in a timely manner (40, 46).

While false beliefs and a lack of understanding of language development in minority-language speakers likely stems from a lack of training, even when training is available practitioners do not consistently *use evidence-based practices*; another barrier emerging in 10 articles. For various reasons, too many practitioners are not using evidence to inform their practice when working with minority-language children with NDDs. For example, Pascoe et al. (47) report that even when best-practice guidelines are available, practitioners do not consistently use them to assess bilingual children. Other studies show that while practitioners may believe that they are using best practices, in reality, the methods they are using do not follow best-practices (42, 47, 54). Crucially, assessing minority-language children (who are either monolinguals or bilinguals) requires different strategies to those used when assessing majority-language children, as well as cultural competency (48) [see (63) for discussion on bilingual assessments]. Yet, many practitioners report relying solely on standardized measures in the majority language when assessing these children (34, 43, 45, 48, 54, 58) and many report not using an interpreter when assessing minority-language children (34, 39, 45, 48). While departures from best practices may be due to a lack of training, they may also be due to external factors like policies requiring standardized scores or time constraints (45). Whatever the underlying cause, such practices can lead to delays in diagnosis or to misdiagnoses (53).

The challenge of offering family-centered services is another barrier in the Provider domain (found in three studies). Indeed, despite recognition of its importance for minority-language speakers with NDDs (36, 54) practitioners can face difficulties adapting to the cultural expectations of each of their clients, being sensitive to diverging attitudes toward disabilities, and in reconciling their own beliefs with those of families [see (54) for an entire list of these challenges]. While these challenges may not be specific to minority-language services, they are likely common when working with minority-language families (36, 54). Practitioners' recommendation against bilingualism (39), despite families wanting to maintain their minority language (39) is also a challenge to family-centered practice, in addition to contravening best-practices. Consequently, community organizations are often called upon to address many cultural and language barriers that are typically ignored by traditional healthcare practitioners, by offering information in the minority language, emotional support, and guidance (36). The need for such services highlights the need for the integration of family-centered approaches within the healthcare system.

The final barrier linked to practitioners is the *lack of resources* (six studies) required to offer high quality services to minority-language speakers with NDDs and their families. This includes financial resources, physical resources (such as assessment tools), and time (34, 39). For example, studies note the prohibitive costs linked to hiring interpreters and practitioners who are trained to offer services to minority-language speakers (34, 39). Other studies note the lack of assessment and intervention tools in minority-languages (40, 45, 46, 49). Finally, while best-practices include working with trained interpreters (64), this practice can be more time consuming than working directly with a caregiver who speaks the majority language (49), as are bilingual language assessments (45). In certain contexts, practitioners may not be allowed this extra time as it may come at the expense of seeing other patients (34, 39).

Family experience

The last of the three domains refers to barriers linked directly to the family's experience accessing and using healthcare services. Two barriers emerged in the literature: (1) distrust toward healthcare practitioners who do not speak their minority language and (2) stigma.

Findings indicate that minority-language families often experience *feelings of distrust toward healthcare practitioners who do not speak their minority language* (three studies). Such distrust is the result of the inappropriate ways in which practitioners interact with caregivers from minority-language backgrounds. First, caregivers report encountering practitioners who are unable to provide information in a way that is culturally and linguistically appropriate (36, 57, 59). Caregivers also report experiencing discrimination from practitioners (36), which likely arises from a lack of cultural sensitivity. Finally, caregivers' distrust of practitioners who do not speak their minority-language may be a result of the frustrations that they experience during the diagnostic process, which in turn leads to doubts as to whether the provider was acting in families' best interests (59). Unfortunately, these feelings may lead to parents delaying acting on the recommendations of practitioners which they deemed less trustworthy (59).

Minority-language speakers with NDDs and their families also experience *stigma* when utilizing healthcare services (in two articles). Indeed, families with limited proficiency in the majority language have reported significantly higher rates of stigmatization compared to individuals who were visible minorities but majority-language speakers (56). The experience of stigma persisted after controlling for socio-economic status and was associated with unmet needs in treatment services (56). Unfortunately, the experience of stigma appears rampant in some communities; 77% of clinicians described their minority-language clients as being at a high risk of stigmatization (48).

Discussion

This scoping review examined barriers to the access and utilization of healthcare services for individuals with NDDs who are minority-language speakers and their families. Findings from the studies reviewed show that individuals in this minority group face numerous barriers in accessing and utilizing healthcare services. Overall, we identified 12 different barriers linked to Systems, Practitioners, and Family Experience, summarized in Table 4.

For the most part, our findings examining barriers faced by minority-language speakers with NDDs converge with those found in studies examining minority-language speakers' access and utilization of healthcare more broadly. Common to both groups is the language discordance between practitioners and patients, which leads to communication breakdowns and increased patient stress (6, 58). Moreover, limited availability of services in minority-languages also leads to lower quality of care and longer wait times (8, 9, 20, 33, 34, 40, 52). Minority-language speakers also consistently report discrimination and negative attitudes on the part of healthcare practitioners (7, 10, 36) and challenges linked to access to qualified interpreters (7, 34, 36). However, our findings highlight that barriers facing minority-language speakers, are also amplified for those with NDDs and their families. Key barriers for this population include practitioners' erroneous beliefs about language development in minority-language children with NDDs, and the lack of training for healthcare professionals to better assess and support the speech and language of individuals in this subgroup.

Evidence shows that erroneous beliefs regarding bilingual language development in children with NDDs leads to several negative consequences for minority-language speakers. First, practitioners often recommend against exposure of these children to their native languages or to the languages used by their family (20, 33, 39, 55). Second, practitioner may delay referrals to developmental or language assessments, or may not correctly identify a child as having a language disorder because they assume in children's language development will be due bilingualism (46, 48, 49, 53). Consequently, minority-language speakers are at increased risk for both delays in receiving an NDD diagnosis and accessing appropriate intervention. Conversely, minority-language speakers are also at risk of being misdiagnosed as having speech and language disorders when they are in fact in the process of acquiring the majority-language.

It is striking that erroneous beliefs about bilingual development persist despite a wealth of evidence highlighting that bilingualism does not cause language delays, that minority-language speakers with NDDs can become bilinguals and that for all children, but that these children's abilities in each of their languages will be influenced by several factors such as the amount of language exposure that they receive in each language (25, 65). Furthermore, in addition to interfering with quality of care, recommendations against these children being exposed to their native language also reduce resilience in subgroups like immigrants and refugees by leading to further loss of kinship and community tradition (66). The persistence of these erroneous practices, despite the seriousness of their consequences likely reflects persistence in biases and/or a lack of training opportunities regarding language development in minority-language children. The fact that many studies in this scoping review focused on autism specifically may suggest that some of the biases faced by minority-language speakers may be greater for individuals with autism from minority families than for other families with NDDs, although the extent to which this is be true is beyond the scope of this review.

Although our review focused on identifying barriers to access and utilization of healthcare services faced by minority language speakers, key solutions that can mitigate these barriers and level the playing field for minority language speakers are equally evident. Next, we present five key areas emerging from our review, where improvements are needed and can be feasibly implemented. Specifically: the development and uptake of policies and guidelines, practitioner training, referral pathways for specialized services, access to appropriate tools and resources, and partnership with caregivers.

First, there is a need for healthcare systems to develop and implement *policies and guidelines that support minority-language speakers*. One proposed policy is to increase the number of bilingual practitioners within the healthcare system by ensuring the inclusion of culturally and linguistically diverse practitioners (34, 41, 57). Where possible, increasing language concordance between users and practitioners may decrease feelings of mistrust toward practitioners who do not speak their minority language (57). Increasing concordance can also improve access to information and resources for minority-language speakers (36, 57) and therefore increase their knowledge about their child's disorder.

Overall, there is also a need for policies that ensure the adoption of evidence and best-practices through the development of evidence-based clinical guidelines where they do not exist, and measures to ensure that practitioners are following existing guidelines (34, 37, 38, 44, 58). New or existing guidelines can also benefit from adopting standards in how bilingualism is defined and how equity in service delivery is achieved for minority-language speakers (45). It is also critical that policies do not contradict best-practices. For example, practitioners should not be required to use specific standardized tests or to provide standardized scores to enable families to access services, financial supports, or reimbursement for services when the use of such measures is counter indicated. Rather qualified practitioners should be authorized to provide other types of information to describe children's language abilities (45).

Second, practitioners who work with minority-language speakers must *receive appropriate mandatory training* (34, 37, 40, 44–46, 48, 53, 54, 60). Given the consequences of practitioners' lack of training discussed above (e.g., delays in services, misdiagnosis), training is key to improving care for minority-language speakers with NDDs and their families. Indeed, findings show that many practitioners feel insufficiently trained to work with minority-language speakers (34, 46, 54) and less competent when they work with this population (37, 46, 47). Other findings indicate a lack of awareness on the part of practitioners regarding their need for training in this area (37). Therefore, mandatory training (as opposed to voluntary training) is likely the best way to counter the Dunning–Kruger effect [i.e., the effect which suggests that individuals with low levels of knowledge overestimate their abilities (67)].

Universities have a key role in preparing future healthcare professionals to work with minority-language speakers, including those with NDDs. To address the shortage of practitioners who speak a minority language, universities can ensure the inclusion of minority-language students in their training programmes (39, 54) and facilitate language learning opportunities for all their students (34). University programs can also enhance their core curriculum to include up to date evidence on bilingual language development in typically developing children and in children with NDDs, as well as cultural competency, among other areas of knowledge and skills relevant for practice with minority-language speakers.

Employers should also ensure that their employees are adequately trained to work with minority-language speakers (with and without NDDs). Innovative approaches to training can improve practitioners' awareness of the challenges of working with minority-language speakers with NDDs and mitigate biases especially during assessments and when working with interpreters (34, 36, 39, 52, 54). Training should also help practitioners develop cultural competency, especially since it is not always possible for practitioners to be fluent in all of the languages they will encounter (52). Finally, training should be appropriate to the practitioners' responsibilities. Thus, some practitioners, like SLPs, may require more specialized training to work with minority-language speakers (for example, having a strong understanding bilingual language develop and of best-practices when assessing minority-language speakers' language abilities).

In sum, training is key to increasing practitioners' understanding of bilingual language development, diminish false-beliefs and ensuring that practitioners are adequately trained given their responsibilities (34, 37, 45, 46, 52). It also decreases the Dunning–Kruger effect, and thus it increases the likelihood that children from minority-language families are adequately referred to practitioners who specialize in assessing and working with minority-language children with NDDs. Training also may go a long way to help diminish stigma by enhancing trust and family-centered care (36, 41, 48, 56, 57, 59).

The third area of improvement is ensuring that practitioner *refer minority-language speakers to specialized practitioner when there is any doubt regarding children's language or general development.* For example, minority-language children with (or suspected of) speech or language disorders, should be referred to an SLP who specializes in working with this population. By referring children to the appropriate services and to professionals who are experienced and knowledgeable in working with minority-language children with NDDs, these children are less likely to be misdiagnosed and are more likely to receive the types of services that they genuinely require (46, 48). It also ensures that minority-language children do not experience additional delays in accessing assessment and intervention services, and that intervention goals, particularly those in the areas of social skills and language, reflect their needs and are adequately targeted. Thus, to facilitate appropriate referral practices, practitioners are strongly encouraged to work collaboratively in multidisciplinary frameworks (48).

The fourth area of improvement is to ensure that practitioners have access to the necessary *tools and resources necessary to enable them to follow best practices learned during training and outlined in guidelines.* This includes access to trained interpreters who can play a variety of roles including facilitating access to services and other community resources, and helping families to communicate with practitioners (36, 45). However, it is important that interpreters be adequately trained [i.e., have knowledge in working with medical professionals, and have knowledge in NDDs (34, 36)] in order to effectively support both families and healthcare professionals. Beyond the access to trained interpreters, there is a need for screening and assessment tools in multiple languages (45, 47, 49). Recent studies across the globe have highlighted the importance of developing standardized tools to support accurate identification of language disorders and other NDDs, while simultaneously favoring culturally sensitive and contextually appropriate use of existing tools, even when those were not normed or developed for a specific language group (68). Such practices, relevant for all NDDs, include working with interpreters during assessments, obtaining information about the client's culture and language exposure, collecting information about the child's productions in their first language, and the use of dynamic testing (39, 42, 45–48, 54). Additionally, while there is a significant amount of available information regarding language development for some languages like English, there are fewer published norms for other languages (47). Therefore, researchers are called upon to develop language norms for these languages and to ensure that these norms are readily available in several languages (rather than only in the target language). Use of such practices and an increase in resource's is likely to reduce the risks of over- or under-diagnosing a language disorder in minority-language children. Employers must also ensure that practitioners are given the time to implement best-practices when working with minority-language individuals with NDD.

Finally, caregivers are a wonderful resource who need to be viewed by healthcare practitioners as *partners*. One important way to increase the partnership between caregivers and practitioners is for healthcare services to engage in stakeholder engagement activities to better identify the barriers in their context and ways to mediate these barriers. Stakeholders should include individuals with NDDs who are minority-language speakers and their caregivers, as well as practitioners and other frontline workers, and administrators. By engaging the different stakeholders, healthcare systems will better understand the situation in their individual context and find solutions that best suit the needs of their stakeholders. Effectively working with caregivers also includes supporting them to work with their children (54). This can include offering caregiver training and using parents as partners in the intervention process, by directly involving them in their child's intervention (54). These strategies increase parents' empowerment through increased understanding of their child's condition and improved access to services and resources. They also help minority-language caregivers implement intervention strategies at home in their minority language.

Taken together, the areas of improvement we proposed would enhance person- and family-centered care, and lead to improvements in the quality and efficiency of healthcare services. More importantly, implementing such solutions will begin to address the clear inequity facing minority language speakers. Future research is also needed to examine the efficacy of the proposed solutions in mitigating barriers in a various regions of the world and in different healthcare contexts. Differences in populations and systems might favor some solutions over others. For example, what are key differences in healthcare services that minority-language children with NDDs receive based on whether their minority-language is an official or unofficial language? New and unique solutions may be required for minority-language speakers who also face significant stigma and marginalization and/or those who are survivors of trauma. Furthermore, our review has highlighted a striking lack of studies with first person perspectives of people with NDDs themselves. Future research can build on recent advocacy and improved methods for capturing these perspectives (69, 70) in order to build a more accurate picture of the lived experience of minority language speakers in healthcare.

Author contributions

MB developed the research question, protocol, wrote the protocol for registration, and wrote the manuscript. MB, KA, and SW developed search terms, inclusion/exclusion criteria, data extraction forms and responsible for major manuscript revisions. KA and SW screened articles and completed data extraction. MB, KA, SW, JL, JS, and ME provided feedback for the protocol and revisions for the protocol and manuscript. All authors contributed to the article and approved the submitted version.

Funding

This project is funded by the following grant: Health Canada, and supported by Dialogue McGill at McGill University, under the Action Plan for Official Languages— 2018–2023: Investing in Our Future, a grant from the Fonds de Research du Québec en Sciences (#34734), and by the *Toronto Dominion Bank Post-Doctoral Fellowship in Child Health Research Excellence.*

Acknowledgments

The authors would like to thank Amy Bergeron, librarian at the McGill University Health Centre Library for her advice and support in preparing this review. We would also like to thank Jovia Wong for her help in reviewing some of the background literature.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

The reviewer FG declared a shared parent affiliation with the authors MB, KA, SW, JS, and ME to the handling editor at the time of review.

Publisher's note

References

1. Grosjean F. *Life With Two Languages: An Introduction to Bilingualism*. Cambridge, MA: Harvard University Press (1984), p. 384.

2. Paradis J, Genesee F, Crago MB. *Dual Language Development and Disorders*, 2nd ed. Baltimore, MD: Brookes Pub (2021), p. 296.

3. Bouchard L, Desmeules M. Linguistic minorities in Canada and health. *Health Policy*. (2013) 9(Spec Issue):38–47. doi: 10.12927/hcpol.2013.23589

4. Brisset C, Leanza Y, Rosenberg E, Vissandjée B, Kirmayer LJ, Muckle G, et al. Language barriers in mental health care: a survey of primary care practitioners. *J Immigr Minor Health*. (2014) 16:1238–46. doi: 10.1007/s10903-013-9971-9

5. Falconer J, Quesnel-Vallée A. Les disparités d'accès aux soins de santé parmi la minorité de langue officielle au Québec. *Rech Sociogr*. (2014) 55:511–29. doi: 10.7202/1028377ar

6. Meuter RF, Gallois C, Segalowitz NS, Ryder AG, Hocking J. Overcoming language barriers in healthcare: a protocol for investigating safe and effective communication when patients or clinicians use a second language. *BMC Health Serv Res*. (2015) 15:371. doi: 10.1186/s12913-015-1024-8

7. Free C, Green J. Bhavnani V, Newman A. Bilingual young people's experiences of interpreting in primary care: a qualitative study. *Br J Gen Pract*. (2003) 53:530–5.

8. Djiadeu P, Yusuf A, Ongolo-Zogo C, Nguemo J, Odhiambo AJ, Mukandoli C, et al. Barriers in accessing HIV care for Francophone African, Caribbean and Black people living with HIV in Canada: a scoping review. *BMJ Open*. (2020) 10:e036885. doi: 10.1136/bmjopen-2020-036885

9. Seiber EE, Smith CM, Tanenbaum SJ. Who treats limited English proficient patients? Implications for linguistic access initiatives. *Ethn Dis*. (2009) 19: 433–8.

10. Blignault I, Ponzio V, Rong Y, Eisenbruch M. A qualitative study of barriers to mental health services utilisation among migrants from mainland China in south-east Sydney. *Int J Soc Psychiatry*. (2008) 54:180–90. doi: 10.1177/0020764007085872

11. Flores G, Abreu M, Tomany-Korman SC. Limited English proficiency,primary language at home, and disparities in children's health care: how language barriers are measured matters. *Public Health Rep*. (2005) 120:418–30. doi: 10.1177/003335490512000409

12. American Psychiatric Association. *Diagnostic and Statistical Manual of Mental Disorders* (DSM−5). Washington, DC: Author (2013). doi: 10.1176/appi.books.9780890425596

13. Zablotsky B, Black LI, Maenner MJ, Schieve LA, Danielson ML, Bitsko RH, et al. Prevalence and trends of developmental disabilities among children in the United States: 2009-2017. *Pediatrics*. (2019) 144:e20190811. doi: 10.1542/peds.2019-0811

14. Singh JS, Bunyak G. Autism disparities: a systematic review and meta-ethnography of qualitative research. *Qual Health Res*. (2019) 29:796–808. doi: 10.1177/1049732318808245

15. Magaña S, Lopez K, Aguinaga A, Morton H. Access to diagnosis and treatment services among Latino children with autism spectrum disorders. *Intellect Dev Disabil.* (2013) 51:141–53. doi: 10.1352/1934-9556-51.3.141

16. Khanlou N, Haque N, Mustafa N, Vazquez LM, Mantini A, Weiss J. Access barriers to services by immigrant mothers of children with Autism in Canada. *Int J Ment Health Addict.* (2017) 15:239–59. doi: 10.1007/s11469-017-9732-4

17. Rivard M. Millau, M, Magnan C, Mello C, Boulé M. Snakes and ladders: barriers and facilitators experienced by immigrant families when accessing an autism spectrum disorder diagnosis. *J Dev Phys Disabil.* (2019) 31:519–39. doi: 10.1007/s10882-018-9653-6

18. Gardiner E, French C. The relevance of cultural sensitivity in early intervention. *Except Educ Int.* (2011) 21:34–49. doi: 10.5206/eei.v21i3.7681

19. Beauchamp ML, MacLeod AA. Bilingualism in children with autism spectrum disorder: Making evidence-based recommendations. *Can Psychol.* (2017) 58:250–62. doi: 10.1037/cap0000122

20. Yu B. Issues in bilingualism and heritage language maintenance: perspectives of minority-language mothers of children with autism spectrum disorders. *Am J Speech Lang Pathol.* (2013) 22:10–24. doi: 10.1044/1058-0360(2012/10-0078)

21. Beauchamp ML, Rezzonico S, MacLeod AA. Bilingualism in school-aged children with ASD: a pilot study. *J Autism Dev Disord.* (2020) 50:4433–48. doi: 10.1007/s10803-020-04501-8

22. Ohashi JK, Mirenda P, Marinova-Todd S, Hambly C, Fombonne E, Szatmari P, et al. Comparing early language development in monolingual-and bilingual-exposed young children with autism spectrum disorders. *Res Autism Spectr Disord.* (2012) 6:890–7. doi: 10.1016/j.rasd.2011.12.002

23. Paradis J, Crago M, Genesee F, Rice M. French-English bilingual children with SLI: how do they compare with their monolingual peers? *J Speech Lang Hear Res.* (2003) 46:113–27. doi: 10.1044/1092-4388(2003/009)

24. Bird EK, Cleave P, Trudeau N, Thordardottir E, Sutton A, Thorpe A. The language abilities of bilingual children with Down syndrome. *Am J Speech Lang Pathol.* (2005) 14:187–99. doi: 10.1044/1058-0360(2005/019)

25. Pearson BZ. Social factors in childhood bilingualism in the United States. *Appl Psycholinguist*. (2007) 28:399-410. doi: 10.1017/S014271640707021X

26. Thordardottir E. The relationship between bilingual exposure and vocabulary development. *Int J Biling.* (2011) 15:426–45. doi: 10.1177/1367006911403202

27. Thordardottir E. The relationship between bilingual exposure and morphosyntactic development. *Int J Speech Lang Pathol.* (2015) 17:97–114. doi: 10.3109/17549507.2014.923509

28. Thordardottir E. Amount trumps timing in bilingual vocabulary acquisition: effects of input in simultaneous and sequential school-age bilinguals. *Int J Biling.* (2017) 23:236–55. doi: 10.1177/1367006917722418

29. Ouzzani M, Hammady H, Fedorowicz Z, Elmagarmid A. Rayyan — a web and mobile app for systematic reviews. *Syst Rev.* (2016) 5:210. doi: 10.1186/s13643-016-0384-4

30. Page MJ, McKenzie JE, Bossuyt PM, Boutron I, Hoffmann TC, Mulrow CD, et al. The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. *BMJ.* (2021) 29;372:n71. doi: 10.1136/bmj.n71

31. Tricco AC, Lillie E, Zarin W, O'Brien KK, Colquhoun H, Levac D, et al. PRISMA extension for scoping reviews (PRISMA-ScR): checklist and explanation. *Ann Intern Med.* (2018) 169:467–73. doi: 10.7326/M18-0850

32. Bergeron A, Beauregard F. Perceptions of six immigrant parents of their child's language disorder and care experience. *Can J Speech Lang Pathol Audiol.* (2018) 42:145–60.

33. Bird EK, Lamond E, Holden J. Survey of bilingualism in autism spectrum disorders. *Int J Lang Commun Disord.* (2012) 47:52–64. doi: 10.1111/j.1460-6984.2011.00071.x

34. Caesar LG, Kohler PD. The state of school-based bilingual assessment: actual practice versus recommended guideline. *Lang Speech Hear Serv Sch.* (2007) 38:190–200. doi: 10.1044/0161-1461(2007/020)

35. de Valenzuela JS, Bird EK, Parkington K, Mirenda P, Cain K, MacLeod AA, et al. Access to opportunities for bilingualism for individuals with developmental disabilities: key informant interviews. *J Commun Disord.* (2016) 63:32–46. doi: 10.1016/j.jcomdis.2016.05.005

36. Fong VC, Lee BS, Iarocci G. A community-engaged approach to examining barriers and facilitators to accessing autism services in Korean immigrant families. *Autism.* (2022) 26:525–37. doi: 10.1177/13623613211034067

37. Hammer CS, Detwiler JS, Detwiler J, Blood GW, Dean Qualls C. Speech-language pathologists' training and confidence in serving Spanish-English bilingual children. *J Commun Disord.* (2004) 37:91–108. doi: 10.1016/j.jcomdis.2003.07.002

38. Jimenez ME, Martinez Alcaraz E, Williams J, Strom BL. Access to developmental pediatrics evaluations for at-risk children. *J Dev Behav Pediatr.* (2017) 38:228–32. doi: 10.1097/DBP.0000000000000427

39. Jordaan H. Clinical intervention for bilingual children: an international survey. *Folia Phoniatr Logop.* (2008) 60:97–105. doi: 10.1159/000114652

40. Kritikos EP. Speech-language pathologists' beliefs about language assessment of bilingual/bicultural individuals. *Am J Speech Lang Pathol.* (2003) 12:73–91. doi: 10.1044/1058-0360(2003/054)

41. Kuhn J, Levinson J, Udhnani MD, Wallis K, Hickey E, Bennett S, et al. What happens after a positive primary care autism screen among historically underserved families? Predictors of evaluation and autism diagnosis. *J Dev Behav Pediatr.* (2021) 42:515–23. doi: 10.1097/DBP.0000000000000928

42. Marinova-Todd SH, Colozzo P, Mirenda P, Stahl H, Kay-Raining Bird E, Parkington K, et al. Professional practices and opinions about services available to bilingual children with developmental disabilities: an international study. *J Commun Disord.* (2016) 63:47–62. doi: 10.1016/j.jcomdis.2016.05.004

43. Mcleod S, Baker E. Speech-language pathologists' practices regarding assessment, analysis, target selection, intervention, and service delivery for children with speech sound disorders. *Clin Linguist Phon.* (2014) 28:508–31. doi: 10.3109/02699206.2014.926994

44. Mennen I, Stansfield J. Speech and language therapy service delivery for bilingual children: a survey of three cities in Great Britain. *Int J Lang Commun Disord.* (2006) 41:635–52. doi: 10.1080/13682820600623911

45. Mulgrew L, Duffy O, Kennedy L. Assessment of minority language skills in English-Irish-speaking bilingual children: a survey of SLT perspectives and current practices. *Int J Lang Commun Disord.* (2022) 57:63–77. doi: 10.1111/1460-6984.12674

46. Nayeb L, Wallby T, Westerlund M, Salameh EK, Sarkadi A. Child healthcare nurses believe that bilingual children show slower language development, simplify screening procedures and delay referrals. *Acta Paediatr.* (2015) 104:198–205. doi: 10.1111/apa.12834

47. Pascoe M, Maphalala Z, Ebrahim A, Hime D, Mdladla B, Mohamed N, et al. Children with speech difficulties: an exploratory survey of clinical practice in the Western Cape. *S Afr J Commun Disord.* (2010) 57:66–75. doi: 10.4102/sajcd.v57i1.51

48. Scharff Rethfeldt W. Speech and language therapy services for multilingual children with migration background: a cross-sectional survey in Germany. *Folia Phoniatr Logop.* (2019) 71:116–26. doi: 10.1159/000495565

49. Knuti Rodrigues K, Hambidge SJ, Dickinson M, Richardson DB, Davidson AJ. Developmental screening disparities for languages other than English and Spanish. *Acad Pediatr.* (2016) 16:653–9. doi: 10.1016/j.acap.2015.12.007

50. St Amant HG, Schrager SM, Peña-Ricardo C, Williams ME, Vanderbilt DL. Language barriers impact access to services for children with autism spectrum disorders. *J Autism Dev Disord.* (2018) 48:333–40. doi: 10.1007/s10803-017-3330-y

51. Vanegas SB. Examining factors related to the age of diagnosis of children with autism spectrum disorder from immigrant and non-immigrant backgrounds in a diverse clinical sample. *Autism Res.* (2021) 14:1260–70. doi: 10.1002/aur.2489

52. Verdon S, McLeod S, McDonald S. A geographical analysis of speech-language pathology services to support multilingual children. *Int J Speech Lang Pathol.* (2014) 16:304–16. doi: 10.3109/17549507.2013.868036

53. Wiefferink K, van Beugen C, Sleeswikj B, Gerrits E. Children with language delay referred to Dutch speech and hearing centres: caseload characteristics. *Int J Lang Commun Disord.* (2020) 55:573–82. doi: 10.1111/1460-6984.12540

54. Williams CJ, McLeod S. Speech-language pathologists' assessment and intervention practices with multilingual children. *Int J Speech Lang Pathol.* (2012) 14:292–305. doi: 10.3109/17549507.2011.636071

55. Yu B, Hsia S. Inclusion of heritage language learners on the autism spectrum: Lessons from second-generation parents. *Int J Appl Linguist.* (2019) 29:356–69. doi: 10.1111/ijal.12233

56. Zuckerman KE, Lindly OJ, Reyes NM, Chavez AE, Cobian M, Macias K, et al. Parent perceptions of community autism spectrum disorder stigma: measure validation and associations in a multi-site sample. *J Autism Dev Disord.* (2018) 48:3199–209. doi: 10.1007/s10803-018-3586-x

57. Zuckerman KE, Lindly OJ, Reyes NM, Chavez AE, Macias K, Smith KN, et al. Disparities in diagnosis and treatment of autism in Latino and Non-Latino White families. *Pediatrics.* (2017) 139:e20163010. doi: 10.1542/peds.2016-3010

58. Zuckerman KE, Mattox K, Donelan K, Batbayar O, Baghaee A, Bethell C. Pediatrician identification of Latino children at risk for autism spectrum disorder. *Pediatrics.* (2013) 132:445–53. doi: 10.1542/peds.2013-0383

59. Zuckerman KE, Sinche B, Mejia A, Cobian M, Becker T, Nicolaidis C. Latino parents' perspectives on barriers to autism diagnosis. *Acad Pediatr*. (2014) 14:301–8. doi: 10.1016/j.acap.2013.12.004

60. Zuckerman KE, Sinche B, Cobian M, Cervantes M, Mejia A, Becker T, et al. Conceptualization of autism in the Latino community and its relationship with early diagnosis. *J Dev Behav Pediatr*. (2014) 35:522–32. doi: 10.1097/DBP.0000000000000091

61. USA Census Bureau. *Quick Facts: Denver County Colorado*. (2020). Available online at: https://www.census.gov/quickfacts/denvercountycolorado (accessed March 20, 2022).

62. Pew Research Center. *Use of Spanish declines among Latinos in major U.S. metros*. (2017). Available online at: https://www.pewresearch.org/fact-tank/2017/10/31/use-of-spanish-declines-among-latinos-in-major-u-s-metros/#spanish-language-use-in-major-u-s-metro-areas (accessed March 20, 2022).

63. De Lamo White C, Jin L. Evaluation of speech and language assessment approaches with bilingual children. *Int J Lang Commun Disord*. (2011) 46:613–27. doi: 10.1111/j.1460-6984.2011.00049.x

64. The Royal College of Speech and Language Therapists. *RCSLT Good Practice Guidelines*. Milton Keynes: Speechmark Publishing Ltd (2005).

65. Beauchamp ML, MacLeod, AA. The role of exposure on school-aged bilingual language abilities– it depends on what you measure. *J Biling Educ Biling*. (2022) 1–16. doi: 10.1080/13670050.2022.2062217

66. Ungar M, Theron L. Resilience and mental health: how multisystemic processes contribute to positive outcomes. *Lancet Psychiatry*. (2020) 7:441–8. doi: 10.1016/S2215-0366(19)30434-1

67. Kruger J, Dunning D. Unskilled and unaware of it: how difficulties in recognizing one's own incompetence lead to inflated self-assessments. *J Pers Soc Psychol*. (1999) 77:1121–34. doi: 10.1037/0022-3514.77.6.1121

68. Zeidan J, Fombonne E, Scorah J, Ibrahim A, Durkin MS, Saxena S, et al. Global prevalence of autism: a systematic review update. *Autism Res*. (2022) 15:778–90. doi: 10.1002/aur.2696

69. Tesfaye R, Courchesne V, Yusuf A, Savion-Lemieux T, Singh I, Shikako-Thomas K, et al. Assuming ability of youth with autism: synthesis of methods capturing the first-person perspectives of children and youth with disabilities. *Autism*. (2019) 23:1882–96. doi: 10.1177/1362361319831487

70. Courchesne V, Tesfaye R, Mirenda P, Nicholas D, Mitchell W, Singh I, et al. Autism voices: a novel method to access first-person perspective of autistic youth. *Autism*. (2021) 1–14. doi: 10.1177/13623613211042128

frontiers | Frontiers in Psychiatry

TYPE Original Research
PUBLISHED 29 August 2022
DOI 10.3389/fpsyt.2022.909947

Check for updates

OPEN ACCESS

EDITED BY
Diana Schendel,
Drexel University, United States

REVIEWED BY
Jonna Bobzien,
Old Dominion University, United States
Tingyu Li,
Chongqing Medical University, China

*CORRESPONDENCE
Cecilia Montiel-Nava
cecilia.montielnava@utrgv.edu

SPECIALTY SECTION
This article was submitted to
Autism,
a section of the journal
Frontiers in Psychiatry

RECEIVED 31 March 2022
ACCEPTED 04 August 2022
PUBLISHED 29 August 2022

CITATION
Montiel-Nava C, Tregnago M,
Marshall J, Sohl K, Curran AB,
Mahurin M, Warne-Griggs M, WHO
CST Team and Dixon P (2022)
Implementing the WHO caregivers
skills training program with caregivers
of autistic children *via* telehealth in
rural communities.
Front. Psychiatry 13:909947.
doi: 10.3389/fpsyt.2022.909947

Implementing the WHO caregivers skills training program with caregivers of autistic children *via* telehealth in rural communities

Cecilia Montiel-Nava[1]*, Megan Tregnago[2], Jeanne Marshall[2], Kristin Sohl[3,4], Alicia Brewer Curran[4], Melissa Mahurin[4], Melissa Warne-Griggs[5], WHO CST Team[6] and Pamela Dixon[7]

[1]Department of Psychological Science, University of Texas Rio Grande Valley-Edinburg, Edinburg, TX, United States, [2]Easterseals Midwest, Columbia, MO, United States, [3]Department of Child Health, University of Missouri School of Medicine, Columbia, MO, United States, [4]ECHO Autism Communities, University of Missouri School of Medicine, Columbia, MO, United States, [5]Missouri Telehealth Network, School of Medicine University of Missouri, Columbia, MO, United States, [6]Department of Mental Health and Substance Use, World Health Organization, Geneva, Switzerland, [7]Autism Speaks, Princeton, NJ, United States

Background: For families with autistic children living in rural areas, limited access to services partly results from a shortage of providers and extensive travel time. Telehealth brings the possibility of implementing alternative delivery modalities of Parent Mediated Interventions (PMIs) with the potential to decrease barriers to accessing services. This study aimed to evaluate the feasibility and acceptability of implementing the World Health Organization-Caregivers Skills Training program (WHO-CST) *via* an online, synchronous group format in rural Missouri.

Methods: We used a mixed methods design to collect qualitative and quantitative data from caregivers and program facilitators at baseline and the end of the program, following the last home visit. Caregivers of 14 autistic children (3–7 years), residents of rural Missouri, completed nine virtual sessions and four virtual home visits.

Results: Four main themes emerged from the focus groups: changes resulting from the WHO-CST, beneficial aspects of the program, advantages and disadvantages of the online format, and challenges to implementing the WHO-CST *via* telehealth. The most liked activity was the demonstration (36%), and the least liked was the practice with other caregivers. From baseline to week 12, communication skills improved in both frequency ($p < 0.05$) and impact ($p < 0.01$), while atypical behaviors decreased ($p < 0.01$). For caregivers' outcomes, only confidence in skills ($p < 0.05$) and parental sense of competence ($p < 0.05$) showed a positive change.

Conclusion: Our results support the feasibility of implementing the WHO-CST program *via* telehealth in a US rural setting. Caregivers found strategies easy to follow, incorporated the program into their family routines, and valued the group meetings that allowed them to connect with other families. A PMI such as the WHO-CST, with cultural and linguistic adaptations and greater accessibility *via* telehealth-plays an essential role in closing the treatment gap and empowering caregivers of autistic children.

KEYWORDS

Autism Spectrum Disorder, parent-mediated behavioral intervention, parenting skills training program, ECHO Autism, rural, telehealth

Introduction

Autism Spectrum Disorder (ASD) is a neurodevelopmental disorder characterized by social-communication impairment and repetitive/restrictive patterns of behavior (1). Parents of autistic[1] children can learn techniques to address the ASD core characteristics, such as promoting communication and social skills, joint attention, positive behaviors, and decreasing restricted and repetitive behaviors in children (3). Parent-Mediated Interventions (PMIs) refer to a group of interventions in which parents are taught strategies typically used by therapists that they can implement with their child in everyday situations. An increasing body of evidence shows PMIs' effectiveness in increasing social interaction and communication, and decreasing atypical behaviors in autistic children (4). In addition, they are frequently used as early intervention protocols for children on the spectrum (3). Common elements of evidence-based PMIs include goal setting, use of behavioral principles, a focus on naturalistic settings and interactions, and systematic evaluation of outcomes (5). The literature indicates that parents of autistic children can learn techniques to promote development and positive behaviors in their children. Previous studies have shown that parents can implement treatment strategies to improve or increase communication skills (6–9), social skills (10) and joint attention (11–13). Besides effectively reducing challenging behaviors and restricted and repetitive behaviors in autistic children (14, 15), PMIs also increase their self-help skills (16). In addition, training in behavioral interventions promotes self-efficacy in parents (17, 18), and parent self-efficacy is associated with positive treatment outcomes for children (11, 19–21). In the U.S., children with an ASD diagnosis between the ages of three and five are also eligible to receive early childhood special education services (22), including speech-language therapy, occupational therapy, and therapy based on the applied behavior analysis (ABA) principles. However, current knowledge about the amounts and types of special education services autistic children have access to is limited (23).

Autism prevalence has doubled in the last decade, increasing service demands and becoming a public health concern (24, 25). Despite the compelling evidence for the efficacy of PMIs, most studies have included parents living in urban areas with more access to services, suggesting that samples do not represent the broader and diverse US population. Consequently, the dissemination of evidence-based interventions is limited among rural communities in the US. Furthermore, there are inequalities in accessing services for autistic individuals living in rural or low-income neighborhoods compared to those living in metropolitan areas (26). Such disparities have been attributed to limited health care resources, shortage of specialized health professionals, and structural factors such as travel distance to service and costs (27). According to the Census Bureau (28), 60 million or 1 in every 5 Americans live in rural areas. Many rural regions have been categorized as shortage areas: geographic areas, populations, and facilities with too few primary care, dental and mental health providers and services [HRSA] (29). As of December 2021, it is estimated that there are 5,999 mental health professional shortage areas in the US, wherein 136 million people reside, with an estimated need of 6,806 mental health practitioners (29). For families with autistic children living in rural areas, limited access to services is partly a function of a shortage of providers and extensive travel time, both for in-home service providers and parents driving to hospitals and clinics that offer services (30). Missouri is among the states with a shortage of health and mental health professionals in the US (31). Children in rural areas are more likely to live in poverty than children in urban settings affecting the health and mental health outcomes of autistic children (32, 33). Therefore, interventions aiming to address such inequalities and social determinants of mental health might differ from families residing in urban or rural settings in the same country (34). Many families with autistic children living in rural areas in the US might not have access to experienced healthcare providers who could make appropriate referrals or offer quality parent-skills training programs. Participation in those services may improve child outcomes, decrease parental stress, and increase parent competency and efficacy (20). Consequently, living in a rural area in the US could be considered a risk factor

---

[1] We are aware of the diverse opinions regarding the terminology used to refer to individuals on the spectrum. We have elected to use identity-first instead of person-first language following the suggestions to avoid ableist language (2).

for diminished access to evidence-based early interventions for autistic children, thus increasing the odds for poorer outcomes in this vulnerable population.

As a response to the global treatment gap for children with developmental disabilities, especially those in low-resource and underserved populations, The World Health Organization (WHO) developed the Caregiver Skills Training (CST) program. WHO-CST is a parent-mediated intervention, freely available and adapted to various settings and levels of care, that aims to decrease the treatment gap for children with developmental disabilities globally, especially those in low-income and underserved settings. The WHO-CST program was developed through extensive stakeholder consultation and an iterative revision, increasing its external validity (35). The WHO-CST program takes (a) a *task-shifting approach*: non-specialists (e.g., social workers and trained community volunteers, caregivers) can deliver this program, (b) a *trans-diagnostic approach:* it does not require a diagnosis to qualify for treatment, and (c) a *common elements approach*: its content focuses on strategies that can benefit a group of caregivers with diverse needs (36, 37). The WHO-CST is delivered *via* nine group sessions and three home visits, providing caregivers with skills that can be used in daily home and play routines. Skills taught in the nine group sessions target social communication, adaptive behavior, and behavior management. The WHO-CST program was developed with the expectation that a community or country will translate and adapt the materials to be culturally relevant without changing the core content. Specific guidance for adaptation is provided with the field-test version of the WHO-CST materials (WHO-CST Team, unpublished). The WHO-CST is currently being adapted and implemented in more than 30 countries worldwide. Outcome evaluations in Ethiopia (38), India (39) and Italy (40) indicate that the WHO-CST is valued as a positive intervention for caregivers and community stakeholders with minimal sociocultural barriers (38–40). The CST's preliminary data from different geographical regions emphasize the adaptation process as essential to ensure its implementation and sustainability. The program was designed to be implemented globally and suitable for low-resource contexts and has shown good acceptability in high-income settings too (40, 41). Thus, the WHO-CST is a sustainable and valid program to implement in the rural US.

Telehealth brings the possibility of implementing alternative delivery modalities of PMIs to decrease barriers to accessing services, such as the limited health care resources and structural factors mentioned above. Telehealth strategies incorporating technology to provide health care services have been explored as a potential solution to the challenges of reaching families in rural settings, resulting in positive caregiver outcomes and satisfaction with services (42, 43). As a result of restrictions associated with the COVID-19 pandemic, caregivers of children with disabilities across geographic settings (i.e., urban, rural) experienced difficulties accessing services. A recent review of telehealth applications for ASD (44) indicates that studies involving PMIs use a variety of approaches to teaching parents strategies to promote child engagement and communication and manage challenging behaviors. Strategies include using video conferencing technology to train parents individually, in groups, and through self-guided websites. Reported outcomes include high levels of parent satisfaction (29, 45, 46), reductions in challenging behaviors (42, 47) and parental stress (48, 49), and improvements in child adaptive functioning (50). Overall, telehealth interventions are well-received by parents and have comparable outcomes to in-person services, providing some components of individual support and coaching (44, 51). The COVID-19 pandemic has exposed the power and usefulness of remote parent support in families with autistic children (39). Empirical data support the efficacy of using PMIs with parents of autistic children, as well as telehealth approaches to deliver PMIs (19, 42, 45, 52–56).

Given the effectiveness of PMIs, the existing barriers to accessing services for families with an autistic child living in rural settings, and the growing body of research supporting the use of telehealth to deliver PMIs, this study aimed to evaluate the feasibility and acceptability of implementing the WHO-CST program *via* an online synchronous group format in rural. Outcomes from this study will support a much larger implementation trial in rural settings.

Materials and methods

Study design

The study used a mixed-methods design and collected qualitative and quantitative data from caregivers and program facilitators. A phenomenological framework, as well as a cross sectional survey design, examined the feasibility and acceptability of the WHO-CST in a rural US setting using an online delivery format. In addition, a one-group pretest-posttest with matching design was used to examine if there exists a difference in child and caregivers' outcomes after the program implementation. Data was collected at baseline and at the end of the program, following the last home visit.

Potential participants were referred to research staff at the University of Missouri–Columbia by ECHO Autism clinicians, Missouri Regional Offices and Easterseals Midwest staff. Caregivers who expressed interest in participating in the study were referred to a team member. All participants were contacted by research staff at the University of Missouri–Columbia who provided detailed information about the study. The consent form was verbally reviewed over the telephone for caregivers who wanted to participate. Study data were collected and managed using Research Electronic Data Capture (REDCap) (57) hosted at the University of Missouri–Columbia. REDCap was used to send caregivers a unique link to complete the electronic consent and additional questionnaires.

Following recruitment and consent, participants completed the pre-intervention questionnaires. Each caregiver was assigned to one of three different groups, depending on the time of the scheduled sessions they selected during recruitment. Starting and finish weeks were the same for all groups, but each group offered different days and times for their sessions. Participants attended nine, 90-min group sessions in an online format (conducted through Zoom) and three, 60 to 90-min virtual home visits. One additional 15-min virtual home visit occurred for families after session one to review a goal setting sheet and answer questions the family had regarding their first session.

At the end of the nine group sessions and the four home visits, participants completed the post-intervention questionnaires. Attendance and post session feedback were collected at every encounter. Master Trainers and caregivers participated in separate post-intervention focus groups during December 2020. Group sessions lasted between 40 and 60 min each and were conducted by an experienced independent qualitative researcher who was not a facilitator of any group in the study.

All study procedures were approved by the University of Missouri-Columbia Institutional Review Board, and caregivers, Master Trainers, and Facilitators provided informed consent before collecting study data.

Participants

Eligibility for the study included caregivers of children between 24 months and 9 years of age diagnosed with ASD. In addition, to be included in the study, caregivers and children were required to (1) be residents of one of Easterseals Midwest's rural catchment areas at the time of the study (i.e., Central, Northeast, or Southeast Missouri), (2) have a reliable internet connection, and (3) access to Zoom through a desktop computer, laptop, or tablet with a video camera embedded or attached, (4) consent to videotaping of virtual home visits, and (5) be fluent in English. There were no additional exclusion criteria.

Caregivers of 18 children completed the initial screening procedures and signed electronic consent forms. Sixteen caregivers completed baseline outcome measures, but only 15 completed the program (one caregiver did not complete post-intervention measures). Table 1 presents the demographic characteristics of the sample. Most caregivers were female (92.9%, $n = 13$), White-non-Hispanic (78.6%, $n = 11$) and had some college education (78.6%, $n = 11$). Half of the sample had received prior training on similar topics (50%, $n = 7$). Children in the study were mostly male (64.3%, $n = 9$), with a mean age of 4.5 years (SD = 1.63). In terms of the history of services, 79 % ($n = 11$) reported having received support from the schools. However, the questionnaire did not inquire about the type of support that was offered. Caregivers also indicated that children had received behavior therapy (57%, $n = 8$), speech–language therapy (71%, $n = 10$), and medication (29%, $n = 4$); but without specification of where that service was provided (school, clinic, or home).

TABLE 1 Demographic characteristics of the participants.

Characteristics	***N* = 14**
Caregivers demographics	
Gender	
Female	13 (92.9%)
Male	1 (7.1%)
Age, mean ± SD	37.07 ± 5.51
Caregivers education	
High school diploma	1 (7.1%)
Some college	6 (42.9%)
Bachelor's degree	3 (21.4%)
Graduate degree	4 (28.6%)
Ethnicity	
Non-hispanic/non-latino origin	14 (100%)
Race	
White, non-hispanic	11 (78.6%)
African American, non-hispanic	2 (14.3)
White, hispanic	0 (0%)
African American, hispanic	0 (0%)
Not reported	1 (7.1%)
Marital status	
Single	2 (14.3%)
Married	8 (57.1%)
Divorced	3 (21.4%)
Living with romantic partner	1 (7.1%)
Children under 18 at home	
1	1 (7.1%)
2	8 (57.1%)
3	2 (14.3%)
4+	3 (21.4%)
Other children with developmental delays	2 (14.3%)
Relationship with child with developmental delays	
Mother	12 (85.7%)
Father	1 (7.1%)
Grandmother	1 (7.1%)
Had received training on similar topics	7 (50%)
Had received information on this topic	11 (78.6)
Child demographics	
Gender	
Female	5 (35.7%)
Male	9 (64.3%)
Age, mean ± SD	4.53±1.63
History of Services	
School support	11 (79%)
Speech/language therapy	10 (71%)
Behavior therapy	8 (57%)
Medication	4 (29%)

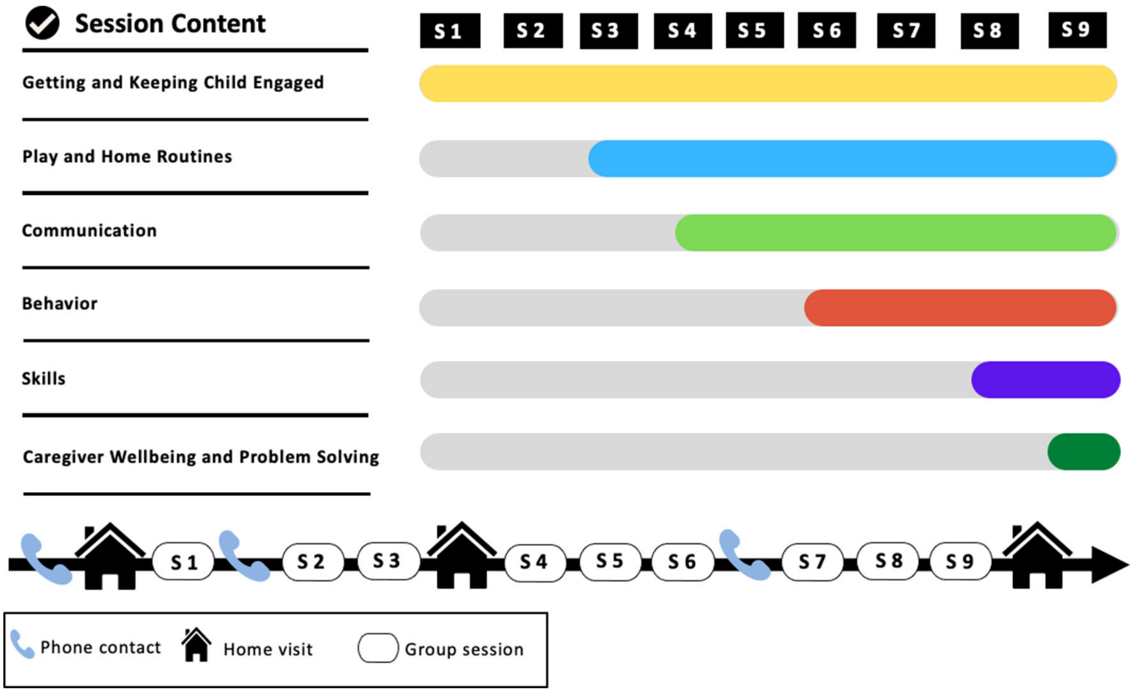


FIGURE 1
WHO-CST curriculum.

Intervention

The WHO-CST program was designed to teach caregivers strategies to engage their child in communication and play, to promote adaptive behaviors and learning, and to reduce challenging behavior (35). Its content is based on principles of social learning theory, positive parenting, ABA, and developmental theories. The program consists of a combination of nine group sessions for caregivers and three individual home visits. The content of each of the nine sessions is as follows: (a) introduction and psychoeducation, (b) engaging with the child, (c) helping children share engagement, (d) understanding communication, (e) promoting communication, (f) preventing challenging behavior, (g) responding to challenging behavior, (h) learning new skills, and (i) caregiver problem solving and self-care (Figure 1). For the group sessions, Facilitators implement various techniques, including modeling, role-play, demonstrations, group discussions, and case vignettes. Each session includes homework assignments to encourage caregivers to implement the learned skills in everyday home situations. Before starting the program, Facilitators complete a home visit to define specific goals and targets for each family, explore the presence of additional health problems the child may have, observe the caregiver and child's interactions, inform and engage other caregivers, and answer questions about the program. For the other two home visits occurring halfway through the program and after the last group session, Facilitators focus on coaching the caregiver and providing tailored support, evaluating progress, troubleshooting, and identifying possible additional support needs.

Adaptation and pre-pilot testing

A group of stakeholders met to review the materials and assess the need for adaptations for the rural Midwest setting. Stakeholders included four parent trainers with a minimum of 6 years of experience teaching caregivers with autistic children. At this beginning stage, adaptations were mainly linguistic (i.e., changing British spellings to American English) and adapting examples and names to be more culturally representative of the population, consistent with the adaptation guide for the WHO-CST (WHO-CST Team, unpublished). These four parent trainers simultaneously completed a 5-day in-person intensive training with the World Health Organization-Autism Speaks (WHO-AS) team. This training included reviewing the WHO-CST materials, role-playing the sessions, and conducting live practice sessions with mothers and their autistic children. Each Master Trainer met treatment fidelity of at least 80% with the WHO-AS team. Another adaptation to the WHO-CST training of Master Trainers was the supervision component. Typically, Master Trainers send implementation videos of the WHO-WHO-CST model to the WHO-AS trainers and one-on-one feedback is provided. During this pilot, the team adapted the supervision process by leveraging the Extension for Community Healthcare Outcomes (ECHO) model® to aid Master Trainers in achieving fidelity to the WHO-CST model.

ECHO is a model that utilizes video-conferencing technology to provide professionals, such as clinicians, educators, and advocates with the knowledge and guided practice needed to further develop professional expertise. The model has a "hub and spoke" framework that allows spokes, or professionals, to present cases to an interdisciplinary "hub team" of experts. Hub team members mentor and coach spokes to improve spoke knowledge and confidence in their ability to provide best practice care and build a community of practice. The core components of the ECHO model® include a cased-based presentation and a brief didactic. This model has been successfully applied to autism, and the ECHO Autism framework has shown to improve the self-efficacy of community-based clinicians, creating access to high-quality autism care for autistic people and their families in local communities (58).

For this study, the WHO-CST utilized the ECHO Autism framework for supervision and training of Master Trainers (58, 59). WHO-CST teleECHO sessions were led by a hub team of WHO-CST experts that included two WHO-AS trainers and two to four global focal points (i.e., other Master Trainers around the world). Weekly ECHO Autism: WHO-CST teleECHO sessions were hosted by the expert hub team. During the teleECHO sessions, one of the WHO-AS trainers presented a 10-min didactic regarding one of the WHO-CST sessions or primary components. Videos submitted by the Master Trainers were watched by all participants and feedback was given as a group to promote discussion and improved skill application. Fidelity of WHO-CST skills and strategies was evaluated by two clinical psychologists with expertise in implementing the WHO-CST using the WHO-CST Adult/Child Interaction Fidelity scale v1.0 (WHO-CST Team, unpublished). All ECHO Autism teleECHO sessions were completed prior to starting the pre-pilot WHO-CST group.

Facilitator training

After completing the pre-pilot testing, the four master trainers conducted a training of four WHO-CST facilitators. The four Master Trainers delivered a 16-h online (i.e., Zoom) training for the facilitators that covered the content of the WHO-CST sessions, group facilitation behaviors, video reviews of the strategies, an introduction to the ECHO Autism framework, and telehealth protocols. Due to COVID-19 protocols in place at that time, Facilitators were not able to practice WHO-CST skills with children in person. As a replacement method, video examples of Master Trainers exhibiting these skills during their fidelity training were used for teaching.

The team leveraged the ECHO Autism-WHO-CST program to provide supervision to Facilitators. ECHO Autism teleECHO sessions occurred once per week while the telehealth WHO-CST program was being delivered, for a total of 14 sessions. During the ECHO Autism teleECHO sessions, one of the Hub Team Master Trainers presented a didactic lesson on the upcoming week's WHO-CST session or Home Visit with the Facilitators. Additionally, Facilitators were scheduled to present two cases. One case presentation focused on the dynamics of the previous week's WHO-CST session, and the second case presentation focused on the experiences the Facilitators had with one caregiver-child dyad that they supported during a home visit. Following rich case discussions generated by the case presentations, the hub team provided Facilitators with feedback and recommendations about how to address any challenges faced during WHO-CST sessions or virtual home visits.

Measures

During the different stages of the project, caregivers completed a variety of measures to document feasibility and acceptability of the program as well as preliminary efficacy of this delivery method (telehealth) (Table 2), the majority of which were derived from the WHO-CST monitoring-evaluation framework (WHO-CST Team, unpublished).

Demographic and service history information

At study entry, caregivers were asked to complete a questionnaire electronically to obtain basic information about the dyad such as the child's gender, age, diagnosis, language spoken at home, ethnicity, and caregiver demographics including gender, relationship to the child, birthdate, and occupation. The caregiver was also asked to report the child's history of medical services and psychological interventions.

Feasibility and acceptability outcomes

The following measures were obtained to determine whether the treatment was acceptable and feasible to caregivers and Master Trainers.

(a) **WHO-CST Session Attendance**- Caregivers' attendance at the group and individual sessions was tracked by the program Facilitators.
(b) **Focus Groups**– Focus groups were conducted with WHO-CST Master Trainers and participants. Focus groups followed a guide adapted from questions designed by the CST-WHO developers, consisting of open-ended questions related to expectations, positive experiences, and

TABLE 2 Description of the instruments and timeline.

Purpose	Data type	Timeline
Demographics and service history	Electronic intake questionnaire	Study entry
Feasibility outcomes	Attendance	Recorded after each session
	Focus groups	Study exit
	WHO-CST post session feedback form from facilitators	Recorded after each session
	Caregivers knowledge and confidence in skills	Study entry and study exit
Acceptability outcomes	WHO-CST post session feedback form from caregivers	Recorded after each session
Preliminary efficacy outcomes	Autism Treatment and Evaluation Checklist (ATEC) Autism Impact Measure (AIM) Brief Family Distress Scale (BFDS) Kessler screening scale for psychological distress Parental stress scale Parenting Sense of Competence (PSOC)	Study entry and study exit

opportunities for improvement. Additional questions were included to inquire about the online format of the program, and follow-up questions were used to stimulate discussion. All focus groups were conducted in December 2020, and audio recorded *via* Zoom.

(c) **Caregivers Knowledge and Skills Test** (WHO-CST Team, unpublished)– A questionnaire with three sections was given at study entry and study exit to assess caregivers' knowledge and skills related to the WHO-CST content. The first section includes 38 statements about the main skills taught by the WHO-CST. Caregivers are asked to indicate the degree to which they agree with each statement using a 5-point Likert scale (1-Strongly Disagree-5 Strongly Agree). The total score on this subscale ranged from 38 to 190. The second part includes 13 questions about caregivers' confidence in applying some of the skills and knowledge taught by WHO-CST (1-Not at all confident-5 Very Confident), with scores ranging from 13 to 65. The third section is comprised of three vignettes followed by a request a list of three suggestions in how the caregivers in the vignette could respond to the specific situation. For the purposes of this study, sections one and two were included in the analysis. Higher scores are associated with greater knowledge of concepts and strategies taught by the WHO-CST, and confidence in applying those skills with their children.

(d) **Post-session Feedback Form from Facilitators** [adapted from Kasari et al. (11)]-it renders information about acceptability and feasibility of the group sessions according to the Facilitators. Facilitators complete it immediately after each session. Using a 5-point Likert scale, Facilitators will rate the complexity of the content, amount of content for the time of the session, and their perceived preparedness to run the session (feasibility). On the other hand, it requires the Facilitator to rate if the session's content was relevant for caregivers, caregivers' agreement with the ideas presented and their participation and engagement during the sessions (acceptability). At the end it has two open-ended questions about suggestions to improve their preparedness and changes they would make to the session.

(e) **Post session Feedback Form from Caregivers** [adapted from Kasari et al. (11)]- it is a 14-item form that caregivers complete after each session to measure acceptability of group sessions. Using a 5-point Likert scale, caregivers rate the difficulty level of the content ("Did you find this session easy to understand?"), its relevance ("How well do you think the information in this session applies to you and your child?"), usefulness ("How useful will the key messages and tips be to you and your child?") and alignment with values ("The messages in this session are in conflict with what I believe is good and important"). Another set of questions (multiple choice) inquired about the most and least liked learning activity during the session. With a 3-point Likert scale, 3 questions asked about the length of the whole session, of sharing experiences and discussions, and of practice in pairs (1-Too long 2–Too short 3-Just right). In addition, another section requires the caregivers to rate the usefulness of the WHO-CST tips or strategies that were included in each section. Lastly, there was a space for caregivers to write suggestions to improve the delivery or content of the session ("What could be done differently to improve the session?").

Clinical measures

Several caregiver-report measures were used to examine caregiver and child responses to the WHO-CST program.

(a) **The Autism Treatment and Evaluation Checklist (ATEC)** (60)- This questionnaire was developed to measure changes in response to treatment (60). The ATEC is a one-page 77-item checklist, completed by caregivers, assessing developmental skills and severity of symptoms of developmental delays. It includes four subtests (1) speech/language communication (14 items), (2) sociability

(20 items), (3) sensory-cognitive awareness (18 items), and (4) Health/physical behavior. ATEC has been found to be responsive to change in children with autism and shows high internal consistency in the English original and cross-cultural translation (61, 62). Total scores range from 0 to 180, and the higher the score the worse the symptoms.

(b) **The Autism Impact Measure (AIM) (63)**– The AIM is a caregiver-reported questionnaire assessing autism symptom frequency and impact in children. It was designed specifically for treatment-outcome assessment in children with ASD, focusing on treatment-relevant aspects of symptom presentation and efficient detection of short-term improvement (64). The AIM has 41 parent-rated items, each requiring two corresponding 5-point ratings (frequency and impact). Items reflect either the presence of a maladaptive behavior or the absence of an expected skill.

(c) **Brief Family Distress Scale** (BFDS) (65)– The BFDS is a 10-item parent-report scale designed to indicate the family's level of stress and crisis. A score of one represents perception of positive coping while a score of 10 indicates a marked level of distress where the caregiver perceives the family is currently in crisis. The BFDS has been examined with families of children with ASD. High scores are correlated with problematic coping while low scores are correlated with positive adjustment and coping.

(d) **The Kessler Screening Scale for Psychological Distress (K6)** (66)– The Kessler Psychological Distress Scale (K6+) is a six-item self-report measure of psychological distress intended to be used as a quick tool to assess risk for serious mental illness in the general population, and in this study, it is used in caregivers. It was developed for use in the annual US National Health Interview Survey and National Household Survey on Drug Abuse (66). A cut-point of $K6 \geq 13$ is the accepted score for serious mental illness (67).

(e) **Parental Stress Scale** (68)– The Parental Stress Scale is a self-report scale that contains 18 items representing pleasure or positive themes of parenthood (emotional benefits, self-enrichment, personal development) and negative components (demands on resources, opportunity costs and restrictions). Respondents are asked to agree or disagree with items in terms of their typical relationship with their child or children and to rate each item on a five-point scale: strongly disagree (1), disagree (2), undecided (3), agree (4), and strongly agree (5). The eight positive items are reverse scored so that possible scores on the scale can range between 18 and 90. Higher scores on the scale indicate greater stress. This scale was designed to assess outcomes of interventions designed to support parenting efficacy of caregivers.

(f) **Parenting Sense of Competence (PSOC)** (69)– The PSOC is a 17-item scale designed to measure parents' satisfaction with parenting and their self-efficacy in the parenting role. Parents indicate their level of agreement with each item by circling a number between 1 (strongly agree) and 6 (strongly disagree). Eight items are reverse scored so that high scores indicate positive parental experience. It has two different subscales: (1) parenting satisfaction (PSOC-S) defined as the person's liking of the parenting role, and (2) parenting efficacy (PSOC-E), defined as the person's perceived competence in the parenting role (69). For this study, we used the Parental Efficacy subscale (PSOC-E).

Data analysis

Statistical analysis

Statistical significance was evaluated at the 0.05 level, and data analyses were performed using IBM SPSS, version 26. Child and caregiver characteristics were calculated for the full sample using means and standard deviations for continuous variables and frequencies and percentages for categorical measures. To evaluate change in outcomes among participants, baseline scores of child and caregivers' outcomes and post intervention scores were compared with paired *t*-tests.

Qualitative analysis

Audio recordings of interviews were transcribed using an automated transcription service (Temi.com). To maintain participant privacy, recordings were de-identified before placing them into a secure folder shared with the research team. To ensure accuracy of transcription, the research team compared each transcript to its recording and made any needed edits to match the audio. After each focus group or interview, the focus group facilitator noted important points and lasting impressions from the interview. To inductively derive themes from the data, de-identified transcripts of the audio recordings were used to conduct a content analysis. Two members of the research team conducted the thematic analysis using Excel to organize the data. Although the aim of this study is not to produce grounded theory, we used the technique of constant comparison to identify emerging themes in focus groups and interviews (70). After reviewing these codes, the researchers refined them by consolidating and sorting into broader themes. New codes were added, as needed. Once the analysis was completed, the research team met to develop a summative grid of the emerging themes. Researchers reviewed 273 decision points and found initial agreement of 65% across all decision points. Discussion further clarified code definitions and researchers consulted the original transcripts as needed on areas requiring discussion. Discussion resulted in 100% agreement for the qualitative analysis. The over-coding and review provide evidence that the qualitative analysis and presentation of findings accurately represent the voices of the WHO-CST participants. These processes were aimed at establishing trustworthiness of the data analysis and results.

TABLE 3 Facilitators-rated perceived feasibility of delivery of group session.

Session	*N*	Complexity of ideas				Amount of content for the time				Preparedness to conduct the session	
		Too simple		*Too advanced*		*Too little*		*Too much*		*Inadequate*	
		n	%	*n*	%	*n*	%	*n*	%	*n*	%
1	8	1	12.25	1	12.25	2	25	4	50	3	37.5
2	8	0	0	1	12.25	0	0	6	75	1	12.5
3	8	0	0	0	0	0	0	6	75	0	0
4	8	0	0	0	0	1	12.5	7	87.5	0	0
5	8	0	0	0	0	1	12.5	4	50	0	0
6	8	0	0	2	25	0	0	3	37.5	0	0
7	7	1	14.29	0	0	0	0	5	71.4	0	0
8	8	0	0	0	0	0	0	0	0	0	0
9	8	0	0	0	0	0	0	2	25	0	0

Results

Feasibility and acceptability outcomes

For this pilot study, feasibility outcomes were measured by parental attendance to WHO-CST sessions, attrition, caregiver's knowledge, and confidence in skills, WHO-CST Post Session Feedback form from Facilitators. Acceptability was measured by the WHO-CST Post Session Feedback form from Caregivers and Facilitators, as described in the measures section. Focus groups offered information for both feasibility and acceptability as well as recommendations to improve the program.

Feasibility

Attendance and attrition

The program included nine group-sessions and three home visits. Attendance rate for the group-sessions was 96.8%, and a 100% for the three home visits. None of the caregivers dropped out, attaining a 0-attrition rate for the online program.

Facilitators' ratings of feasibility of delivery of group sessions

Table 3 reports frequencies of ratings in the insufficient ($\leq$2) or excessive ($\geq$4) in the dimensions of complexity of ideas (content of sessions), amount of content for the allotted time, and preparedness to conduct the session according to the Facilitators' perception.

Caregivers' knowledge and confidence in skills test

As shown in Table 4, caregivers did not exhibit differences in the knowledge pre and posttest ($t = 0.121$, a $\geq$ 0.05), but they did exhibit differences in confidence scores ($t = 2.11$, a $\leq$ 0.05). There was no correlation between knowledge gained in the WHO-CST and having participated in previous training or having received similar information in the past.

Acceptability

Caregiver ratings of acceptability of group sessions

In general, caregivers considered the content somewhat easy, somewhat relevant, very useful and not in conflict with their values (Table 5). Only one parent reported that the content of session 6 (Preventing Challenging Behavior, Helping Children Stay Engaged and Regulated) was slightly in conflict with his/her values. Although the caregivers rated the duration of group sessions as "just right," the specific time allotted for sharing experiences and discussions was considered too short across sections but more frequently for the first three sessions (Table 6).

Facilitator ratings of perceived acceptability of group sessions to caregivers

Table 7 shows Facilitators ratings of perceived relevance and acceptability of the sessions' content for the caregivers, agreement with ideas presented during the sessions and caregiver's participation and engagement during the sessions. Frequencies reported are of unsatisfactory ratings ($\leq$ 3).

WHO-CST post session feedback form from caregivers

Caregivers' feedback included content's complexity, relevance, usefulness, and alignment with family values. In addition, it measured the parent's preparedness to practice the learned strategies at home. For all the dimensions assessed across the nine sessions, caregivers offered scores equal to or $>$ 3 (neutral), with no scores of 2 or 1 (unsatisfactory cuts off). Most of the caregivers' ratings of content's complexity (73%), relevance (66%), and usefulness (60%) were greater than or equal than 4 (somewhat easy) for all nine sessions. The most liked

TABLE 4 Changes in clinical measures: Child and caregivers outcomes.

	Baseline		End of program		Baseline to end of program mean difference (SE)	Paired t-test
	Mean	SD	Mean	SD		
Child outcomes						
ATEC						
Communication	17.57	5.14	14.14	9.07	3.43 (4.40)	2.92*
Sociability	16.54	4.67	14.64	6.91	2.00 (5.97)	1.25
Sensory/Cognitive	19.64	6.63	20.50	7.40	−0.86 (3.48)	−0.92
Health/Physical	24.79	9.10	26.36	11.31	−1.57 (11.63)	−0.506
Total	78.64	12.16	75.64	11.32	3.00 (14.09)	0.80
Autism Impact Measure (AIM)						
Frequency						
Communication	19.86	3.98	17.21	4.54	2.64 (1.04)	2.54*
Repetitive behavior	22.21	5.95	22.14	6.53	0.07 (5.84)	0.46
Social reciprocity	15.29	4.25	15.43	4.03	−0.14 (3.86)	−0.14
Peer interaction	10.79	3.33	11.00	2.22	−0.21 (2.72)	−0.29
Atypical behavior	16.21	4.02	15.50	4.97	0.71 (4.94)	0.54
Total	116.5	16.25	111.14	15.89	5.36 (5.52)	1.23
Impact						
Communication	18.79	5.51	14.31	5.17	4.46 (2.88)	5.59**
Repetitive behavior	15.00	5.11	14.69	6.60	0.31 (5.26)	3.49
Social reciprocity	10.86	4.20	11.14	4.55	−0.29 (4.23)	−0.253
Peer interaction	9.54	5.11	8.08	3.57	1.46 (3.48)	1.52
Atypical behavior	18.00	5.45	12.61	5.12	5.38 (5.52)	3.52**
Total	95.46	29.09	82.15	30.52	13.31 (20.36)	2.36*
Caregiver outcomes						
Brief family distress scale	2.86	1.29	2.50	1.34	0.36 (1.15)	1.16
Parental stress scale	62.29	4.85	63.07	4.01	−0.79 (9.36)	−0.58
Kessler-psychological distress	25.36	3.37	24.86	2.38	0.50 (1.52)	0.82
Caregivers knowledge	82.43	3.41	82.29	2.95	0.14 (4.40)	0.12
Caregivers confidence in skills	46.79	9.06	52.07	7.23	−5.28 (9.36)	−2.11*
Parental sense of confidence	34.14	5.60	37.29	5.70	−3.14 (5.08)	−2.31*

*$p < 0.05$. **$p < 0.01$.

activity throughout the program was the demonstration (36%), and the least liked was the practice with other caregivers.

Focus groups: Feasibility and acceptability

All Master Trainers were interviewed ($n = 4$). Although all caregivers were invited, 10 out of 16 participated in the focus groups. There were between four and six participants in each group, which is consistent with recommended sizes and availability of the participants (71). The thematic analysis of the focus groups with participant caregivers and Master Trainers identified four main themes: (1) Changes resulting from the CST, (2) beneficial aspects of CST, (3) advantages and disadvantages of the online format, and (4) challenges to implementing WHO-CST *via* telehealth (Table 8). Such themes were further qualified according to feasibility, acceptability, and suggestions following the method used by Salomone et al. (40). Excerpts presented are from various participants [caregivers (CST) and Master Trainers (MT)] and in that way present a broad range of experiences and meanings to illustrate the themes and subthemes of this sample.

Changes resulting from the CST. Both the caregivers and Master Trainers discussed changes that the caregiver or child had made due to the program including changed thinking, behavior, and attitudes. For example, noticing less miscommunication between caregiver and child. All Master Trainers felt that the program had benefited

TABLE 5 Caregivers' rated acceptability of contents of the sessions.

Session	N	Difficulty[a]		Relevance[b]		Usefulness[c]		Alignment with values[d]	
		n	%	n	%	n	%	n	%
1	13	0	0	1	7.7	0	0	0	0
2	15	0	0	0	0	1	6.7	0	0
3	7	0	0	0	0	0	0	0	0
4	12	0	0	0	0	0	0	0	0
5	12	0	0	0	0	1	8.3	0	0
6	11	0	0	0	0	0	0	1	9.1
7	12	0	0	0	0	0	0	0	0
8	11	0	0	0	0	0	0	0	0
9	8	0	0	0	0	0	0	0	0

[a] ≤ Neither easy nor difficult.
[b] ≤ Neither relevant nor irrelevant.
[c] ≤ Neither useful nor useless.
[d] ≥ Neither agree nor disagree-there were neither conflict nor not in conflict.

TABLE 6 Caregivers-rated acceptability of the duration of the different activities of groups sessions.

Session	N	Group sessions				Sharing experiences/discussion				Practice in pairs			
		Too long		Too short		Too long		Too short		Too long		Too short	
		n	%	n	%	n	%	n	%	n	%	n	%
1	13	1	7.7	0	0	0	0	4	30.7	0	0	2	15.4
2	15	1	6.7	0	0	1	6.7	5	33.3	0	0	2	13.3
3	7	0	0	2		0	0	3	43	0	0	1	14.3
4	12	0	0	0	0	0	0	2	16.7	0	0	0	0
5	12	0	0	0	0	0	0	2	16.7	0	0	0	0
6	11	0	0	0	0	0	0	2	18.2	0	0	1	9.1
7	12	1	8.3	0	0	0	0	0	0	0	0	0	0
8	11	0	0	0	0	0	0	0	0	0	0	0	0
9	8	0	0	0	0	0	0	0	0	0	0	0	0

participants and they could see real changes. The families were engaged and implemented the program into their family routines.

> *One of the most positive things was just the end result with the families. Once we implemented it with them and got their feedback and saw how comfortable they were with the information and thankful they were probably the most positive thing that we experienced. (MT,2)*

Some caregivers began to feel more confident in their parenting. Others realized they have an active role in communicating with their child and that they can make changes to their own behaviors that could make a difference in the quality of communication with their child. Caregivers adjusted expectations, started using visuals when they hadn't before, and started paying more attention to "the small things." One parent mentioned shifting their focus from changing behavior to engaging with their child. Changes in children included fewer tantrums, positive reactions to strategies, increased independence for the child, and increased self-expression.

> *I've never really paid attention to how much she may have noticed me putting up the groceries or fixing her food or we definitely didn't play games... I'm not sure if she was able to play games, but just doing those simple engagements; it helped her to be more independent, to speak more independently: "Can we go outside now?", "Can I have a sandwich?" Just by engaging her a little bit more than, "Okay, are you hungry?" And giving her choices definitely would. Giving her choices instead of saying, "Okay, we're going to have lunch." (CST2,14)*

TABLE 7 Facilitator-rated perceived acceptability of the group sessions to caregivers.

Session	*N*	Relevance[a]		Acceptability[b]		Agreement[c]		Participation[d]	
		n	%	*n*	%	*n*	%	*n*	%
1	8	0	0	0	0	1	12.5	3	37.5
2	8	0	0	2	25	3	37.5	5	71.4
3	8	0	0	0	0	0	0	1	12.5
4	8	0	0	0	0	0	0	1	12.5
5	8	0	0	0	0	0	0	2	25
6	8	0	0	0	0	0	0	1	12.5
7	7	0	0	0	0	0	0	2	25
8	8	1	12.5	0	0	0	0	1	12.5
9	8	1	12.5	0	0	1	12.5	3	37.5

[a] ≤ Somewhat relevant to most participants.
[b] ≤ A few ideas are not acceptable.
[c] ≤ Did not express positive or negative opinions about the material.
[d] ≤ Responded and participated only when prompted.

TABLE 8 Feasibility, acceptability, and suggestions: themes developed from focus groups with caregivers and Master Trainers.

Domain	Themes			
	Changes resulting from the WHO-CST	*Beneficial Aspects of WHO-CST*	*Advantages and disadvantages of the online format*	*Challenges to Implementing WHO-CST via Telehealth*
Acceptability	Parents increased their confidence in their parenting skills. Changes in children included fewer tantrums, positive reactions to strategies, increased independence for the child, and increased self-expression.	The information is accessible, easy to use, and language was easy to receive. Most useful aspects were visual strategies (i.e., Thermometer), giving the child choices, and breathing exercises. The discussion portion of the group sessions and the Home Visits were the most useful aspects of the program.	Participants thought the online format was convenient. The virtual format made the program accessible to caregivers. Allows participation of caregivers from all over the state.	Too much content to cover in Home Visit 1.
Feasibility	Families were engaged and implemented the program into their family routines.	Strategies easy to follow. Learning from and connecting with other families. Some topics were too simple.	Trade-off between community building and accessibility was worth it. Less opportunity to build rapport. Parents seemed to be more focused on getting the content than in sharing with one another. Poor internet connection.	Maintain and practice the learned strategies. Topics or suggestions no applicable to their child. Model working with a child in an online format.
Suggestions	Add information that would provide a longer-term perspective; information that would help them know what they might expect down the road. Incorporate a planned follow up.	Update some of the examples to be more relevant to the US context. More coaching time for caregivers. Add information about how to engage schools and how to modify environments for learning–especially IEPS.	Start Facilitators' training in-person and give them a chance to practice in-person with families.	Create a video library as a resource to show an example of the content. Splitting up the first home visit into two visits. Guidance on training Facilitators.

According to most caregivers, the program would benefit from having information about how to engage schools and how to modify environments for learning–especially IEPs.

> *Maybe to give us more information of how to address some issues that you may run into with having your child in the educational environment, how to address a teacher or school. I would have, liked more information about that. I would have liked to maybe have a small message with tips of how to maybe help your child to modify themselves for that type of environment of school when you cannot be there. (CST2,44)*

Master Trainers suggested the incorporation of a planned follow-up, which could be in the form of monthly group check-in meetings or perhaps visits. This was in response to MTs observations that caregivers seemed to "get it" during their second home-visit, but often reverted to previous habits during the third visit. It seemed caregivers would benefit from sustained support to turn learned strategies into lasting changes in interactions with their children.

> *Some more home visits or a different structure. I don't know exactly what, but you know, more, face-to-face more coaching, more time doing that. I think our parents didn't, I mean, they didn't want the program to end. . . . All our families were working families and they got home, and they got dinner started and they jumped on our call and like had a lot of things going on, but they still like wanted to be there and didn't want it to end. So, there's some kind of better fade out maybe. . . we were like "there -adios". That's always an uncomfortable transition. So maybe at the end there could be some different types of transition. (MT,43)*

Beneficial aspects of CST. Caregivers spoke positively about WHO-CST content, noting the information was accessible, easy to use, and language was easy to receive. Participants felt the program helped them understand and communicate with their children. They stated connecting with other families that are dealing with the same things was important to them and especially appreciated being able to talk to people who can understand what they are experiencing without judgment. This was mentioned by both caregivers as well as Master Trainers:

> *Hearing from other families that are having you know the same kind of issues that you are makes you feel less isolated, and you know, just, it, it feels good to make connections with people who can understand what you're going through. (CST1,10)*

Master Trainers also found communication techniques to be the most useful strategy for caregivers. In addition, they mentioned how caregivers took advantage of learning new techniques, or even revisited things that they had seen before but hadn't tried.

> *I think... their communication with the parents, the caregivers (was very important); understanding that they have a role in that. It's not just about like their child asking them or telling them, but it's also about how they set the foundation upon which they can do that. Whether it's the way they communicate, the supports, environmental supports they're providing those kinds of things. (MT,38)*

In addition, all the caregivers felt like the discussion portion of the group trainings was the most useful aspect of the program. The discussions helped caregivers think through the strategies they were learning. Caregivers appreciated getting others' feedback (including Facilitators) on the weekly plans they developed, including hearing about others' experiences. They especially liked hearing what worked and didn't work with other people's children. On the other hand, some caregivers felt that some training topics were a little simple, but they understood that people might be coming in with varying levels of education and experience on the issues.

> *So, I think that the most useful part of the program for me was the discussions that we had. I felt like the, the lessons were a little bit elementary, and I understand the need for that because, you know, the people that you're working with, aren't always at the same educational level and things, but I personally found the lessons kind of like, okay, this is what we're doing, but the discussion afterward related to those subjects was really good. (CST,39)*

Caregivers frequently mentioned virtual home visits as a key component of the WHO-CST experience and expressed that these visits provided an opportunity to get one-on-one attention and ask questions specific to their child and their needs as a caregiver.

> *It gave you an opportunity to have that one-on-one where somebody could help you understand, that (you) might have a question that you don't want to ask in front of a group of people. And so, it made it feel like a safer space in regard to, if you had something that necessarily you didn't want to share...With the larger group. (CST2,35)*

Master Trainers were also largely supportive of the content. They did not suggest removing anything form the program but updating some of the examples to be more relevant to the American context. An example might be changing the images in the participants' booklets.

> *I think because of the picture, the illustrations, there's only so many different directions we could take it.... we're not washing our hands in a water basin in the United States. So,*

> *like, that's kind of like, "Oh, okay. That's interesting". I mean, some of the examples we could switch up, some of them we couldn't. And it's not to say it's bad or wrong, but some of those things would be helpful I think, to be updated. (MT,46)*

Master Trainers also made suggestions regarding the training of Facilitators.

> *I do wish that there was more guidance on how to properly train the Facilitator. Cause I felt like we were just kind of treading water to figure out what it looks like, reading something there, there wasn't really guidelines on what it should look like, what they need to know, what they don't need to know. So, it was just kind of thrown at us. (MT,31)*

The other recurrent suggestion was to incorporate more coaching to caregivers and maybe more virtual home visits

> *So, I think a lot of coaching during that time or more home visits. What I observed was, you know, you had the initial visit where they didn't know anything. You had the midway through visit, and you could see all these changes. And then you have a last home visit. And it was like, all the parents totally forgot what you taught them. It was really interesting. It was like the program's done. And then we came a week or two later and they weren't in it anymore. So, I thought that was really interesting that I did not see it, but when they were in the group, it was phenomenal. So, I think more coaching is what I would like to see. (MT,40)*

Advantages and disadvantages of the online format. Participants felt the virtual format made the program accessible to them. They could participate in a comfortable space, not have to travel, or not need to find childcare. Most participants felt like the trade-off between community building and accessibility was worth it.

> *I really enjoyed it. I was able to fit it in with all the other services and everything else that we have going on. It was a lot more convenient to schedule. I didn't have to be at a specific location, you know, on top of time travel and all that. It was a lot easier to work into my schedule. (CST1,30)*
>
> *I feel like I was able to fit [it in]. I'm a stay-at-home mom. So, I was able to make every session versus probably having to be like, 'I can't find a babysitter', especially during these times. So, I think that worked well. (CST2,25)*

Master Trainers indicated that the Zoom format allowed people from all over the state to come together, so it is possible for people to participate regardless of their location relative to program Facilitators.

> *I think the advantages is that like our group, we have families from all across the state, you know, so I live up in northern Missouri, and originally that was kind of the recruitment area. And I think we only ended up maybe having one or maybe two out of five families that were actually in that county coverage area. And so, they just kind of picked what day of the week worked best for them. So that was the beauty of the tele-health portion. (MT,30)*

In general participants thought the online format was *convenient*. The only down sides were the lack of human contact, and some tech issues. The main disadvantage discussed by both groups was less opportunity to build rapport.

> *If you were actually meeting in person, it would allow you the opportunity to possibly meet somebody with a child with that's the same age and possibly create a friendship out of that. But I mean, that's not even necessarily something that wouldn't happen (online), but I find that when you have a child that you have, you know, different, differently-abled, it's harder to come by those kinds of relationships at times. So that's something that would be positive from that experience, and they can relate. (CST2,28)*

Other issues included technology issues which were mainly problems with internet connections, some awkwardness in interactions such as lulls because people weren't sure who was talking or who might want to go next and dealing with some additional distractions when participating from home.

> *Sometimes when it would cut out, it was challenging, but my main thing was on the virtual. I don't even know how the classroom setting would work either, but like pinpointing a specific person to talk because it kind of creates a lot of lull time when everybody's just waiting to see you. And then if you have somebody who's more interested in, you know, communicating everything, you know, it causes other people not to have their perspective looked at and then something that could actually help everybody is missing out on. (CST2,19)*

Challenges to implementing WHO-CST via telehealth. At least three caregivers mentioned at some point that there were topics or suggestions that were not applicable to their child, since they thought the suggestion would not work with their children.

> *I think sometimes there were some suggestions that I was like, (my son) is never going to do that. Nope. He's not going to cooperate with that. And, you know, I mean, we all know our kids best, so it's worth a try, you know, of course. And then you're like, well, I, I tried it, it did not work and we're going to move on and try something else. Hmm. (CST1,25)*

Participants also found it challenging to maintain and practice the strategies they were taught.

> *The only hardest thing was actually finding time to remove all objects out of your area to find time to initiate the engagement for her. That was my still kind of is my biggest problem, finding the time to create engaging time with her. And I'm still kind of looking at her behavior to kind of build and modify some of the messages and tips that were, was given throughout the program. But I think that's still, like my biggest hurdle is finding time to engage and utilize and put the skills to work. (CST2,40)*

Master Trainers mentioned that feedback to Facilitators is not built into how the WHO-CST runs yet. They did not feel this was covered in their Master Trainer training. Most felt coaching Facilitators over Zoom was sufficient, but it was challenging to address ongoing problems. It was also difficult to model working with a child *via* Zoom.

> *I had a difficult experience with my Facilitator because I felt like I needed to give her feedback about how she presented and things like that. And that's not really built into the way that WHO-CST is run yet when we're helping to support those Facilitators. So, adding a piece to help Master Trainers do that would be nice to see. (MT,8)*

Although caregivers and Master Trainers considered virtual home visits as one of the most helpful components of the program, Master Trainers felt that the one-first home visit model meant there was a lot for caregivers to digest in one visit. There was not enough time to build rapport, so doing observations felt somewhat awkward. Their suggestion was to focus on program information during the first visit and then do the parenting observation in the second visit. However, caregivers did not mention this as problematic from their point of view.

> *[The first visit] was just so much... I think part of that had to do with the recruiting piece because we don't know what they know and they don't know what they don't know, you know? So, it's like trying to figure out what information they have and what they need to expect. And, and that first like home visit, I mean, you have to, you know, we've all been on the receiving end of like coaching that first time you get coaching or someone's watching you and you're trying to, you know, be productive and, you know, effective in what you're doing. And I think families just, they naturally are very hesitant and embarrassed. (MT,25)*
>
> *I think splitting up the first home visit into two visits, I think would have just kind of like set them on a different path and like trajectory, because I feel like they were all, our group was pretty good about communicating with us and they seemed like they were getting the information, but I feel like, like it would have brought it to the next level if we really figured out how to build that rapport. (MT,26)*

Master Trainers also suggested creating a video library as a resource for next telehealth WHO-CST

> *In-person, we were able to like model examples and basically role play what it would look like when working with the child. Whereas on Zoom, it wasn't that easy and there also wasn't videos available. So, that would be something I would suggest for telehealth is if they could have a video library ready to go for the next go around that Master Trainers can use to show an example of that, that material that we just went over. (MT,22)*

Changes in clinical measures

Testing of changes on clinical measures focused on parental knowledge about ASD, parental stress, self-efficacy, and parental reports of child changes as measured by validated instruments.

Child outcomes

From baseline to week 12, ATEC communication scores decreased from 17.57 (SD 5.14) from baseline to 14.14 (SD 9.07) at week 12 ($t = 2.92$, $p < 0.01$). Communication scores also improved according to the AIM, in both frequency ($t = 2.54$, $p < 0.05$) and impact ($t = 5.59$, $p < 0.01$). For the frequency communication domain, the scores decreased from 19.86 (SD: 3.98) at baseline to 17.21 (SD: 4.54) at week 12; while for the impact domain they went from 18.79 (SD: 5.51) to 14.31 (SD: 5.17). The AIM impact scores also showed a reduction between baseline and week 12 for atypical behavior ($t = 3.52$, $p < 0.01$) and the total impact score ($t = 2.36$, $p < 0.05$). Atypical behavior impact scores went from 18.00 at baseline to 12.61 at week 12; while the total impact scores decreased from 95.46 (SD 29.09) at baseline to 82.15 (SD: 30.52) at week 12.

Caregiver outcomes

Although there was a reduction in all the caregivers' measures, there were statistically significant differences from baseline and week 12 only for the caregiver's confidence in skills ($t = -2.11$, $p < 0.05$) and the parental sense of competence efficacy subscale ($t = 2.31$, $p < 0.05$). Parental sense of competence went from 34.14 (SD: 5.60) at baseline to 37.29 (SD: 5.70) at week 12, indicating an increase in parenting efficacy at the end of the program. Regarding caregivers' confidence in skills, scores went from 46.79 (SD: 9.06) to 52.07 (SD: 7.23) showing an increased confidence in using the learned skills.

There were no significant differences for the brief family distress scales, the caregiver's knowledge, or the parental stress scale (Table 4).

Discussion

This study's primary aim was to evaluate the feasibility and acceptability of implementing the WHO-CST program *via* an online, live group format in rural Missouri. Globally, most research on PMI has been conducted and evaluated in high-income and high-resource settings, leaving out non-urban settings with limited access to services (4). Our results support the feasibility of implementing the WHO-CST program *via* telehealth in a US rural setting. Attendance has been referred to as one of the main barriers to PMI implementation. A zero-attrition rate, 96.8% attendance for the group sessions, and 100% for the four virtual home visits are evidence of the feasibility of implementing the WHO-CST in a telehealth format in a US rural setting. In addition, data collection of outcome measures over 12 weeks was 100%, supporting the feasibility of this format, too. The program was considered feasible by both caregivers and Master Trainers. They reported that the strategies were easy to follow, and that caregivers were engaged and incorporated the program into their family routines. Caregivers also valued the group meetings that allowed them to learn from and connect with other families.

The COVID-19 pandemic enlarged health disparities for ASD services in low-resourced areas (39). Telehealth has been rapidly growing, and the COVID-19 pandemic increased its use and acceptance and showed us that remote PMI was a possibility (47, 72). Even before the pandemic, some research showed that PMI *via* telehealth is an effective delivery modality for addressing core symptoms and challenging behaviors in autistic children (39). The benefits are seen in many aspects, including scheduling, costs, and better use of resources. Other PMI implementation studies have shown that participation barriers are parents' time and childcare for non-autistic children and transportation (38). For our research, the online format diminished such hurdles as parents expressed.

The telehealth format made the WHO-CST program accessible to rural caregivers, allowing participation of caregivers from across the state. According to reports from the caregivers, the drawback of this format is that it offered less opportunity to build rapport among the participants. In addition, technical issues like internet connectivity, screen freeze, and timing lags were seen as challenges; however, caregivers stated that the benefits outweighed the challenges. Telehealth could be a feasible alternative for those families who experience the most common barriers to accessing services. The remote model allowed families from remote rural places to join the sessions, making WHO-CST available where previously there were no services. Sengupta et al. (39) commented on the need to offer options for the in-home visits in settings where cultural or contextual barriers might hinder the physical home visits. Our results support the use of telehealth for such purposes.

Caregivers rated the program content and its activities as comprehensible, relevant to them, and aligned with their values across sessions and home visits, which supports its acceptability in the context of rural American Mid-West. Results from different measures show that the demonstration was the most liked activity, whereas practice with pairs was the least liked one. These are similar findings to those from Salomone et al. (40) in Italy's WHO-CST field trial. For this study, one of the adaptations was to use prerecorded video for the demonstration activities, so Facilitators could pause, rewind, or replay it to illustrate or explain the content. The practice in pairs activity was challenging to complete in this virtual format. Caregivers rated the time devoted for the pair's activities as too short. Thus, the combination of the remote effect and not enough time to complete the activity could have resulted in its dislike. Salomone et al. (40) reported similar findings in their in-person trial. Being an essential part of the training and a good learning strategy for non-specialists, role-playing activities need to be revised to find different formats to present them in a more engaging and timely fashion. Caregivers reported that one of the main strengths of the WHO-CST was the opportunity to share with other caregivers that experience the same concerns, making it clear that the interaction is not the barrier here but the way the activity is presented. Outside COVID-19 circumstances, remote WHO-CST would offer caregivers more options to access services. WHO-CST might constitute an added option in service providers' portfolios that could eventually reduce operational costs by pairing the remote format (online) with the WHO-CST curriculum, thus increasing the probability for attendance and positive caregiver and child outcomes. Facilitator ratings of the feasibility of remote delivery of group sessions showed that sessions 2 (Keeping Children Engaged), 3 (Helping Children Share Engagement in Play and Home Routines), 4 (Understanding Communication), and 7 (Teaching Alternatives to Challenging Behavior) had too much content to present in the time allotted for each of the sessions. However, caregivers rated the level of difficulty of such sessions as adequate or somewhat easy, and the length of the sessions as 'just right. The delivery method (online) might have placed a burden on Facilitators, making them feel they did not have enough time to present all the content. However, one of the adaptations made for this study was to reduce the length of the sessions to avoid participants' fatigue. During the focus groups, Facilitators recommended using videos to demonstrate the content or present them as examples. Such a modification might ease the load of Facilitators to deliver the content in the planned time. Likewise, the WHO-CST implementation in northern Italy showed that some sessions were also considered as too packed with information. Such results suggest the need to revise those sessions for future implementations. Also, alternative instructional strategies like videos or more guided role-playing might diminish caregivers and Facilitators burden.

The other feasibility measure was knowledge of the skills related to WHO-CST content. Caregivers did not show any difference in pre-WHO-CST knowledge and post CST. However, the sense of confidence significantly improved, implying that the WHO-CST program might have an influence on learning how to use the knowledge, and applying the learned knowledge. Knowledge might not have been new for caregivers but practicing the concepts during group sessions or virtual home visits might have given caregivers confidence in using the skills. The characteristics of our sample (42.9% had some college, 21.4% had a college degree, 78.6% had previously received information on similar topics) might have impacted the results. The social determinants of mental health framework proposes that the circumstances in which people live, and work shape their health outcomes (73). Social determinants include SES, education, physical environment, access to healthcare (74). The high baseline knowledge scores may also indicate that the instrument's questions were too easy for the participants. The questionnaire was designed to be administered in low resourced/low-and middle-income countries; hence, it might need to be adapted to reflect the characteristics of a more educated sample of caregivers. Even if most caregivers had already participated in similar training or educational sessions on autism and parenting, they found the program's content beneficial.

During the focus groups, caregivers and Facilitators considered virtual home visits an essential element of the program. In that way, we might see the impact of live coaching on caregivers, which is an added value of the WHO-CST compared with self-paced PMI (75). In addition, naturalistic interventions like CST, which embed learning and practice opportunities into the child's daily routines, increase skill generalization (19). Facilitators perceived that the information gathered in the first home visit was too much for the caregivers to process. They suggested splitting it in two, so the first home visit could be devoted to discussing program information and the second home visit could focus on observing the caregiver-child interaction. The adapted curriculum added a 15-min home visit for families after session 1 to review a goal-setting sheet and answer questions the family had regarding their first session. In that way, the Facilitators enhanced the opportunities to interact with caregivers, clarify the goals for each family, and answer questions the participants might have had. Facilitators indicated the benefit of including a planned follow-up, which could be in the form of monthly group check-in meetings or perhaps visits. They supported the suggestion by stating that caregivers might understand the content during the second visit, but that learning is not present during the third visit. They conclude that caregivers would benefit from sustained support to turn learned strategies into lasting changes in interactions with their children. Results from the Indian WHO-CST trial also suggested the need for ongoing support for parents to build their competency in implementing strategies in the form of booster sessions post-implementation (39).

Parents have expressed high levels of satisfaction with therapist-assisted telehealth interventions. Such interventions have been associated with acquiring knowledge of behavioral interventions strategies from such programs (76). In other studies, parents reported reduced stress and increased self-efficacy after participating in a telehealth PMI (19). Iadarola et al. (20) provided outcome data on caregiver stress and parenting sense of confidence. Parents in the treatment arm of the study reported greater improvement on the parenting sense of confidence (effect size 0.34) than parents in the parent education program (effect size 0.34). Conversely, Bradshaw et al. (77) showed that parents in the parent education program reported a significant reduction in parenting stress and increased parenting sense of competence. These results, aligned with ours, might suggest that participation in group parent training could be a valuable tool that could increase self-efficacy and reduce parental stress.

Regarding participants' suggestions to improve the program, caregivers proposed having sessions with content related to engaging their child's school and longer-term perspectives on autism to help them prepare for the future. In addition, Facilitators indicated that having a video library would be beneficial when presenting new content or demonstrating/teaching new skills to caregivers. Both Facilitators and caregivers suggested updating some of the examples to be more relevant to the American context. Specifically, they commented that session two content relative to witchcraft or demon possessions was not relevant for the type of families in this geographic location, mainly white American Midwest. According to the adaptation guidance, this information should be used to further adapt the program to the context in Missouri. Compared to the Salomone et al. (40, 41) study, the only published field trial in a high-income country, caregivers, and Facilitators voiced similar concerns with the caregivers' stories. For example, one of the Facilitators pointed out that people do not use a basin to wash their hands in the US, while a caregiver pointed out the absence of fathers in most of the stories. Such comments highlight the relevance and need for an adaptation process aligned with cultural practices and values in the community in which the WHO-CST will be implemented.

It is possible that caregiver stories may not require changes in other settings in which the story contents are a well-recognized and culturally relevant tool (38, 40). For the WHO-CST implementation in rural Missouri, the central adaptations were linguistic: changing British spelling to American English, as well as examples and names to have culturally and linguistically valid materials. Overall, caregivers identified implementation barriers related to maintenance and practice of the learned strategies and the use of some contents that did not apply to their child's developmental level. Facilitators referred to challenges in terms of the amount of content for the first home visit and how to model some WHO-CST strategies to the caregivers *via* an online format. Besides the provided examples, caregivers and facilitators deemed the adapted WHO-CST materials acceptable and relevant for the American rural Midwest.

Although not the primary aim of the study, analysis of clinical measures from our sample suggests that the efficacy of the WHO-CST program delivered *via* telehealth is promising. However, due to the design, the non-probabilistic sampling process, and other limitations of the study, these results need to be considered as preliminary. The significant results aligned with the primary outcomes of the program: provide caregivers with strategies to support their children's development by engaging children in everyday activities and applying strategies to support the development of the child's communication skills and reduce challenging behavior. Results showed a significant reduction in the AIM atypical behavior impact scores but not in the frequency, suggesting that the stigma experienced by parents comes from atypical behaviors (78). Using the skills learned during the WHO-CST program, parents might have learned how to redirect, manage, and perceive the behaviors. In addition, there was a significant decrease in the AIM Total impact scores, but not for the frequency. This change may represent how the WHO-CST might have impacted the way caregivers interpret behaviors. Children might still exhibit the same behavior, but caregivers are given a different meaning to such behaviors due to their participation in the WHO-CST program. Nevertheless, it is important to mention that some of the participants had received services, limiting our ability to make inferences about the factors impacting the clinical outcomes.

There were no significant changes for the social interaction measures. For families and autistic individuals' global events like the pandemic could add distress to an already complex scenario that might have impacted children's opportunities for social interaction. Additionally, the interruption of face-to-face schooling, leisure activities, and reduced access to services represented a disruption of routine, which might partly be responsible for the absence of changes in the social interaction area (79, 80). In summary, interventions deployed to control the spread of the COVID-19 pandemic such as social distancing and the use of face masks, may conflict with interventions aimed to improve the wellbeing of autistic children (81), such as the WHO-CST program. WHO-CST is a naturalistic behavioral intervention, and as such, implementation takes place during naturally occurring home and play routines requiring high levels of clinical judgment. In consequence, training of non-specialists might need more practice and coaching than more directive and structured interventions (82–84). Other studies have found that administrative support, the interactive nature of the training, and the compatibility of the training model with Facilitators' current practices facilitate the training process (54). Although due to the COVID-19 pandemic, our Facilitators received remote training; all Master Trainers and Facilitators had extensive training and experience with children with neurodevelopmental disorders and behavioral interventions. Their current activities included parent training sessions and in-home consultations. WHO-CST was designed to be implemented by non-specialists, but on our site, it was delivered by behavioral health workers with extensive experience in child development and autism; as was the case of Italy (40, 41) and India (39). Therefore, our results are limited to settings that use Facilitators with similar educational and experience backgrounds.

The results of this study need to be interpreted in light of its limitations. First, there was a small sample and no control group. However, there were four dyads of Master Trainer-Facilitator implementing the program independent of each other. Second, we did not have independent raters of children and caregivers' behaviors, which could have biased the results. Third, we used a mixture of qualitative and standardized quantitative self-report measures to evaluate changes in clinical outcomes rather than performing an independent objective examination of knowledge and skills. However, there is no consensus on what measures to use for the range of potentially relevant outcomes to evaluate intervention effectiveness for autistic children (85). Finally, all families were paid for participation in all assessments, impacting the attrition rate and data collection rates. In terms of Facilitator training, due to COVID-19, Facilitators completed their entire training *via* Zoom, preventing them from obtaining hands-on training with families, including the opportunity to see interactions with families. Master trainers agreed that some in-person training would be beneficial. However, the results from this study suggest that remote training for Facilitators might also be an option with some adaptations to the programmed practice. Other studies have found that telehealth is an effective way for training non-specialists in delivering PMIs, and also for providing supervision and coaching (42).

Albeit these limitations, this study has several strengths. It is the first implementation of a telehealth PMI conducted with non-specialists in a rural US setting; and could be generalizable to similar rural settings. Results from this study-an adapted parent-mediated intervention-could impact public policy by offering a scalable and sustainable program to bridge the gap between research and community implementation. Furthermore, this study innovates by (1) including a unique sampling framework to represent caregivers from an underserved rural setting, (2) using the ECHO Autism model to prepare WHO-CST group Facilitators in a cost effective and efficient manner while allowing for iterative guidance with WHO-CST implementation, and (3) applying a mixed methods design to inform the process. Results would serve as a baseline for future autism studies and service interventions with underserved rural families in other rural US areas. Our mixed-methods approach using focus groups ensures the adaptations made to the WHO-CST are relevant to rural families of autistic children.

Glasgow and Emmons (86) advocate using practical trials with both quantitative and qualitative methods to assess multiple outcomes relevant to community implementation since delivery of professional services is complex and more so in a naturalistic setting such as the presented trial. These preliminary results using a mix-methods design do not allow us to draw conclusions about the efficacy of the CST-online format. Nevertheless, these findings provide information about the feasibility of implementing the WHO-CST program *via* telehealth in an underserved rural setting, contributing to the global field trials of the program, and serving as preliminary data for a larger randomized control trial (RCT) to explore its efficacy. According to Bearss (42), remote PMI is an effective delivery modality for core symptoms and challenging behaviors in autistic children. In a systematic review and metanalysis, Deb et al. (87) urged experts to standardize a PMI for autistic children and carry out a large-scale RCT to assess its clinical and economic effectiveness. WHO-CST might be that tool since it: (a) uses evidence-based procedures, (b) is open access, (c) can be administered by non-specialists, (d) requires cultural and linguistic adaptation, (e) uses a community based-participatory framework increasing the odds for sustainability and scalability, (f) does not require a diagnosis, decreasing time for accessing services, (g) is a low dose-low intensity program, (i) could be used in both low-and-middle income and high income countries, and (j) has promising evidence of its effectiveness when used *via* telehealth.

For implementing a naturalistic intervention, such as WHO-CST, in a rural setting, several factors unrelated to the PMI need to be identified to increase the success of its implementation. Among such factors, engagement of community stakeholders and partnership with a specialized autism center is essential to offer support to Facilitators and participants from rural areas. In our case, both Easterseals Midwest and the ECHO Autism at the University of Missouri-Columbia provided the knowledge, supervision, and administrative capabilities to conduct the trial. In addition, ECHO Autism clinicians provided a healthcare network that allowed for the referral of families to the program. Lastly, a reliable internet connection for caregivers and service providers is needed, as well as engaging caregivers with technical knowledge to access the sessions' links and materials.

High-income-countries have many low-resource contexts, such as rural areas and the health professional shortage areas in the US (29), in which families have limited access to services. Although the source and degree of disparities might differ, low-resourced communities in high-income-countries share similar characteristics of low-and-middle-income-countries in terms of barriers accessing timely and evidence-based interventions and shortage of trained professionals to identify and treat children with neurodevelopmental conditions (88). In these instances, a PMI-such as the WHO-CST with cultural and linguistic adaptations and greater accessibility *via* telehealth-plays an essential role by closing the treatment gap and empowering caregivers of autistic children.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

All study procedures were approved by the University of Missouri-Columbia Institutional Review Board. The patients/participants provided their written informed consent to participate in this study.

Author contributions

AC, CM-N, KS, JM, MT, and PD contributed to conception and design of the study. AC, JM, MT, and MM collected the data. CM-N organized the database, performed the statistical analysis, and wrote the first draft of the manuscript. AC, MT, MW-G, and PD wrote sections of the manuscript. All authors contributed to manuscript revision, read, and approved the submitted version.

Funding

This project received funding from Autism Speaks.

Acknowledgments

WHO-CST Team Consortium Member List (in alphabetical order): Felicity L. Brown, Laura Pacione, Erica Salomone, Stephanie Shire, and Chiara Servili.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

References

1. American Psychiatric Association. *Diagnostic and statistical manual of mental disorders. 5th ed*. Arlington, VA: American Psychiatric Publishing (2013).

2. Bottema-Beutel K, Kapp SK, Lester JN, Sasson NJ, Hand BN. Avoiding ableist language: suggestions for autism researchers. *Autism Adulthood.* (2021) 3:18–29. doi: 10.1089/aut.2020.0014

3. Oono IP, Honey EJ, McConachie H. Parent-mediated early intervention for young children with autism spectrum disorders (ASD). *Evidence-Based Child Health: A Cochrane Review Journal.* (2013) 8:2380–479. doi: 10.1002/ebch.1952

4. Dawson-Squibb J-J, de Vries PJ. Developing an evaluation framework for parent education and training in autism spectrum disorder: results of a multi-stakeholder process. *J Autism Dev Disord.* (2019) 49:4468–81. doi: 10.1007/s10803-019-04176-w

5. Schreibman L, Dawson G, Stahmer AC, Landa R, Rogers SJ, McGee GG, et al. Naturalistic developmental behavioral interventions: empirically validated treatments for autism spectrum disorder. *J Autism Dev Disord.* (2015) 45:2411–28. doi: 10.1007/s10803-015-2407-8

6. Anderson SR, Romanczyk RG. Early intervention for young children with autism: continuum-based behavioral models. *J Assoc Pers Sev Handicaps.* (1999) 24:162–73. doi: 10.2511/rpsd.24.3.162

7. Koegel LK, Koegel RL, Harrower JK, Carter CM. Pivotal response intervention I: overview of approach. *J Assoc Pers Sev Handicaps.* (1999) 24:174–85. doi: 10.2511/rpsd.24.3.174

8. McGee GG, Morrier MJ, Daly T. An incidental teaching approach to early intervention for toddlers with autism. *J Assoc Pers Sev Handicaps.* (1999) 24:133–46. doi: 10.2511/rpsd.24.3.133

9. Nevill RE, Lecavalier L, Stratis EA. Meta-analysis of parent-mediated interventions for young children with autism spectrum disorder. *Autism.* (2018) 22:84–98. doi: 10.1177/1362361316677838

10. Sofronoff K, Leslie A, Brown W. Parent management training and Asperger syndrome: a randomized controlled trial to evaluate a parent based intervention. *Autism.* (2004) 8:301–17. doi: 10.1177/1362361304045215

11. Kasari C, Gulsrud AC, Wong C, Kwon S, Locke J. Randomized controlled caregiver mediated joint engagement intervention for toddlers with autism. *J Autism Dev Disord.* (2010) 40:1045–56. doi: 10.1007/s10803-010-0955-5

12. Vismara LA, Lyons GL. Using perseverative interests to elicit joint attention behaviors in young children with autism: theoretical and clinical implications for understanding motivation. *J Posit Behav Interv.* (2007) 9:214–28. doi: 10.1177/10983007070090040401

13. Harrop C. Evidence-based, parent-mediated interventions for young children with autism spectrum disorder: the case of restricted and repetitive behaviors. *Autism.* (2015) 19:662–72. doi: 10.1177/1362361314545685

14. Pickles A, Le Couteur A, Leadbitter K, Salomone E, Cole-Fletcher R, Tobin H, et al. Parent-mediated social communication therapy for young children with autism (PACT): long-term follow-up of a randomised controlled trial. *Lancet.* (2016) 388:2501–9. doi: 10.1016/S0140-6736(16)31229-6

15. Dawson G, Rogers S, Munson J, Smith M, Winter J, Greenson J, et al. Randomized, controlled trial of an intervention for toddlers with autism: the Early Start Denver Model. *Pediatrics.* (2010) 125:e17–23. doi: 10.1542/peds.2009-0958

16. Kroeger K, Sorensen R, A. parent training model for toilet training children with autism. *J Intellect Disabil Res.* (2010) 54:556–67. doi: 10.1111/j.1365-2788.2010.01286.x

17. Boyle CL, Sanders MR, Lutzker JR, Prinz RJ, Shapiro C, Whitaker DJ. An analysis of training, generalization, and maintenance effects of Primary Care Triple P for parents of preschool-aged children with disruptive behavior. *Child Psychiatry Hum Dev.* (2010) 41:114–31. doi: 10.1007/s10578-009-0156-7

18. Solish A, Perry A. Parents' involvement in their children's behavioral intervention programs: parent and therapist perspectives. *Res Autism Spectr Disord.* (2008) 2:728–38. doi: 10.1016/j.rasd.2008.03.001

19. Dai YG, Thomas RP, Brennan L, Helt MS, Barton ML, Dumont-Mathieu T, et al. Development and acceptability of a new program for caregivers of children with autism spectrum disorder: online parent training in early behavioral intervention. *J Autism Dev Disord.* (2021) 51:1–20. doi: 10.1007/s10803-020-04863-z

20. Iadarola S, Levato L, Harrison B, Smith T, Lecavalier L, Johnson C, et al. Teaching parents behavioral strategies for autism spectrum disorder (ASD): effects on stress, strain, and competence. *J Autism Dev Disord.* (2018) 48:1031–40. doi: 10.1007/s10803-017-3339-2

21. Casagrande KA, Ingersoll BR. Service delivery outcomes in ASD: role of parent education, empowerment, and professional partnerships. *J Child Fam Stud.* (2017) 26:2386–95. doi: 10.1007/s10826-017-0759-8

22. United S. *Individuals with Disabilities Education Improvement Act of 2004.* Bethesda, MD: ProQuest (2011).

23. McDonald CA, Donnelly JP, Feldman-Alguire AL, Rodgers JD, Lopata C, Thomeer ML. Special education service use by children with autism spectrum disorder. *J Autism Dev Disord.* (2019) 49:2437–46. doi: 10.1007/s10803-019-03997-z

24. Maenner MJ, Shaw KA, Baio J. Prevalence of autism spectrum disorder among children aged 8 years—autism and developmental disabilities monitoring network, 11 sites, United States, 2016. *MMWR Surveillance Summaries.* (2020) 69:1. doi: 10.15585/mmwr.ss6904a1

25. Zeidan J, Fombonne E, Scorah J, Ibrahim A, Durkin MS, Saxena S, et al. Global prevalence of autism: a systematic review update. *Autism Res.* (2022) 15:778–90. doi: 10.1002/aur.2696

26. Stahmer AC, Dababnah S, Rieth SR. Considerations in implementing evidence-based early autism spectrum disorder interventions in community settings. *Pediatr Med.* (2019) 2:18. doi: 10.21037/pm.2019.05.01

27. Kohlhoff J, Cibralic S, Horswood D, Turnell A, Maiuolo M, Morgan S. Feasibility and acceptability of internet-delivered parent-child interaction therapy for rural Australian families: a qualitative investigation. *Rural Remote Health.* (2020) 20:5306. doi: 10.22605/RRH5306

28. Ratcliffe M, Burd C, Holder K, Fields A. Defining rural at the US Census Bureau. *American community survey and geography brief.* (2016) 1:1–8. Available online at: https://www2.census.gov/geo/pdfs/reference/ua/Defining_Rural.pdf

29. Administration HRaS. Shortage areas (2021). Available online at: https://data.hrsa.gov/topics/health-workforce/shortage-areas

30. Hoogsteen L, Woodgate RL. Embracing autism in C anadian rural communities. *Aust J Rural Health.* (2013) 21:178–82. doi: 10.1111/ajr.12030

31. Hub RHI. Health Professional Shortage Areas: Mental Health, by County, 2021-Nonmetropolitan (2021). Available online at: https://www.ruralhealthinfo.org/charts/7?state=MO

32. Cason J. A pilot telerehabilitation program: delivering early intervention services to rural families. *Int J Telerehabilitation.* (2009) 1:29. doi: 10.5195/ijt.2009.6007

33. Olsen S, Fiechtl B, Rule S. An evaluation of virtual home visits in early intervention: feasibility of "virtual intervention". *Volta Rev.* (2012) 112:267. doi: 10.17955/tvr.112.3.m.702

34. Compton MT, Shim RS. The social determinants of mental health. *Focus.* (2015) 13:419–25. doi: 10.1176/appi.focus.20150017

35. Salomone E, Pacione L, Shire S, Brown FL, Reichow B, Servili C. Development of the WHO caregiver skills training program for developmental disorders or delays. *Front Psychiatry.* (2019) 10:769. doi: 10.3389/fpsyt.2019.00769

36. Hamdani S, Akhtar P, Nazir H, Minhas F, Sikander S, Wang D, et al. WHO parents skills training (PST) programme for children with developmental disorders and delays delivered by Family Volunteers in rural Pakistan: study protocol for effectiveness implementation hybrid cluster randomized controlled trial. *Glob Ment Health.* (2017) 4:e11. doi: 10.1017/gmh.2017.7

37. Salomone E, Reichow B, Pacione L, Shire S, Shih A, Servili C. Training caregivers to transform children's lives. *Early Child Matters.* (2018) 127:74–7. Available online at: https://earlychildhoodmatters.online/2018/training-caregivers-to-transform-childrens-lives

38. Tekola B, Girma F, Kinfe M, Abdurahman R, Tesfaye M, Yenus Z, et al. Adapting and pre-testing the World Health Organization's Caregiver Skills Training programme for autism and other developmental disorders in a very low-resource setting: findings from Ethiopia. *Autism.* (2020) 24:51–63. doi: 10.1177/1362361319848532

39. Sengupta K, Shah H, Ghosh S, Sanghvi D, Mahadik S, Dani A, et al. World Health Organisation-Caregiver Skills Training (WHO-CST) program: feasibility of delivery by non-specialist providers in real-world urban settings in India. *J Autism Dev Disord.* (2021) 1–18. doi: 10.1007/s10803-021-05367-0

40. Salomone E, Ferrante C, Salandin A, Ferrara F, Torchio E, Foletti G, et al. Acceptability and feasibility of the World Health Organization's caregiver skills training implemented in the Italian national health system. *Autism.* (2022) 26:859–74. doi: 10.1177/13623613211035228

41. Salomone E, Settanni M, McConachie H, Suma K, Ferrara F, Foletti G, et al. Pilot randomized controlled trial of the WHO caregiver skills

training in public health services in Italy. *J Autism Dev Disord.* (2021) 1–15. doi: 10.1007/s10803-021-05297-x

42. Bearss K, Burrell TL, Challa SA, Postorino V, Gillespie SE, Crooks C, et al. Feasibility of parent training *via* telehealth for children with autism spectrum disorder and disruptive behavior: a demonstration pilot. *J Autism Dev Disord.* (2018) 48:1020–30. doi: 10.1007/s10803-017-3363-2

43. Chi N-C, Demiris G. A systematic review of telehealth tools and interventions to support family caregivers. *J Telemed Telecare.* (2015) 21:37–44. doi: 10.1177/1357633X14562734

44. Ellison KS, Guidry J, Picou P, Adenuga P, Davis TE. Telehealth and autism prior to and in the age of COVID-19: a systematic and critical review of the last decade. *Clin Child Fam Psychol Rev.* (2021) 24:1–32. doi: 10.1007/s10567-021-00358-0

45. Vismara LA, McCormick CE, Wagner AL, Monlux K, Nadhan A, Young GS. Telehealth parent training in the early start denver model: results from a randomized controlled study. *Focus Autism Other Dev Disabl.* (2018) 33:67–79. doi: 10.1177/1088357616651064

46. Suess AN, Romani PW, Wacker DP, Dyson SM, Kuhle JL, Lee JF, et al. Evaluating the treatment fidelity of parents who conduct in-home functional communication training with coaching via telehealth. *J Behav Educ.* (2014) 23:34–59. doi: 10.1007/s10864-013-9183-3

47. Wacker DP, Lee JF, Dalmau YCP, Kopelman TG, Lindgren SD, Kuhle J, et al. Conducting functional communication training via telehealth to reduce the problem behavior of young children with autism. *J Dev Phys Disabil.* (2013) 25:35–48. doi: 10.1007/s10882-012-9314-0

48. Ingersoll B, Wainer AL, Berger NI, Pickard KE, Bonter N. Comparison of a self-directed and therapist-assisted telehealth parent-mediated intervention for children with ASD: a pilot RCT. *J Autism Dev Disord.* (2016) 46:2275–84. doi: 10.1007/s10803-016-2755-z

49. Kuravackel GM, Ruble LA, Reese RJ, Ables AP, Rodgers AD, Toland MD. Compass for hope: evaluating the effectiveness of a parent training and support program for children with ASD. *J Autism Dev Disord.* (2018) 48:404–16. doi: 10.1007/s10803-017-3333-8

50. Gerow S, Radhakrishnan S, Akers JS, McGinnis K, Swensson R. Telehealth parent coaching to improve daily living skills for children with ASD. *J Appl Behav Anal.* (2021) 54:566–81. doi: 10.1002/jaba.813

51. Akamoglu Y, Meadan H. Parent-implemented communication strategies during storybook reading. *J Early Interv.* (2019) 41:300–20. doi: 10.1177/1053815119855007

52. Burrell AJ, Pellegrini B, Salimi F, Begum H, Broadley T, Campbell LT, et al. Outcomes for patients with COVID-19 admitted to Australian intensive care units during the first four months of the pandemic. *Med J Aust.* (2021) 214:23–30. doi: 10.5694/mja2.50883

53. Vismara LA, McCormick C, Young GS, Nadhan A, Monlux K. Preliminary findings of a telehealth approach to parent training in autism. *J Autism Dev Disord.* (2013) 43:2953–69. doi: 10.1007/s10803-013-1841-8

54. Wainer AL, Arnold ZE, Leonczyk C, Soorya LV. Examining a stepped-care telehealth program for parents of young children with autism: a proof-of-concept trial. *Mol Autism.* (2021) 12:1–14. doi: 10.1186/s13229-021-00443-9

55. Hao Y, Franco JH, Sundarrajan M, Chen Y, A. pilot study comparing tele-therapy and in-person therapy: perspectives from parent-mediated intervention for children with autism spectrum disorders. *J Autism Dev Disord.* (2021) 51:129–43. doi: 10.1007/s10803-020-04439-x

56. Tsami L, Lerman D, Toper-Korkmaz O. Effectiveness and acceptability of parent training via telehealth among families around the world. *J Appl Behav Anal.* (2019) 52:1113–29. doi: 10.1002/jaba.645

57. Harris PA, Taylor R, Minor BL, Elliott V, Fernandez M, O'Neal L, et al. The REDCap consortium: building an international community of software platform partners. *J Biomed Inform.* (2019) 95:103208. doi: 10.1016/j.jbi.2019.103208

58. Mazurek MO, Brown R, Curran A, Sohl K. ECHO autism: a new model for training primary care providers in best-practice care for children with autism. *Clin Pediatr.* (2017) 56:247–56. doi: 10.1177/0009922816648288

59. Sohl K, Mazurek MO, Brown R. ECHO autism: using technology and mentorship to bridge gaps, increase access to care, and bring best practice autism care to primary care. *Clin Pediatr.* (2017) 56:509–11. doi: 10.1177/0009922817691825

60. Rimland B, Edelson M. *The Autism Treatment Evaluation Checklist (ATEC).* San Diego, CA: Autism Research Institute. Available online at: https://www.autism.org/autism-treatment-evaluation-checklist/

61. Magiati I, Moss J, Yates R, Charman T, Howlin P. Is the autism treatment evaluation checklist a useful tool for monitoring progress in children with autism spectrum disorders? *J Intellect Disabil Res.* (2011) 55:302–12. doi: 10.1111/j.1365-2788.2010.01359.x

62. Memari AH, Shayestehfar M, Mirfazeli F-S, Rashidi T, Ghanouni P, Hafizi S. Cross-cultural adaptation, reliability, and validity of the autism treatment evaluation checklist in Persian. *Iran J Pediatr.* (2013) 23:269. Available online at: https://brieflands.com/articles/ijp-1655.pdf

63. Kanne SM, Mazurek MO, Sikora D, Bellando J, Branum-Martin L, Handen B, et al. The Autism Impact Measure (AIM): initial development of a new tool for treatment outcome measurement. *J Autism Dev Disord.* (2014) 44:168–79. doi: 10.1007/s10803-013-1862-3

64. Mazurek MO, Carlson C, Baker-Ericzén M, Butter E, Norris M, Barr C, et al. The autism impact measure (AIM): examination of sensitivity to change. *Autism Res.* (2020) 13:1867–79. doi: 10.1002/aur.2397

65. Weiss JA, Lunsky Y. The brief family distress scale: A measure of crisis in caregivers of individuals with autism spectrum disorders. *J Child Fam Stud.* (2011) 20:521–8. doi: 10.1007/s10826-010-9419-y

66. Kessler RC, Green JG, Gruber MJ, Sampson NA, Bromet E, Cuitan M, et al. Screening for serious mental illness in the general population with the K6 screening scale: results from the WHO World Mental Health (WMH) survey initiative. *Int J Methods Psychiatr Res.* (2010) 19:4–22. doi: 10.1002/mpr.310

67. Prochaska JJ, Sung HY, Max W, Shi Y, Ong M. Validity study of the K6 scale as a measure of moderate mental distress based on mental health treatment need and utilization. *Int J Methods Psychiatr Res.* (2012) 21:88–97. doi: 10.1002/mpr.1349

68. Berry JO, Jones WH. The parental stress scale: Initial psychometric evidence. *J Soc Pers Relat.* (1995) 12:463–72. doi: 10.1177/0265407595123009

69. Johnston C, Mash EJ, A. measure of parenting satisfaction and efficacy. *J Clin Child Psychol.* (1989) 18:167–75. doi: 10.1207/s15374424jccp1802_8

70. Corbin J, Strauss A. *Basics of qualitative research: Techniques and procedures for developing grounded theory*. Sage publications (2014).

71. Krueger RA, Casey MA. Focus group interviewing. Handbook of practical program evaluation. 2015:506–34. doi: 10.1002/9781119171386.ch20

72. Parsons D, Cordier R, Vaz S, Lee HC. Parent-mediated intervention training delivered remotely for children with autism spectrum disorder living outside of urban areas: systematic review. *J Med Internet Res.* (2017) 19:e6651. doi: 10.2196/jmir.6651

73. Marmot M, Bell R. Social inequalities in health: a proper concern of epidemiology. *Ann Epidemiol.* (2016) 26:238–40. doi: 10.1016/j.annepidem.2016.02.003

74. World health Organization. *Social determinants of health.* (2021). Available online at: https://www.who.int/health-topics/social-determinants-of-health#tab=tab_1

75. Tomeny KR, McWilliam R, Tomeny TS. Caregiver-implemented intervention for young children with autism spectrum disorder: a systematic review of coaching components. *Rev J Autism Dev Disord.* (2020) 7:168–81. doi: 10.1007/s40489-019-00186-7

76. Ingersoll B, Berger NI. Parent engagement with a telehealth-based parent-mediated intervention program for children with autism spectrum disorders: Predictors of program use and parent outcomes. *J Med Internet Res.* (2015) 17:e227. doi: 10.2196/jmir.4913

77. Bradshaw J, Bearss K, McCracken C, Smith T, Johnson C, Lecavalier L, et al. Parent education for young children with autism and disruptive behavior: response to active control treatment. *J Clin Child Adolesc Psychol.* (2018) 47:S445–S55. doi: 10.1080/15374416.2017.1381913

78. Montenegro MC, Abdul-Chani M, Valdez D, Rosoli A, Garrido G, Cukier S, et al. Perceived stigma and barriers to accessing services: experience of caregivers of autistic children residing in Latin America. *Res Dev Disabil.* (2022) 120:104123. doi: 10.1016/j.ridd.2021.104123

79. Baweja R, Brown SL, Edwards EM, Murray MJ. COVID-19 pandemic and impact on patients with autism spectrum disorder. *J Autism Dev Disord.* (2021) 1–10. doi: 10.1007/s10803-021-04950-9

80. Demetriou EA, Lampit A, Quintana D, Naismith S, Song Y, Pye J, et al. Autism spectrum disorders: a meta-analysis of executive function. *Mol Psychiatry.* (2018) 23:1198–204. doi: 10.1038/mp.2017.75

81. Trabacca A, Craig F. Social distancing and social interaction in children with autism spectrum disorder during the Covid-19 pandemic: a clinical oxymoron. *Minerva pediatrics.* (2021) 74:481–2. doi: 10.23736/S2724-5276.21.06057-2

82. Pellecchia M, Beidas RS, Mandell DS, Cannuscio CC, Dunst CJ, Stahmer AC. Parent empowerment and coaching in early intervention: study protocol for a feasibility study. *Pilot Feasibility Stud.* (2020) 6:1–12. doi: 10.1186/s40814-020-00568-3

83. Rogers SJ, Stahmer A, Talbott M, Young G, Fuller E, Pellecchia M, et al. Feasibility of delivering parent-implemented NDBI interventions in low resource regions: a pilot randomized controlled study. (2020) 14:3. doi: 10.21203/rs.3.rs-105121/v1

84. Shire SY, Worthman LB, Arbuckle S, A. Technology-enabled adaptation of face-to-face caregiver-mediated JASPER intervention: preliminary examination of video conferenced caregiver coaching. *Am J Intellect Dev Disabil.* (2021) 126:421–34. doi: 10.1352/1944-7558-126.5.421

85. Sandbank M, Bottema-Beutel K, Crowley S, Cassidy M, Dunham K, Feldman JI, et al. Project AIM: autism intervention meta-analysis for studies of young children. *Psychol Bull.* (2020) 146:1. doi: 10.1037/bul0000215

86. Glasgow RE, Emmons KM. How can we increase translation of research into practice? Types of evidence needed. *Annu Rev Public Health.* (2007) 28:413–33. doi: 10.1146/annurev.publhealth.28.021406.144145

87. Deb SS, Retzer A, Roy M, Acharya R, Limbu B, Roy A. The effectiveness of parent training for children with autism spectrum disorder: a systematic review and meta-analyses. *BMC Psychiatry.* (2020) 20:1–24. doi: 10.1186/s12888-020-02973-7

88. Durkin MS, Elsabbagh M, Barbaro J, Gladstone M, Happe F, Hoekstra RA, et al. Autism screening and diagnosis in low resource settings: Challenges and opportunities to enhance research and services worldwide. *Autism Research.* (2015) 8:473–6. doi: 10.1002/aur.1575

frontiers | Frontiers in Psychiatry

TYPE Original Research
PUBLISHED 31 August 2022
DOI 10.3389/fpsyt.2022.904380

EDITED BY
Costanza Colombi,
Stella Maris Foundation (IRCCS), Italy

REVIEWED BY
Cheng-Fang Yen,
Kaohsiung Medical University, Taiwan
Lu Qu,
East China Normal University, China

*CORRESPONDENCE
Wei-Tsuen Soong
soongwt@gmail.com

SPECIALTY SECTION
This article was submitted to
Autism,
a section of the journal
Frontiers in Psychiatry

RECEIVED 25 March 2022
ACCEPTED 03 August 2022
PUBLISHED 31 August 2022

CITATION
Seng G-J, Chiu Y-N, Tsai W-C, Lin H-Y, Li S-C, Hsiao M-N, Liu T-J, Chen H-M, Shih A, Chang Y-C, WHO CST Team and Soong W-T (2022) Promotion and implementation effectiveness of World Health Organization's Caregiver Skills Training program in Taiwan.
Front. Psychiatry 13:904380.
doi: 10.3389/fpsyt.2022.904380

Promotion and implementation effectiveness of World Health Organization's Caregiver Skills Training program in Taiwan

Guan-Jye Seng[1,2], Yen-Nan Chiu[1,3], Wen-Che Tsai[1,3], Hsiang-Yuan Lin[4], Su-Chen Li[1], Mei-Ni Hsiao[1,5], Tseng-Jung Liu[6], Heng-Man Chen[6,7], Andy Shih[8], Ya-Chih Chang[9], WHO CST Team[10] and Wei-Tsuen Soong[1,3]*

[1]Department of Psychiatry, National Taiwan University Hospital, Taipei, Taiwan, [2]Graduate Institute of Brain and Mind Sciences, College of Medicine, National Taiwan University, Taipei, Taiwan, [3]Department of Psychiatry, College of Medicine, National Taiwan University, Taipei, Taiwan, [4]Azrieli Adult Neurodevelopmental Centre, Centre for Addiction and Mental Health, Department of Psychiatry, University of Toronto, Toronto, ON, Canada, [5]School of Occupational Therapy, College of Medicine, National Taiwan University, Taipei, Taiwan, [6]Foundation for Autistic Children and Adults in Taiwan, Taipei, Taiwan, [7]Department of Special Education, National Taipei University of Education, Taipei, Taiwan, [8]Autism Speaks, New York City, NY, United States, [9]Department of Special Education and Counseling, California State University, Los Angeles, CA, United States, [10]Department of Mental Health and Substance Use, World Health Organization, Geneva, Switzerland

The World Health Organization (WHO) developed the Caregiver Skills Training for Families of Children with Developmental Delays and Disabilities (CST) with support from Autism Speaks to address the resource gaps and worldwide needs for interventions for children with developmental disorders or delays, especially those with autism spectrum disorder (ASD), and their families. Evidence has indicated that parent-mediated interventions benefit both caregivers and children by strengthening caregivers' knowledge and confidence and children's social communication skills and behavioral regulation. The CST-Taiwan team began the prepilot field trial in 2017 and developed the project to serve families in various locations. This study (1) delineated the adaptations and promotion of CST-Taiwan; (2) determined the program's effectiveness in the promotional stage, in terms of caregiver and child outcomes, and (3) examined the maintenance of its effects. The materials, delivery, and facilitator training procedure of the original CST were adapted to Taiwan. The quantitative data indicated that CST-Taiwan is a promising program, it positively affected caregiver knowledge and confidence and reduced the severity of the children's autistic symptoms. The 3-month follow-up results suggested that the effects persisted. Thus, CST-Taiwan, and its promotional strategies are feasible and effective.

KEYWORDS

autism spectrum disorder, developmental delays, World Health Organization Caregiver Skills Training, promotion, effectiveness

Introduction

Neurodevelopmental disorders are characterized by early childhood onset, alterations in central nervous system development, and functional impairments. Autism spectrum disorder (ASD) is a neurodevelopmental disorder characterized by persistent deficits in social communication and interaction and restricted and repetitive behaviors and interests (1). The prevalence is 1%—2% globally (2, 3) and 1% in Taiwan according to a nationally representative sample (4). The increasing prevalence (5) has placed a considerable financial and psychosocial burden on individuals, families and the society (6–8).

Early intervention is beneficial for autistic children in various regards, including social functioning, adaptive behaviors and behavioral regulation (9–11). Early intervention for developmental delays is highly demanded in Taiwan, with approximately 20,000 (1.5%) children younger than 6 years of age requiring such services every year (16,584 children in 2017, 19,103 children in 2018, 19,407 children in 2019, and 19,723 children in 2020) (12). In Taiwan, most early interventions are implemented in medical institutions in National Health Insurance system and nursery institutions in the social welfare system (13). Among early intervention services in Taiwan in 2020, 73% were implemented in medical institutions whose health care services are solely covered by the National Health Insurance, and 15.6% were implemented in nursery institutions (12). However, Taiwan's National Health Insurance only allows early interventions to be reimbursed as a non-intensive therapy (i.e., only one 30-min therapy session can be charged per person/week/institution). As such, daycares and child development centers that provide intensive treatment and care cannot satisfy the demand for interventions (3.7% of early intervention services in 2020) (12). Families often receive early intervention services at multiple institutions to increase treatment hours for their children. This distinct phenomenon in Taiwan results in inconsistency among the interventions (i.e., goals, theoretical bases, treatment principles, and skills) and requires time for travel, thus imposing a considerable extra burden on families, especially those living in rural area with limited access to services (14).

Parent-mediated interventions may compensate for this problem. Such programs require two key components: providing parents with information regarding ASD and helping them develop parenting skills (15). Parents' involvement in interventions for ASD improves parent–child interaction, generalization of the skills to daily living, increases consistency of interventions chose by the families, and enables parents to receive psychosocial support (16–18). In addition, parents trained in interventions can continue employing strategies in their children's daily lives (19, 20), thus generalizing and maintaining the treatment effect (21). Moreover, parent-mediated interventions increase parents' confidence in caregiving, reduce parental stress, improve quality of life, and lead to better parent-child relationship (16, 21–23). Improvement in parent–child interaction is a potential mediator in reducing autistic symptoms (24). Meta-analyses have revealed that parent-mediated interventions significantly reduced clinical symptom severity of the children (e.g., small effect size on parent-reported communication skills and medium effect size on challenging behaviors) (16–18). Earlier empirical studies have also revealed effects on children's sociability (25) and emotional regulation (26).

The World Health Organization (WHO) developed the Caregiver Skills Training Program for Families of Children with Developmental Delays and Disabilities (originally Caregiver Skills Training Program for Families of Children with Developmental Disorders or Delays, CST) with support from Autism Speaks (AS) (27) to improve caregiver skills and facilitate caregiver–child interaction, according to the WHO's mhGAP Intervention Guide (28). The program is effective, freely available, deliverable by non-specialists, feasible with limited resource, and can be integrated into other services (29). The WHO CST is open to all caregivers (not limited to parents) of children with developmental disorders or delays. The CST program consists of nine caregiver group sessions with manualized guides, three home visits, and three telephone calls and is based on the principles of natural developmental behavioral interventions (NDBIs) (29). The program is delivered according to established manuals by facilitators who are trained through WHO CST training program (i.e., Training of Trainers, ToT). The group sessions cover specific topics, including engagement, building up routines, communication, behavioral regulations and management, learning new skills, and caregiver's well-being (29). The caregiver role-play during group sessions and at-home practices after group sessions are used to strengthen skills from each session. Home visits are conducted prior to, midway through, and at the end of the group sessions to engage, coach, and support families, set goals, and evaluate the progress. The telephone calls are implemented after group sessions to clarify the group's focus and offer support. Although the WHO CST program was designed for global use, the materials and delivery process should be adapted for each context and to suit local needs (29, 30) to make the program relevant and feasible for implementation. The process of evaluating the feasibility and accessibility of the WHO CST consists of three stages: adaptation, prepilot and pilot stages (29, 31). Ideally, the adaptations were made prior to and within the prepilot stage. The adaptations were tested in the pilot stage to ensure the program is feasible and acceptable for dissemination in the local context. The program had been adapted and implemented in both low-income countries and high-income countries (30–32). The previous studies indicated that the WHO CST program is both acceptable and relevant to the low-resource contexts (e.g., Ethopia and India) (30, 31) and to the high-resource context (e.g., Italy) (33). A pilot randomized controlled trial study in Italy reported the promising effectiveness on joint engagement of caregiver–child interaction, parent stress, parenting self-efficacy and child gestures (32). The Taiwan CST team began adaptation and field testing in 2016, completed these steps in early 2019, and advanced to promotion in 2019. This study explored (1) the adaptations of CST-Taiwan, (2) its promotion, and (3) its effectiveness during the promotion stage.

Materials and methods

Adaptation process

This study is a part of the CST Taiwan Adaptation and Implementation Project launched by the Department of Psychiatry at National Taiwan University Hospital (NTUH) and the Foundation for Autistic Children and Adults in Taiwan (FACT). The core local team, consisting clinicians at NTUH and workers at FACT, was established in 2016 which lead the work of translation of "WHO CST Field Test version 1.0". The materials, namely the facilitator guide, participant booklet, home visit guide, consent forms for families, and the monitoring and evaluation framework, were translated into Traditional Chinese (WHO CST Traditional Chinese version 1). Four master trainers were trained by the WHO-AS CST team and delivered the prepilot groups in 2017 in two institutions (one medical institution and one social welfare institution) in Taipei City, a metropolis. Before the prepilot groups, some adaptations of CST-Taiwan were developed on the basis of preparation meetings and rehearsals, yielding the WHO CST Traditional Chinese version 2, which was used in the prepilot groups. Because the WHO CST team released the WHO CST Field Test version 2.06 in 2018, our new version, based on WHO CST Field Test version 2.06 and a review of the prepilot groups in Taiwan, were used in the pilot groups in 2018 (i.e., the WHO CST Traditional Chinese version 3). Minor adaptations were made on the basis of the pilot study, leading to the latest version used in the promotion, the WHO CST Traditional Chinese version 4. In 2017 and 2018 (i.e., prepilot and pilot stages), CST-Taiwan implemented six caregiver groups by 12 facilitators (including the four master trainers) in five institutions, three in Taipei city and two in Kaohsiung (a city in southern Taiwan). We recruited 31 families (caregivers: $n = 31$, mean age=39.4 years, standard deviation [SD] = 6.1; children: $n = 31$, mean age = 4.0 years, [SD] = 1.0] during the prepilot and pilot stages in 2017 and 2018. Local adaptations in Taiwan were submitted to the WHO CST team for approval. The qualitative data collected during the adaptation process will be reported separately.

Training of trainers (ToT)

In 2017, WHO-AS CST trainers held a 5-day ToT in Taiwan. Four specialists (one clinical psychologist, two occupational therapists, and one early intervention teacher) participated in live practice with children and submitted the Adult-Child Interaction fidelity videos to a WHO-AS CST trainer for verification. The WHO-AS CST trainer supported the subsequent posttraining practices as a technical consultant. After completion of the Adult-Child Interaction fidelity verification and the prepilot groups, the four master trainers led the first ToT for facilitators in Taiwan in 2018 (i.e., the pilot stage) and collected feedback from the facilitators for further adaptation. Thereafter, eight facilitators delivered four CST groups (two facilitators jointly delivered one group in pair) in Taipei and Kaohsiung at the pilot stage under the full supervision of the master trainers to ensure the procedural fidelity and to collect information to verify the local adaptations. The WHO-AS CST consultant verified the fidelity videos. ToT required adaptation for Taiwan because of the master trainers' workload, geographic restrictions, and the need for facilitators for promotion. Adapted ToT in Taiwan were held annually to scale up the promotion, and 15 facilitators maximum were trained a year.

We started the promotion stage in 2019. New institutions were invited to participate, and the four master trainers trained new facilitators each year. We targeted two cities or counties each year. Ideally, we sought to invite one medical institution and one social welfare institution (e.g., preschool education and early intervention organizations) in each targeted city or county; this was because early intervention services are provided in various institutions in Taiwan, and thus, we sought for representativeness of institutions (13). In the new institutions, newly trained facilitators would hold one CST group under the master trainers' supervision. The group enrollment ranged from four to six families. Institutions and facilitators who had previously participated were invited to continue the program in the following years after accreditation.

A review meeting was held at the end of each year of promotion. The CST-Taiwan core team, master trainers, facilitators, the leaders of the institutions, and the WHO-AS CST consultants attended the meetings. The implementation process, feedbacks, and challenges were reviewed. Suggestions of revision for future use were discussed and finalized, after approval by the WHO-AS consultants.

Enrollment of families

Information regarding the Taiwan CST program was shared on the FACT website and advertised by the participating institutions. The families registered by themselves with or without referrals. To participate, families were required to have (1) children aged 2–6 years with (2) clear evidence of developmental delays, such as a confirmed clinical diagnosis (ASD was prioritized), developmental evaluation reports, or a disability certification; (3) the caregivers' commitment to participate in the group sessions and home visits; and (4) a basic level of reading and spoken Mandarin Chinese. However, we provided a brief translation of the materials (i.e., the whole participant booklet was translated into Simplified Chinese and the Key Messages and the Tips were translated into Vietnamese) to help caregivers who unfamiliar with Traditional Chinese. Usually, the main caregivers are the parents of children with developmental delays. Nonetheless, aunts, uncles, or grandparents could be the caregivers for some families because of the distinct family dynamics. Recruitment required the collective agreement of the Taiwan core team and participating institutions. During promotion stage in 2019 and 2020, we recruited 91 families. The data collected during the promotion stage were used in the analysis of effectiveness. The demographic data of the participants consisted of the child's age, sex, and treated/non-treated prior to the CST (i.e., whether the child had been treated through any type of early interventions; dichotomous variable) and the caregiver's age, relation to the child, educational level, and ethnicity.

All of the procedures during the prepilot and pilot field trials were approved by NTUH Institutional Review Board (#201703123RIND). We obtained written informed consent from the parent or substitute decision maker of each participated child after explaining the present objectives and procedures.

Measurements

WHO caregiver knowledge and skills test

The WHO Caregiver Knowledge and Skills Test is a 5-point caregiver-reported questionnaire developed by the WHO CST team (unpublished) and consists of 24 items measuring the caregivers' understanding of the key principles of CST (i.e., knowledge). The higher score represents a stronger understanding of WHO CST caregiving (31). The internal consistency was high in our sample who completed baseline, postintervention, and follow-up tests, indicating high reliability (Cronbach's α of baseline/postintervention/follow-up test was 0.78/0.78/0.80).

Caregiver self-efficacy questionnaire

The Caregiver Self-Efficacy Questionnaire (CSQ) is a caregiver-reported questionnaire developed by the WHO CST team (unpublished) and consists of 13 items scored from 0 to 5 points to measure caregivers' confidence in the strategies delivered by WHO CST. The internal consistency was high in our sample who completed baseline, postintervention, and follow-up tests, indicating high reliability of this measure (Cronbach's α of baseline/postintervention/follow-up test was 0.89/0.91/0.93).

Family empowerment scale

The Family Empowerment Scale (FES) is used to measure the family empowerment in caregiving for children with special needs. It is a 5-point caregiver-reported questionnaire consisting of 34 items (34). The internal consistency in our sample who completed baseline, postintervention, and follow-up tests was high, indicating high reliability (Cronbach's α of pre-/post-/follow up test was 0.91/0.94/0.94).

Autism treatment evaluation checklist

The Autism Treatment Evaluation Checklist (ATEC) is a caregiver-reported questionnaire suggested as a comprehensive tool to monitor the treatment effect and progress of children with ASD. As a caregiver-reported measure, it has been validated in comparison of professional-rated measures with a well-established validity (35, 36). Four subscales are used to assess child outcomes, namely speech/language/communication (14 items, 3-point scale), sociability (20 items, 3-point scale), sensory/cognitive awareness (18 items, 3-point scale) and health/physical behavior (25 items, 4-point scale). The scores for the speech/language/communication scale and sensory/cognitive awareness scales were reversed to match the other two scales. Higher scores represented more severe autistic symptoms. The internal consistency in our sample who completed baseline, postintervention, and follow-up tests, indicating high reliability of the Chinese version (Cronbach's α of baseline/postintervention/follow-up test was 0.94/0.94/0.94 in the first subscale; 0.92/0.94/0.90 in the second subscale; 0.91/0.93/0.92 in the third subscale; 0.84/0.85/0.82 in the fourth subscale).

Quantitative data collection

The Caregiver Knowledge and Skills Test, CSQ, ATEC were also translated into Traditional Chinese by the CST-Taiwan team during the adaptation process, while the Chinese version of FES was translated and validated in 2011 (37). The participants completed all measurements before the intervention (i.e., the questionnaires were given to the caregivers at the first home visits and sent back to facilitators at the first group session, which was the baseline), immediately after the intervention (postintervention), and 3-months after the intervention (follow-up). For families with two caregivers who participated in all sessions, the two caregivers completed separate questionnaires for caregiver-related outcomes.

Statistical analysis

The demographics of the participants and their children are presented descriptively. The missing value rate of the questionnaires was 0.2%. Multiple imputation was used to replace missing values (38, 39) if the missing values presented randomly. A repeated-measures analysis of covariance (ANCOVA) was performed to compare the baseline, postintervention, and follow-up data with adjusting for caregiver age and educational level on WHO Caregiver Knowledge and Skills Test, CSQ and FES scores, and child age, sex, and treatment history and caregiver age on ATEC. We used Tukey's test to conduct *post hoc* analysis for the variables with significant differences.

To address the concerns regarding differences between families with whom we did not follow up, which may influence the interpretation of the follow-up data, we compared the demographics, baseline of caregiver and child outcomes between these two groups. In addition, the facilitators delivering the CST had three experience levels (i.e., A: two experienced facilitators, B: one new facilitator and one experienced facilitator, and C: two new facilitators; detailed in Section Adaptations of training of trainers (ToT) and supervision). Thus, we preformed ANCOVA to compared the changes between baseline and postintervention among these groups and to justify the analyses in which the effects of the facilitators' experience level on effectiveness were not controlled for. The baseline scores and Baseline× Group interaction term were included as covariates in the ANCOVA (40). These analyses are presented in the Supplementary materials. Providing the exploratory nature of this study, the presented results were not corrected for multiple tests.

Results

Local adaptations of CST-Taiwan

Several local adaptations were developed from the experience with the prepilot and pilot field trials, focus groups with facilitators and caregivers, and the comments during the review meetings for the core local team. The adaptations are summarized in Table 1.

Adaptations of WHO CST materials

Adaptations were made to the contents of the WHO CST facilitator guide and the participant booklet, namely linguistic and sociocultural adaptation and the formatting. The "characters" and "locations" in the stories and demonstration scripts were changed into local names; however, the illustrations were retained to depict families of numerous ethnic origins. The facilitator guide and the participant booklet were reformatted to make the Key Messages, Tips, and facilitation notes more recognizable to readers (e.g., highlighting passages).

Adaptations of WHO CST delivery

The age range of recruited children was adapted to 2–6 years (2–9 years in the original WHO CST program). A major adaptation in the Taiwan CST delivery was the use of prerecorded videos as substitutes for live demonstrations. The two facilitators recorded the videos beforehand to ensure that they understood the strategies demonstrated. The number of telephone calls was increased from three in the original WHO CST program to seven (i.e., after every session without a home visit). Also, we added coaching and troubleshooting for any difficulties among home practice to the telephone calls which last at least 30 min. The caregivers were welcomed to send home practice videos for facilitators' review before telephone calls. Therefore, we renamed the telephone calls to telephonic sessions.

Adaptations of training of trainers (ToT) and supervision

First, we adapted the selection criteria for the facilitators. Although the WHO CST was originally designed to be delivered by both specialists and nonspecialists, we established selection criteria to optimize the implementation of CST-Taiwan. Institutions recommended facilitators on basis of two criteria: (1) license in a field related to developmental early-intervention or education (e.g., certified therapists in medical institutions, teachers, and practitioners from early intervention organizations) and (2) at least 3 years of experience implementing any type of developmental early interventions.

Second, the training of facilitators, which originally included a 5-day ToT session and post-ToT practices under supervision, was also adapted. The 5-day ToT session was divided into two 2.5-day sessions with 2 weeks apart to suit the clinical workload and schedule and to have time for practice. We added a theoretical introduction to the CST to the facilitator guide to deepen the facilitators' understanding of the core principle. More videos were shared during the training course to illustrate the Key Messages and Tips of CST. The hands-on practice time with children was also increased. The facilitators were invited to use the Adult–Child Interaction Fidelity Scale (WHO CST team, unpublished) to offer feedback to each other, and master trainers provided debriefing sessions immediately after the hands-on practice time with children. The facilitators were required to send Adult-Child Interaction Fidelity videos to verify they meet the WHO CST requirements. We added 3 rehearsal sessions after the 5-day ToT session, prior to the implementation of CST groups. The newly trained facilitators rehearsed all sessions to familiarized themselves with the content and structure of the CST groups. For the post-ToT practices, we used two facilitators joint-delivery under supervision of master trainers, rather than the original model of one master trainer and one new facilitator joint-delivery, which is also an adaptation of WHO CST delivery. The master trainers demonstrated an example of the first home visit for the new facilitators. The

TABLE 1 Adaptation summary of CST-Taiwan.

	Planning and translation (2016)	**Prepilot stage (2017)**			**Pilot stage (2018)**		
	Translation and adaptation	**ToT**	**Prepilot groups**	**Adaptation (2018)**	**ToT**	**Pilot groups**	**Adaptation (2019)**
Purpose	To translate the WHO CST materials to Traditional Chinese for local use.	To investigate the feasibility and the acceptability of the translated/adapted materials and the delivery process. To identify barriers of local implementation which may need adaptations.			To test the feasibility and acceptability of the adaptations based on the prepilot stage. To make further adaptations for promotion stage.		
Personnel	The local core team and 8 child therapists at NTUH.	WHO-AS CST Trainers, the local core team and potential master trainers.	The local core team and 4 master trainers.	The local core team and 4 master trainers.	The local core team, 4 master trainers and 8 facilitators.	The local core team, 4 master trainers and 8 facilitators from 4 institutions.	The local core team and 4 master trainers.
Adapted materials							
Facilitator guide	●	●		●			●
Participant booklet	●	●		●			●
Home visit guide	●	●					
Consent forms	●						
Monitoring and evaluation framework	●						
Version of materials	WHO CST Field Test version 1.0 was translated to WHO CST Traditional Chinese version 1.	Used WHO CST Traditional Chinese version 1, further adapted to version 2 during the rehearsals.	Used WHO CST Traditional Chinese version 2.	Adapted to WHO CST Traditional Chinese version 3.	Used WHO CST Traditional Chinese version 3.	Used WHO CST Traditional Chinese version 3.	Adapted to WHO CST Traditional Chinese version 4.
Summary of adaptations	Translation and sociocultural adaptations.	Unifying the format of facilitator guide and participant booklet.		Updated the version to WHO CST Field Test 2.06; adding theoretical bases to facilitator guide; highlighting and reformatting.			Minor change of wordings to make the texts plain.
Adapted ToT							
Criteria of facilitators					●		
Materials for ToT		●		●			
Format		●			●		
Contents of training					●		

(Continued)

TABLE 1 Continued

	Planning and translation (2016)	Prepilot stage (2017)			Pilot stage (2018)		
	Translation and adaptation	**ToT**	**Prepilot groups**	**Adaptation (2018)**	**ToT**	**Pilot groups**	**Adaptation (2019)**
Summary of adaptations		Establishing the play kits; adding rehearsals.		Translated Adult–Child Interaction Fidelity Scale.	Licenses and experiences required for facilitator; ToT was divided into two 2.5-day sessions 2 weeks apart; adding introduction of theoretical bases; increasing hours for hands-on practice.		
Adapted CST delivery							
Enrollment of participants			●				
Delivery personnel						●	
Group sessions						●	
Telephonic sessions			●				●
Home visits							●
Summary of adaptations			Change the recruitment children's age; 3 telephone calls changed to 7 telephonic sessions.			Two facilitators (rather than one master trainer and one facilitator) joint-delivery under supervision of master trainers; using prerecorded demonstration.	Establishing recording forms for telephonic sessions and home visits.

ToT, training of trainers; NTUH, National Taiwan University Hospital; FACT, Foundation for Autistic Children and Adults in Taiwan.
The core local team of CST-Taiwan consists four child psychiatrists at NTUH (W-TS, Y-NC, W-CT, H-YL), and the chief executive officer at FACT(T-JL).

TABLE 2 Promotion summary of CST-Taiwan.

Stage	Prepilot stage	Pilot stage	Promotion stage	
Year	2017	2018	2019	2020
Regions (City/County)	Taipei	Taipei, Kaohsiung	Taipei, Kaohsiung, Hsinchu, Yilan, Penghu	Taipei, Kaohsiung, Hsinchu, Yilan, Hualien, Taitung
Number of newly trained facilitators	4 (Master trainers)	8	10	12
Number of implemented facilitators	4 (Master trainers)	8	14	21
Number of new institutions	2	3	4	4
Number of implemented institutions	2	4	7	10
Number of CST groups	2	4	7	11
Number of participating families	11	20	37	54

facilitators subsequently implemented their first CST group in pairs under the supervision of master trainers. The facilitators were considered as fully trained after the fidelity verification, rehearsals, and post-ToT practices, consisting of observing the first home visit implemented by master trainers, and their implementation under supervision.

We established a hierarchical supervision model for the facilitators and institutions based on level of experience (i.e., A: online supervision as required for two experienced fully trained facilitators, B: partial supervision for one experienced fully trained facilitator and one new facilitator, and C: full supervision for two new facilitators) to ensure the fidelity of the CST delivery. We also created home visit and telephonic sessions recording forms according to the CST guidelines for home visits and telephonic sessions. The recording forms record information (e.g., the child's competencies, the concerns of challenges, goals and progress of home practice) collected through interview or live interaction with the child. The facilitators were required to complete the forms for master trainers' review.

Promotion of CST-Taiwan

During the study period (i.e., 2019–2020), 27 facilitators in 12 institutions implemented CST-Taiwan through 18 caregiver groups. The institutions were distributed across both urban and rural areas in Taiwan, including Taipei, Kaohsiung, Hsinchu, Yilan, Penghu, Hualien, and Taitung (Table 2 and Figure 1). A total of 33% of the facilitators continued to implement CST in the following years (included 5 facilitators, who were trained in 2018 and continued to implement CST in the following years). The facilitators were clinical psychologists, occupational therapists, physiotherapists, speech therapists, special educators, educators, psychiatrists, early intervention teachers, and nurses (Figure 2). Most facilitators reported a positive experience of implementing CST. Intensive supervision played a critical role in ensuring the new facilitators in correctly followed the CST guidelines, empowered them, and helped them resolve challenging problems in certain families. We used the quantitative data from the first 2 years of promotion (i.e., 2019–2020) to explore the effectiveness.

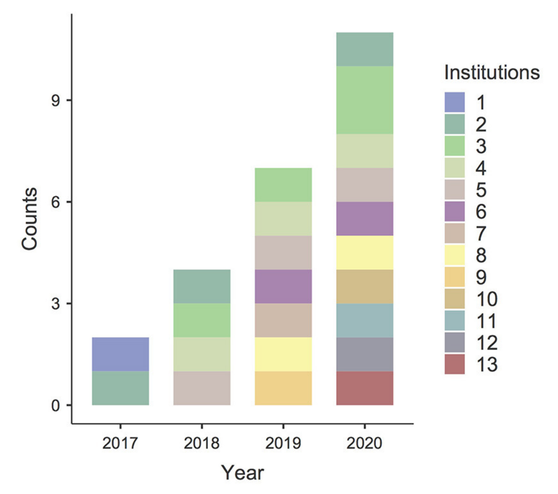


FIGURE 1
Promotion of CST-Taiwan. The number of institutions delivering CST increased over time. Two institutions implemented the first two groups in Taiwan (prepilot stage) in 2017. Four institutions implemented four groups (pilot stage) in 2018. During the promotional stage, seven institutions implemented seven groups in 2019. Ten institutions implemented 11 groups in 2020. By the end of 2020, 13 institutions had joined the CST-Taiwan program and implemented 24 groups.

Demographics of participants during promotion of CST-Taiwan

In 2019 and 2020, CST-Taiwan served 91 families (94 caregivers and 91 children, three families had two caregivers participating all sessions together). The mean age of the caregivers and children was 38.56 (SD = 5.81; range: 27–61)

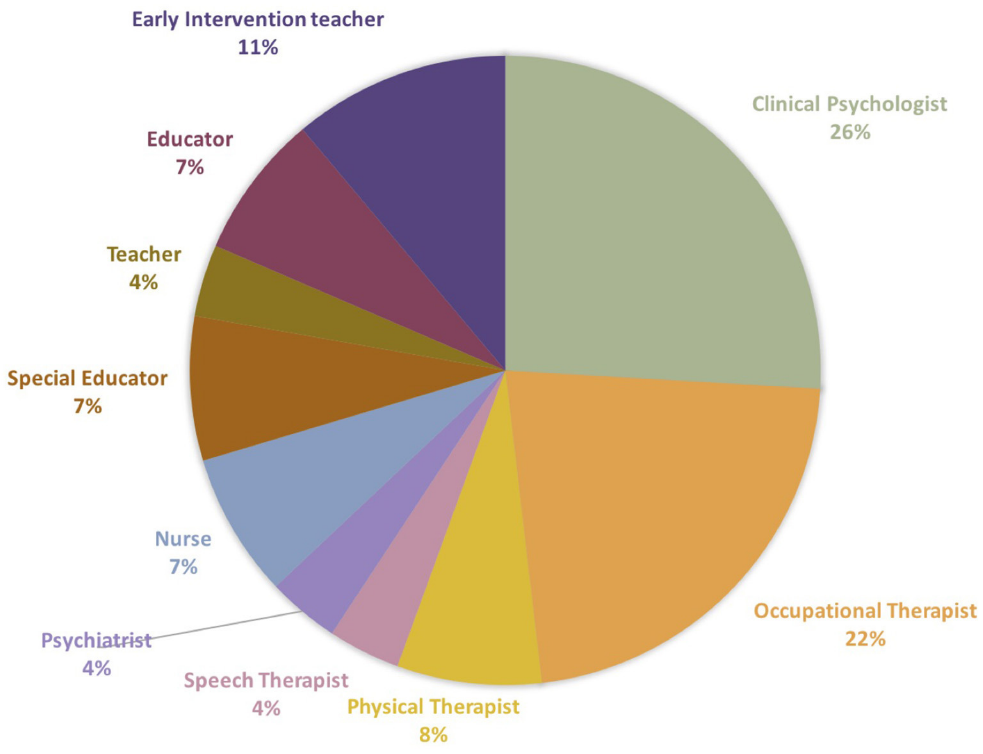


FIGURE 2
Types of the profession of facilitator at the promotional stage. The facilitators were clinical psychologists, occupational therapists, physiotherapists, speech therapists, special educators, educators, psychiatrists, early intervention teachers, and nurses.

and 3.85 (SD = 1.07; range: 1.32–6.84) years, respectively. The caregivers were mostly the children's mothers (87.2%) and held a college degree or higher (81.9%). The baseline caregiver knowledge, confidence, and family empowerment were 96.06 (SD = 8.02), 38.76 (SD = 8.04), and 124.59 (SD = 14.9), respectively. Most of caregivers were Taiwanese (90 caregivers, including 3 indigenous), and four were foreigners. Most children were diagnosed as having ASD, but some were diagnosed as having developmental delays or disorders. Half of the children (51.6%) had been treated through any type of early interventions prior to CST. The mean baseline symptoms severity (total ATEC score) was 62.12 (SD = 25.3). Eighty-nine children in our sample were with a baseline ATEC score ≥20, a suggested minimal ATEC severity for using ATEC as a measurement (41). Only two children with ATEC total score lower than 20 (total score =18 and 19) at baseline, however, the clinical impression of ASD was apparent (Table 3). Only one family dropped out due to the busy schedule of the caregiver. Overall attendance rate was 91.7 % (Sessions 1–9: 95, 92, 95, 87, 91, 93, 91, 88, and 95%, respectively). Figure 3 presents a CONSORT flow diagram of the participants.

Differences among baseline, postintervention and follow-up

A total of 87 WHO Caregiver Knowledge and Skills Test, CSQ and FES questionnaires were completed both at baseline and postintervention, and 48 were completed at follow-up; 85 baseline and postintervention ATECs were completed, and 50 follow-up checklists were completed. We observed no significant difference in demographics and baseline child and caregiver outcomes between participants with whom we followed up and those with whom we did not (Supplementary Table 1). No significant difference in demographics and baseline child and caregiver outcomes was observed among the groups with various facilitator experience levels, but more children had been previously treated in groups delivered by two new facilitators compared to the groups delivered by one new and one experienced facilitator (Supplementary Table 2). We observed no significant difference in caregiver (i.e., knowledge, confidence, and empowerment) or child outcomes (i.e., all ATEC subscales and the total score) among the groups with various facilitator experience levels (Supplementary Table 3).

TABLE 3 Demographics of children and caregivers participating CST-Taiwan at the promotion stage.

Children ($n = 91$)	
Age, year [mean, (SD)]	3.85 (1.07)
Age range	1.32–6.48
Sex [*n*, (%)]	
Male	70 (76.9%)
Female	21 (23.1%)
Treatment history [*n*, (%)]	
Treated	47 (51.6%)
Non-treated	44 (48.4%)
Geographic areas [*n*, (%)]	
Rural	23 (25.3%)
Urban	68 (74.7%)
Baseline autistic symptoms severity[a] [mean, (SD)]	62.12 (25.29)
Baseline autistic symptoms severity[a] range	18–116.2

Caregivers ($n = 94$)	
Age, year [mean, (SD)]	38.56 (5.81)
Age range	27–61
Sex [*n*, (%)]	
Male	7 (7.4%)
Female	87 (92.6%)
Caregiver's relation to the child[b] [*n*, (%)]	
Father	7 (7.4%)
Mother	82 (87.2%)
Grandparents	3 (3.2%)
Other	2 (2.1%)
Caregiver educational levels [*n*, (%)]	
Junior	3 (3.2%)
High	14 (14.9%)
College	55 (58.5%)
Graduate	22 (23.4%)
Ethnicity	
Taiwanese	87 (92.6%)
Indigenous	3 (3.2%)
Foreign	4 (4.3%)
Baseline caregiver knowledge[c] [mean, (SD)]	96.06 (8.02)
Baseline caregiver confidence[c] [mean, (SD)]	38.76 (8.04)
Baseline family empowerment[c] [mean, (SD)]	124.59 (14.94)

SD, standard deviation.

[a] Baseline autistic symptoms were assessed by the total score of Autism Treatment Evaluation Checklist. Only 88 children had this data. [b] Three families had two caregivers participating group sessions together. [c] Only 92 caregivers completed these questionnaires, one father and one mother whose partner also participating groups session did not complete these questionnaires.

Caregiver outcomes

With controlling for caregiver age and educational level, significant difference was observed between baseline and postintervention knowledge [$F_{(1,84)} = 4.78$, $p = 0.032$, $n = 87$, Table 4A]. Significant effect of time on confidence [$F_{(2,90)} = 3.80$, $p = 0.026$, $n = 48$] was observed among difference among baseline, postinvention and follow-up. No significant difference was observed between the postintervention and follow-up results in *post hoc* analysis (Table 4B).

Child outcomes

With controlling for child's age, sex, treatment history and caregiver' age, significant differences between baseline and postintervention on speech/language/communication, sociability, health/physical behavior, and the total score were observed [$F_{(1,80)} = 11.95$, $p < 0.001$; $F_{(1,80)} = 5.04$, $p = 0.028$; $F_{(1,80)} = 4.47$, $p = 0.038$, $F_{(1,80)} = 9.23$, $p = 0.003$, Table 5A). Significant effect of time on speech/language/communication, sociability, and the total score were observed in differences among baseline, postintervention and follow-up [$F_{(1.84,82.70)} = 7.82$, $p = 0.001$; $F_{(2,90)} = 7.60$, $p < 0.001$; $F_{(2,90)} = 3.64$, $p = 0.030$]. In *post hoc* analysis, no significant difference was observed between the postintervention and follow-up results in speech/language/communication and the total score, while no significant difference was observed in the baseline–follow-up and postintervention–follow-up comparisons in sociability (Table 5B).

Discussion

This study revealed that implementing the local adapted WHO CST is feasible in Taiwan. The content, delivery, criteria for facilitators selection, and ToT were adapted. The promotional model of CST-Taiwan expanded the service to cities and counties across rural and urban areas in Taiwan. The quantitative data indicated the preliminary effectiveness of CST-Taiwan in positively affecting the caregivers' knowledge and confidence and the children's autistic symptoms with a maintenance effect.

The CST was adapted in Taiwan to optimize implementation across the country. The content was adapted only slightly, suggesting that the WHO CST materials were already applicable to Taiwan. This is similar to the experiences from several WHO CST sites which suggested that the contents of WHO CST is feasible and acceptable for global contexts (30, 31, 33). In terms of delivery, the use of prerecorded videos and increased telephonic sessions strengthened the effects of the program on the caregivers' learning. As some demonstration scripts were complex for facilitators to act leading to a lower acceptability in caregivers (30, 33), prerecorded demonstration videos eased the preparation pressure of facilitators and increased the understandability of the caregivers. The facilitators emphasized and clarified key concepts by pausing or replaying the videos for the caregivers during the group sessions. The increased

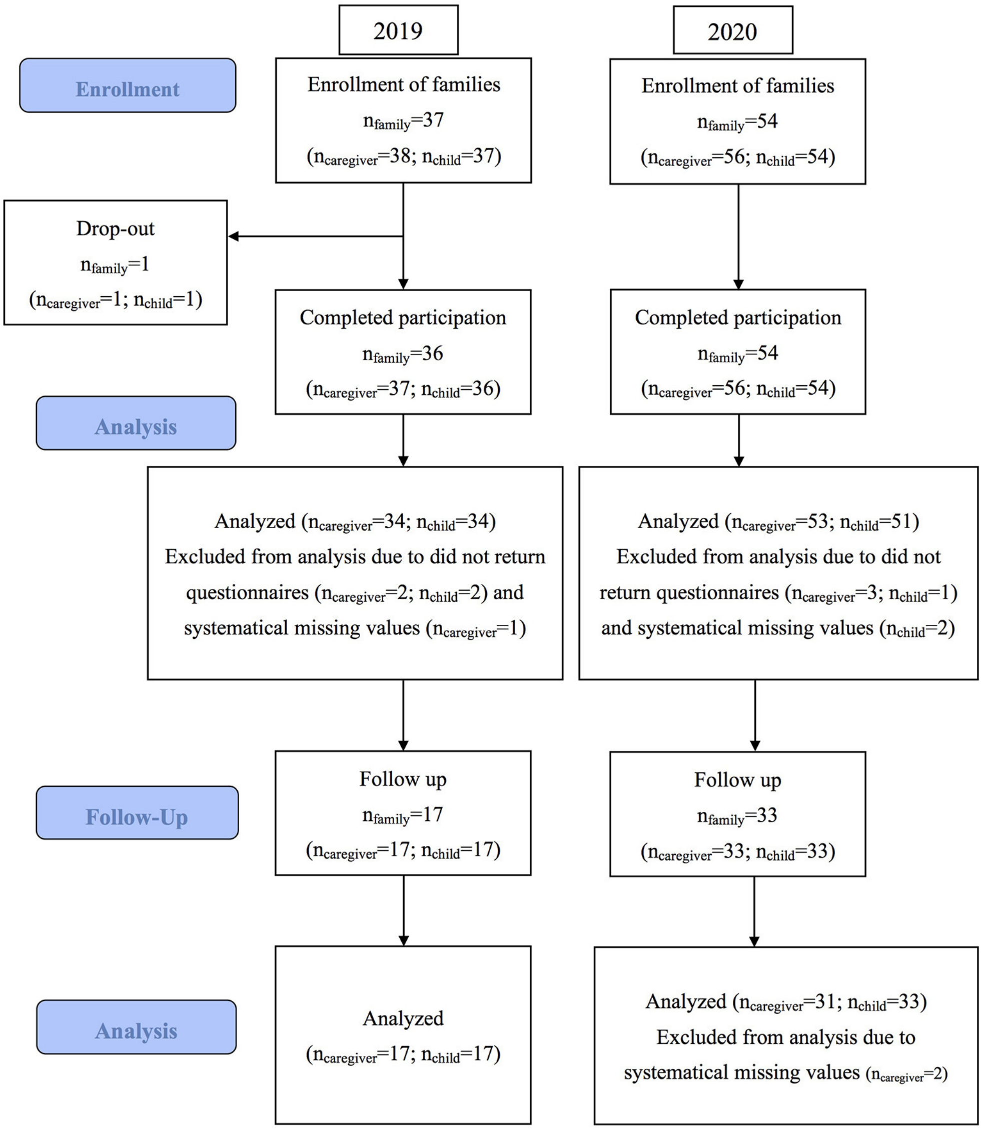


FIGURE 3
CONSORT flow diagram.

telephonic sessions provided one-on-one individual coaching after each group session, which is one of major adaptations of CST-Taiwan responding to the suggestions for intensifying the coaching in Salomone et al.'s work (33). The facilitators encouraged and coached the caregivers to do home practices through intensive follow-ups. When caregivers described challenges they faced during at-home practice between group sessions, the facilitators coached them and offered tips.

Although originally nonspecialists could implement CST, our facilitators were all specialists in developmental early interventions, which is a difference between CST-Taiwan and others (31). Because half of the children had received early interventions, the facilitators with experience in early intervention and related skills felt capable of answering caregivers' questions, and upholding the Adult-Child Interaction fidelity. In addition, CST groups were delivered by two newly trained facilitators in Taiwan, which is different from the WHO CST suggested that facilitators jointly deliver CST groups with master trainers (as a part of post-ToT practices). Thus, we used a different training format to provide more practice opportunities for facilitators. First, we introduced the theoretical bases of CST to the facilitators at the beginning of the ToT because not all were familiar with NDBIs. This benefited facilitators, who had various professional backgrounds, by aligning their professional perspectives with the concepts of NDBIs before learning CST; this may have prevented confusion

TABLE 4 Caregiver outcomes-baseline, postintervention and follow-up differences.

(A) Baseline and postintervention differences

	Mean (SD)		Repeated measured ANCOVA§		
	1 Baseline ($n = 87$)	2 Postintervention ($n = 87$)	Statistic (F)	p–value	Effect size (η^2)
Knowledge	95.94 (7.92)	102.83 (7.41)	4.78	0.032	0.006
Confidence	38.86 (8.08)	49.17 (6.75)	2.94	0.090	0.005
Empowerment	124.71 (15.25)	135.04 (15.87)	<0.001	0.992	<0.001

(B) Baseline, postintervention and follow-up differences

	Mean (SD)			Repeated measured ANCOVA§			*Post-Hoc* (p_{Tukey})
	1 Baseline ($n = 48$)	2 Postintervention ($n = 48$)	3 Follow up ($n = 48$)	Statistic (F)	p-value	Effect size (η^2)	
Knowledge	96.15 (8.28)	102.30 (7.69)	100.86 (8.52)	2.21	0.115	0.006	—
Confidence	39.42 (7.72)	49.28 (6.79)	49.09 (8.14)	3.80	0.026	0.015	1 < 2,3
Empowerment	123.78 (14.8)	136.04 (15.9)	134.04 (16.1)	0.52	0.595	0.001	—

SD, standard deviation; ANCOVA, analysis of covariance.

§caregiver's age and educational level included as covariates.

TABLE 5 Child outcomes-baseline, postintervention and follow-up differences.

(A) Baseline and postintervention differences

	Mean (SD)		Repeated measured ANCOVA§		
	1 Baseline ($n = 85$)	2 Postintervention ($n = 85$)	Statistic (F)	p value	Effect size (η^2)
Speech/language/communication	13.20 (7.95)	10.95 (7.53)	11.95	0.001	0.002
Sociability	13.11 (6.63)	11.63 (7.43)	5.04	0.028	0.003
Sensory/cognitive awareness	15.50 (6.96)	13.49 (7.10)	2.94	0.090	0.001
Health/physical behaviors	20.54(10.93)	18.59 (10.68)	4.47	0.038	0.002
Total	62.34(25.12)	54.66 (25.15)	9.23	0.003	0.004

(B) Baseline, postintervention and follow-up differences

	Mean (SD)			Repeated measured ANCOVA§			*Post-Hoc* (p_{Tukey})
	1 Baseline ($n = 50$)	2 Postintervention ($n = 50$)	3 Follow up ($n = 50$)	Statistic (F)	p value	Effect size (η^2)	
Speech/language/communication	12.86 (7.84)	10.64 (7.44)	9.52 (7.74)	7.82	0.001	0.004	1 > 2,3
Sociability	12.48 (6.60)	11.40 (8.14)	10.90 (7.83)	7.60	<0.001	0.006	1 > 2
Sensory/cognitive awareness	15.66 (7.29)	13.98 (7.59)	12.48 (7.92)	0.34	0.711	<0.001	—
Health/physical behaviors	18.13 (9.11)	17.17 (9.08)	17.10 (8.57)	0.64	0.510	0.001	—
Total	59.15 (23.2)	53.19 (25.2)	49.99 (26.9)	3.64	0.030	0.003	1 > 2,3

SD, standard deviation; ANCOVA, analysis of ovariance.

§child's age, sex, treatment history, and caregiver's age, included as covariates.

and resistance among facilitators. This adaptation echoed Salomone et al.'s work which suggested a booster training module aimed at introducing NDBIs is needed (33). Second, ToT was adapted considerably to provide more opportunities for practice and to strengthen the facilitators' competencies. ToT was divided into two 2.5-day sessions 2 weeks apart; the facilitators were asked to practice what they had learned in the first session of the ToT during these 2 weeks. We also increased the opportunities for hands-on practice with delivering CST and facilitator–child interaction during ToT. In addition, we added three rehearsals before implementation. The unfamiliarity with the program materials may be a challenge for the facilitators when delivering CST (30). The added rehearsals were key to ensuring that the facilitators were well prepared to deliver CST to the families. The first rehearsal, prepared the facilitators for the first home visit and first three group sessions. In the second rehearsal, the facilitators rehearsed the second home visit and the fourth through sixth group sessions. The third rehearsal covered the last home visit and seventh through ninth group sessions. To ensure the facilitators delivered CST faithfully, the master trainers closely supervised the group sessions, home visits, and telephonic sessions. Established recording forms for home visits and telephonic sessions were essential for the master trainers to evaluate and provide feedback. Their supervision encouraged the new facilitators to strengthen their skills and correctly implement all CST procedures.

We adapted ToT on the basis of the learning framework, which involved three key components: knowledge, skills, and attitude (42). Comprehensive background knowledge of NDBIs (provided in the beginning of ToT) facilitated the learning of knowledge of CST. However, developing CST skills and changing attitudes toward early interventions are more challenging than acquiring knowledge. The observation and hands-on practices play a critical role in bridging the gap between "knowing what" and "knowing how" (43). The facilitators received more hands-on practices with children in ToT, observed the master trainers conducting the first home visit and discussed their home visits and telephonic sessions with the master trainers. These practices accelerated and facilitated the development of CST skills and strengthened the facilitators' competencies (44). Thus, our adaptations provided a feasible model to accelerate promotion without sacrificing quality.

CST-Taiwan adopted a progressive promotional model because of the limited numbers of master trainers who could supervise new facilitators. The maximum number of new institutions under simultaneous supervision was limited to eight to consider the workload of the four master trainers. The promotional strategy was to invite and train new facilitators each year to independently implement the CST in the following years of implementation, thereby increasing the number of institutions implementing CST. The one-third facilitators reimplemented CST in the following years, implying the high acceptance among facilitators and local feasibility (33). Most facilitators experienced growth in their professional fields and empowerment by helping the families with needs. The barriers to implementation were mostly practical. The WHO CST is provided to families free of charge, which creates barriers to promotion. Because the WHO CST entails high time and human resource costs, institutions often prefer chargeable services in the health-care system. The Taiwan CST core team asked that FACT subsidize experienced institutions (i.e., the facilitators who had completely led at least one CST group under supervision); however, the program was still not cost effective from the institutions' perspective. A future direction is to embed CST into National Health Insurance in Taiwan to provide CST to communities.

This study demonstrated the effectiveness of CST-Taiwan. The caregiver outcomes indicated the promise of this program. The caregivers learned the core concepts through the nine group sessions, three home visits and seven telephonic sessions. They also reported stronger confidence in caregiving. They felt capable of interacting with their children, facilitating their development, and coping with the stress of caregiving, which is consistent with previous studies (31, 32). This effect persisted for 3 months or more, indicating long-term changes in the caregivers' behaviors and attitude. The construction of WHO CST may have contributed to this promising learning effect. First, the Key Messages and Tips were presented repeatedly during group sessions through various ways including stories, illustrations, demonstrations, discussions and practice in pairs. Individuals are more likely to master knowledge and skills if information is presented in multiple ways (45). The WHO CST also strongly emphasizes the importance of integrating the strategies into daily home routines (29). The newly acquired strategies were immediately applicable to daily life and therefore strengthened the caregivers' knowledge and skills acquisition (46). The intensive one-on-one telephonic sessions after every group sessions, one of our adaptations, may also have contributed to the learning effect (25). The CST program may provide foundational training for families new to the early intervention system. In Taiwan, the accessibility of early intervention services may be higher than that in other countries because those in approved institutions are covered by National Health Insurance and government subsidies (13, 47). However, the search for services may lead to therapy shopping among parents (47), especially those in urban areas. For families who had participated in several interventions, CST may encourage families with children who previously received interventions to select evidence-based and effective interventions for their children (48). On the other hand, receiving CST also ease the caregivers' confusion of whether they are facilitating their child correctly, this is similar to the report from India addressing the positive experience toward CST program from the caregivers (31). The CST program can also complement the intervention service system by strengthening caregivers' skills. The improvement in caregiving knowledge and confidence persisted for all types of caregivers (in terms of age and education level). This indicates that CST can benefit caregivers of all backgrounds.

We observed that the severity of the children's symptoms of autism was significantly lower after the CST intervention, with adjustment for potential confounding factors (except for sensory/cognitive awareness). The change in communication, sociability, and the total score persisted for 3 months or more. The results suggest that CST reduced core autistic symptoms with consideration for potential confounders. The significant improvement of children's communication is also found in the pilot groups in India (31). To be noted, the improvement of children's gesture use in communication, which is a crucial component in CST, was reported in the randomized controlled trial study in Italy (32). However, there was no item assessing the gesture use in the communication subscale of ATEC, yielding a limitation of this study. The significant improvement in communication and sociability but not behaviors may have resulted from the order of the CST sessions and the measurement tool (i.e., ATEC). The sessions addressing communication and sociability were in the first half of the program. The caregivers started to learn and practice strategies to facilitate communication and sociability among the children earlier than they learned strategies for emotional behavioral regulation. The WHO CST sessions are arranged in a hierarchy; earlier sessions form the basis for the latter sessions, that is, the caregivers' role play and at-home practice during each session incorporate all strategies from prior sessions. Most caregivers set communication as their main goals at the first home visit. Because we provided individual sessions after each group sessions, the practice of skills to facilitate the children's communication was frequently addressed in telephonic sessions. Therefore, an accumulated effect of practicing these skills may have contributed to the significant improvement. The "dosage' of practicing behavioral regulation strategies may not have been sufficient to create a significant improvement at the end and follow-up time points. However, further examination to test this hypothesis is required. In addition, more realistic examples of strategies for challenging behaviors in the CST content had been called for (33), indicating that the needs of some caregivers for behavior regulation strategies were not fully covered in the current version of the CST materials. Behavioral outcomes are included in the physical/health/behaviors subscale in ATEC. The subscale consisted of not only items assessing behaviors but also items assessing toileting (not covered in CST), self-care and autistic rigidity. Thus, the nonsignificance of the changes in behavior may have been related to the measurement tool. Future research with different measurements of behavioral regulation should be considered. A longitudinal epidemiological study observed a significant reduction in ATEC scores over time (41, 49); the changes in the total score were large in younger children, who were the majority in this study. Thus, this study cannot rule out the effects of natural developmental factors on effectiveness because we did not have a control group to demonstrate that CST, rather than time, led to improvement. The characteristics of the children and caregivers determine the effectiveness of parent-mediated interventions (50). Additional mediation analyses with detailed characteristics information are required to determine relationships between effectiveness and associated factors.

The strength of this study is the establishment of a strong promotional model to ensure quality and expand the program. Although the results indicate the promise of CST-Taiwan, this study has several limitations. The main limitation of this nonexperimental study is the lack of a control group, which weakens the results. To match the original target population for the WHO CST, CST-Taiwan was open to families with children with developmental concerns. Therefore, a confirmed diagnosis or assessments were not necessarily required, which may have yielded unexpected confounders. Most caregivers had a high education level (the majority had a college degree or higher). Thus, the findings may not be generalizable to caregivers with low education levels. Additional child and caregiver characteristics should be considered in further research (e.g., types and lengths of treatments prior to CST, children's cognitive and language levels of the children, and families' socioeconomic status).

This study revealed that the adapted format of the WHO CST, namely CST-Taiwan, and its promotional model are feasible. CST-Taiwan has significantly improved caregivers' knowledge and confidence. It has also reduced the autistic symptoms among the children overall. The follow-up results suggested that the positive effects remained even after the program.

Data availability statement

The datasets generated and analyzed during the current study are not publicly available due to the research ethics regulation in Taiwan. However, they are available from the corresponding author on reasonable request.

Ethics statement

Ethical review and approval was not required for the study on human participants in accordance with the local legislation and institutional requirements. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

Author contributions

W-TS led the team as the principal investigator of WHO CST Team: Taiwan Adaptation and Implementation Project. Y-NC, W-CT, and H-YL are the co-principal investigators of the project. S-CL, M-NH, G-JS. and H-MC are master trainers providing trainings for facilitators in Taiwan. T-JL contributed to administrative works including seeking for financial resource and connecting with the governmental agency. AS introduced

WHO CST to Taiwan and attended preparatory and review meetings to provide comments for the development of CST-Taiwan. Y-CC contributed to technical supports including training of master trainers, verifying the fidelity of facilitators and master trainers, and approved all technical adaptations of CST-Taiwan. The WHO CST Team developed the CST materials and provided the monitoring and evaluation framework. G-JS and W-TS analyzed the data and wrote the manuscript with input from all authors. All authors contributed to the article and approved the submitted version.

Funding

Open access funding provided by Foundation for Autistic Children and Adults in Taiwan. This study was supported by Social and Family Affairs Administration Ministry of Health and Welfare and Yong-Tai Chen Charitable Trust.

Acknowledgments

The WHO CST Team includes: (in alphabetical order), Felicity Brown, Laura Pacione, Erica Salomone, Stephanie Shire, and Chiara Servili. We thank three WHO-AS CST trainers, Y-CC, Lucia Murillo Chacko, and Melanie Hsu, for training the master trainers in Taiwan. We thank all the institutions and facilitators for implementing CST and their role in data collection at each of the recruiting sites. We also thank the executive assistant, Yu-Ying Chen, for the data collecting and administrative support. Last but not least, the enormous contributions of the participating families in this study are gratefully acknowledged.

Conflict of interest

Author AS was employed by Autism Speaks.

The remaining authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2022.904380/full#supplementary-material

References

1. American Psychiatric Association. *Diagnostic and statistical manual of mental disorders (DSM-5®)*. (Washington, DC: American Psychiatric Pub) (2013).

2. Zeidan J, Fombonne E, Scorah J, Ibrahim A, Durkin MS, Saxena S, et al. Global prevalence of autism: A systematic review update. *Autism Res.* (2022) 15:778–90. doi: 10.1002/aur.2696

3. Lord C, Charman T, Havdahl A, Carbone P, Anagnostou E, Boyd B, et al. The Lancet Commission on the future of care and clinical research in autism. *Lancet.* (2022) 399:271–334. doi: 10.1016/S0140-6736(21)01541-5

4. Chen Y-L, Chen WJ, Lin K-C, Shen L-J, Gau SS-F. Prevalence of DSM-5 mental disorders in a nationally representative sample of children in Taiwan: methodology and main findings. *Epidemiol Psychiatr Sci.* (2019) 29:e15. doi: 10.1017/S2045796018000793

5. Sheldrick RC, Carter AS. State-level trends in the prevalence of autism spectrum disorder (ASD) from 2000 to 2012: a reanalysis of findings from the autism and developmental disabilities network. *J Autism Dev Disord.* (2018) 48:3086–92. doi: 10.1007/s10803-018-3568-z

6. Blaxill M, Rogers T, Nevison C. Autism tsunami: the impact of rising prevalence on the societal cost of autism in the United States. *J Autism Dev Disord.* (2021) 52:2627–43. doi: 10.1007/s10803-021-05120-7

7. Baxter AJ, Brugha T, Erskine HE, Scheurer RW, Vos T, Scott JG. The epidemiology and global burden of autism spectrum disorders. *Psychol Med.* (2015) 45:601–13. doi: 10.1017/S003329171400172X

8. Papadopoulos D. Mothers' experiences and challenges raising a child with autism spectrum disorder: A qualitative study. *Brain Sci.* (2021) 11:309. doi: 10.3390/brainsci11030309

9. Eldevik S, Hastings RP, Hughes JC, Jahr E, Eikeseth S, Cross S. Meta-analysis of early intensive behavioral intervention for children with autism. *J Clin Child Adolesc Psychol.* (2009) 38:439–50. doi: 10.1080/15374410902851739

10. Reichow B, Barton EE, Boyd BA, Hume K. Early intensive behavioral intervention (EIBI) for young children with autism spectrum disorders (ASD). *Cochrane Database Syst Rev.* (2012) 10:CD009260. doi: 10.1002/14651858.CD009260.pub2

11. Tiede G, Walton KM. Meta-analysis of naturalistic developmental behavioral interventions for young children with autism spectrum disorder. *Autism.* (2019) 23:2080–95. doi: 10.1177/1362361319836371

12. Ministry of Health and Welfare in Taiwan. *Number of Children with Developmental Problems for Early Intervention Services.* (2021). Available online at: https://www.mohw.gov.tw/dl-22262-b0439eab-2785-467c-8ad4-d7b83c211914.html (accessed March 11, 2022).

13. Huang P-H. The development and current situation of the early intervention for children with developmental delay in Taiwan. *Int J Child Care Educ Policy.* (2007) 1:45–58. doi: 10.1007/2288-6729-1-1-45

14. Chu C-L, Chiang C-H, Wu C-C, Hou Y-M, Liu J-H. Service system and cognitive outcomes for young children with autism spectrum disorders in a rural area of Taiwan. *Autism.* (2017) 21:581–91. doi: 10.1177/1362361316664867

15. Kaminski JW, Valle LA, Filene JH, Boyle CL, A. meta-analytic review of components associated with parent training program effectiveness. *J Abnorm Child Psychol.* (2008) 36:567–89. doi: 10.1007/s10802-007-9201-9

16. Oono IP, Honey EJ, McConachie H. Parent-mediated early intervention for young children with autism spectrum disorders (ASD). *Evid Based Child Health.* (2013) 8:2380–479. doi: 10.1002/ebch.1952

17. Postorino V, Sharp WG, McCracken CE, Bearss K, Burrell TL, Evans AN, et al. A systematic review and meta-analysis of parent training for disruptive behavior in children with autism spectrum disorder. *Clin Child Fam Psychol Rev.* (2017) 20:391–402. doi: 10.1007/s10567-017-0237-2

18. Nevill RE, Lecavalier L, Stratis EA. Meta-analysis of parent-mediated interventions for young children with autism spectrum disorder. *Autism.* (2018) 22:84–98. doi: 10.1177/1362361316677838

19. Lovaas OI, Koegel R, Simmons JQ, Long JS. Some generalization and follow-up measures on autistic children in behavior therapy. *J Appl Behav Anal.* (1973) 6:131–65. doi: 10.1901/jaba.1973.6-131

20. Reichow B, Kogan C, Barbui C, Smith I, Yasamy MT, Servili C. Parent skills training for parents of children or adults with developmental disorders: systematic review and meta-analysis protocol. *BMJ Open.* (2014) 4:e005799. doi: 10.1136/bmjopen-2014-005799

21. Matson ML, Mahan S, Matson JL. Parent training: a review of methods for children with autism spectrum disorders. *Res Autism Spectr Disord.* (2009) 3:868–75. doi: 10.1016/j.rasd.2009.02.003

22. Lichtlé J, Downes N, Engelberg A, Cappe E. The effects of parent training programs on the quality of life and stress levels of parents raising a child with autism spectrum disorder: A systematic review of the literature. *Rev J Autism Dev.* (2019) 7, 242–62. doi: 10.1007/s40489-019-00190-x

23. McConachie H, Diggle T. Parent implemented early intervention for young children with autism spectrum disorder: a systematic review. *J Eval Clin Pract.* (2007) 13:120–9. doi: 10.1111/j.1365-2753.2006.00674.x

24. Aldred C, Green J, Emsley R, McConachie H. Brief report: Mediation of treatment effect in a communication intervention for pre-school children with autism. *J Autism Dev Disord.* (2012) 42:447–54. doi: 10.1007/s10803-011-1248-3

25. Kasari C, Gulsrud A, Paparella T, Hellemann G, Berry K. Randomized comparative efficacy study of parent-mediated interventions for toddlers with autism. *J Consult Clin Psychol.* (2015) 83:554. doi: 10.1037/a0039080

26. Bearss K, Johnson C, Smith T, Lecavalier L, Swiezy N, Aman M, et al. Effect of parent training vs parent education on behavioral problems in children with autism spectrum disorder: a randomized clinical trial. *JAMA.* (2015) 313:1524–33. doi: 10.1001/jama.2015.3150

27. World Health Organization. *WHO Caregivers Skills Training for Families of Children with Developmental Delays and Disorders*. World Health Organization (n.d.). Available from: https://www.who.int/teams/mental-health-and-substance-use/treatment-care/whocaregivers-skills-training-for-families-of-children-with-developmental-delaysand-disorders (accessed March 11, 2022).

28. World Health Organization. *mhGAP Intervention Guide for Mental, Neurological and Substance Use Disorders in Non-Specialized Health Settings: Mental Health Gap Action Programme (mhGAP)*. (2016). Geneva, Switzerland: World Health Organization.

29. Salomone E, Pacione L, Shire S, Brown FL, Reichow B, Servili C. Development of the WHO caregiver skills training program for developmental disorders or delays. *Front Psychiatry.* (2019) 10:769. doi: 10.3389/fpsyt.2019.00769

30. Tekola B, Girma F, Kinfe M, Abdurahman R, Tesfaye M, Yenus Z, et al. Adapting and pre-testing the World Health Organization's Caregiver Skills Training programme for autism and other developmental disorders in a very low-resource setting: findings from Ethiopia. *Autism.* (2020) 24:51–63. doi: 10.1177/1362361319848532

31. Sengupta K, Shah H, Ghosh S, Sanghvi D, Mahadik S, Dani A, et al. World Health Organisation-Caregiver Skills Training (WHO-CST) Program: feasibility of delivery by non-specialist providers in real-world urban settings in India. *J Autism Dev Disord.* (2021) 1–18. doi: 10.1007/s10803-021-05367-0

32. Salomone E, Settanni M, McConachie H, Suma K, Ferrara F, Foletti G, et al. Pilot randomized controlled trial of the WHO caregiver skills training in public health services in Italy. *J Autism Dev Disord.* (2021):1-15. doi: 10.1007/s10803-021-05297-x

33. Salomone E, Ferrante C, Salandin A, Ferrara F, Torchio E, Foletti G, et al. Acceptability and feasibility of the World Health Organization's caregiver skills training implemented in the italian National Health System. *Autism.* (2022) 26:859–74. doi: 10.1177/13623613211035228

34. Singh NN, Curtis WJ, Ellis CR, Nicholson MW, Villani TM, Wechsler HA. Psychometric analysis of the family empowerment scale. *J Emot Behav Disord.* (1995) 3:85–91. doi: 10.1177/106342669500300203

35. Geier DA, Kern JK, Geier MR, A. Comparison of the Autism Treatment Evaluation Checklist (ATEC) and the Childhood Autism Rating Scale (CARS) for the Quantitative Evaluation of Autism. *J Ment Health Res Intellect Disabil.* (2013) 6:255–67. doi: 10.1080/19315864.2012.681340

36. Magiati I, Moss J, Yates R, Charman T, Howlin P. Is the autism treatment evaluation checklist a useful tool for monitoring progress in children with autism spectrum disorders? *J Intellect Disabil Res.* (2011) 55:302–12. doi: 10.1111/j.1365-2788.2010.01359.x

37. Wang S-C. *A research of social support, empowerment and quality of life of parents with developmental delay children who have accepted early intervention services in Taipei City and New Taipei City*. Taiwan: National Taiwan Normal University (2012).

38. Little RJ, Rubin DB. The analysis of social science data with missing values. *Sociol Methods Res.* (1989) 18:292–326. doi: 10.1177/0049124189018002004

39. White IR, Royston P, Wood AM. Multiple imputation using chained equations: issues and guidance for practice. *Stat Med.* (2011) 30:377–99. doi: 10.1002/sim.4067

40. Jennings MA, Cribbie RA. Comparing pre-post change across groups: Guidelines for choosing between difference scores, ANCOVA, and residual change scores. *J Data Sci.* (2016) 14:205–29. doi: 10.6339/JDS.201604_14(2).0002

41. Mahapatra S, Khokhlovich E, Martinez S, Kannel B, Edelson SM, Vyshedskiy A. Longitudinal epidemiological study of autism subgroups using Autism Treatment Evaluation Checklist (ATEC) Score. *J Autism Dev Disord.* (2020) 50:1497–508. doi: 10.1007/s10803-018-3699-2

42. Kraiger K, Ford JK, Salas E. Application of cognitive, skill-based, and affective theories of learning outcomes to new methods of training evaluation. *J Appl Psychol.* (1993) 78:311–28. doi: 10.1037/0021-9010.78.2.311

43. Geiger KB, LeBlanc LA, Hubik K, Jenkins SR, Carr JE. Live training versus e-learning to teach implementation of listener response programs. *J Appl Behav Anal.* (2018) 51:220–35. doi: 10.1002/jaba.444

44. Kohrt BA, Jordans MJD, Rai S, Shrestha P, Luitel NP, Ramaiya MK, et al. Therapist competence in global mental health: Development of the ENhancing Assessment of Common Therapeutic factors (ENACT) rating scale. *Behav Res Ther.* (2015) 69:11–21. doi: 10.1016/j.brat.2015.03.009

45. Dunst CJ, Trivette CM. Let's be PALS: An evidence-based approach to professional development. *Infants Young Children.* (2009) 22:164–76. doi: 10.1097/IYC.0b013e3181abe169

46. Friedman M, Woods J, Salisbury C. Caregiver coaching strategies for early intervention providers: moving toward operational definitions. *Infants Young Children.* (2012) 25:62–82. doi: 10.1097/IYC.0b013e31823d8f12

47. Chao K-Y, Chang H-L, Chin W-C, Li H-M, Chen S-H. How Taiwanese parents of children with autism spectrum disorder experience the process of obtaining a diagnosis: a descriptive phenomenological analysis. *Autism.* (2018) 22:388–400. doi: 10.1177/1362361316680915

48. Rivard M, Morin M, Mercier C, Terroux A, Mello C, Lépine A. Social validity of a training and coaching program for parents of children with autism spectrum disorder on a waiting list for early behavioral intervention. *J Child Fam Stud.* (2017) 26:877–87. doi: 10.1007/s10826-016-0604-5

49. Mahapatra S, Vyshedsky D, Martinez S, Kannel B, Braverman J, Edelson SM, et al. Autism Treatment Evaluation Checklist (ATEC) Norms: A "Growth Chart" for ATEC Score Changes as a Function of Age. *Children.* (2018) 5:25. doi: 10.3390/children5020025

50. Trembath D, Gurm M, Scheerer NE, Trevisan DA, Paynter J, Bohadana G, et al. Systematic review of factors that may influence the outcomes and generalizability of parent-mediated interventions for young children with autism spectrum disorder. *Autism Res.* (2019) 12:1304–21. doi: 10.1002/aur.2168

frontiers | Frontiers in Psychiatry

TYPE Original Research
PUBLISHED 07 September 2022
DOI 10.3389/fpsyt.2022.915263

Acceptability and feasibility of the World Health Organization's Caregiver Skills Training Programme (WHO CST) delivered *via* eLearning, videoconferencing, and in-person hybrid modalities in Hong Kong

Janet Siu-Ping Lau[1,2]*, Simon Man-Kin Lai[1], Florence To-Sau Ip[1], Paul Wai-Ching Wong[1], WHO CST Team[3], Chiara Servili[3], Erica Salomone[3,4], Laura Pacione[3,5], Stephanie Shire[6] and Felicity L. Brown[7,8]

[1]The University of Hong Kong, Hong Kong, Hong Kong SAR, China, [2]WHO CST Regional Technical Focal Point, Geneva, Switzerland, [3]Department of Mental Health and Substance Use, World Health Organization, Geneva, Switzerland, [4]The University of Milano-Bicocca, Milan, Italy, [5]Division of Child and Youth Mental Health, Department of Psychiatry, University of Toronto, Toronto, ON, Canada, [6]The University of Oregon, Eugene, OR, United States, [7]Research and Development Department, War Child Holland, Amsterdam, Netherlands, [8]Amsterdam Institute of Social Science Research, University of Amsterdam, Amsterdam, Netherlands

OPEN ACCESS

EDITED BY
Maria Luisa Scattoni,
National Institute of Health (ISS), Italy

REVIEWED BY
Yuan Cao,
Hong Kong Polytechnic University,
Hong Kong SAR, China
James Lee,
Juniper Gardens Children's Project,
United States

*CORRESPONDENCE
Janet Siu-Ping Lau
drjanetlau@gmail.com

SPECIALTY SECTION
This article was submitted to Autism, a section of the journal Frontiers in Psychiatry

RECEIVED 07 April 2022
ACCEPTED 27 July 2022
PUBLISHED 07 September 2022

CITATION
Lau J-S-P, Lai S-M-K, Ip F-T-S, Wong P-W-C, WHO CST Team, Servili C, Salomone E, Pacione L, Shire S and Brown FL (2022) Acceptability and feasibility of the World Health Organization's Caregiver Skills Training Programme (WHO CST) delivered *via* eLearning, videoconferencing, and in-person hybrid modalities in Hong Kong. *Front. Psychiatry* 13:915263. doi: 10.3389/fpsyt.2022.915263

Background: Local children with developmental disabilities were deprived of learning opportunities due to recent social and health incidents, resulting in elevating challenging behaviors and familial conflicts. This study explored the acceptability and feasibility of the World Health Organization's Caregiver Skills Training Programme (WHO CST) in alternative delivery modes under new normal and post COVID-19 period.

Method: CST was delivered *via* eLearning (EL), videoconferencing (VC), and in-person hybrid (IP) modes to 34 parent-child dyads, being randomly assigned to modes of asynchronous non-interfering EL ($n = 9$), synchronous with online coaching VC ($n = 7$), synchronous with in-person coaching IP ($n = 9$) and Wait-list Control WLC ($n = 9$). Data from two standardized scales of General Health Questionnaire (GHQ-12) and Strengths and Difficulties Questionnaire (SDQ), and Post-session and Home Visit Feedback Form by Caregivers that included both structured and open-ended questions were collected before and after intervention. Both quantitative and qualitative approaches were used in studying the collected data.

Results: High levels of acceptability and feasibility of the training programme were supported by ratings on comprehensiveness and relevance, agreement with their personal values, duration, and usefulness. IP and VC groups yielded more positive changes than EL and WLC groups with 3, 16, 13, and −3% in General Health Questionnaire (GHQ-12), −13, −15, −6 and 0% in Difficulties-total, and 36.5, 35.5, 5.8 and 2.4% in Prosocial Scale at Strengths and Difficulties Questionnaire (SDQ) for EL, VC, IP, and WLC groups respectively from baseline to 12 weeks after intervention. Results from two standardized scales echoed with qualitative observations that the programme helped improve caregivers' well-being, child's communication, and behaviors across intervention groups.

Conclusions: Current findings revealed that CST delivered in three alternative modes were acceptable and feasible, and yielded positive impacts toward both caregivers and children. In-person coaching, and skill-practicing sessions were effective in mitigating child's challenging behaviors while personal interaction, either face-to-face or virtual, is a significant factor in uplifting caregivers' well-being, whereas the self-learning model was appreciated by the busy caregivers. In clinical practice, needs and goals of families and the constraints of remote interventions at the settings should be balanced.

KEYWORDS

World Health Organization's Caregiver Skills Training Programme, WHO CST, eLearning, videoconferencing, in-person, developmental delays or disabilities, parenting, COVID-19

Introduction

Twenty-four months under restricted movement and social interactions, people in Hong Kong have been exploring new ways of living to restore normal life. However, twenty-four months in early childhood is more than one-third of the "golden development period" (1) which comprises more than 80% of a person's brain development (2, 3). During this critical fraction of development, children can learn and experience rapid development in speech, motor, cognition, and emotions (1, 4) while others require significant support to mitigate their learning needs. Unfortunately, most of the people in Hong Kong have been experiencing an "unprecedentedly stretched lockdown" for over 2 years. The social upheaval due to large protests in June 2019 led to school suspension as transportation was handicapped with potential dangers on the street for more than 6 months. This was followed by social distancing rules that were enacted in Hong Kong due to the pandemic in January of the following year. Therefore, children were kept confined at home and received online schooling for another six months. Lockdown is, indeed, meant to protect the vulnerable, however, for children with developmental disorders or delays the loss of face-to-face schooling and adequate services leads to a dearth of learning opportunities, and oftentimes, elevated unresolved challenging behaviors (5), as well as familial conflicts, including child abuse and domestic violence within the households (6–8). In view of this service gap and elevated parenting needs during this challenging time, this study was designed to examine the acceptability and feasibility of a worldwide, evidence-based parenting programme, the World Health Organization's Caregiver Skills Training Programme (WHO CST or CST), delivered *via* eLearning (EL), videoconferencing (VC), and in-person hybrid (IP) modes in Hong Kong. This study examines alternative delivery practices leveraging remote delivery modalities to provide access to services under challenging conditions to help these parents stay positive and make use of their time for teaching their children at home under the new normal.

Background

"Developmental disabilities" (DD) is a comprehensive term to describe the lifelong chronic neurodevelopmental conditions of early childhood that include but not limited to disorders of intellectual development and autism spectrum disorder (ASD), which share common features: discrepancies in physical, cognitive, linguistic, social, and adaptive functioning from the normative developmental milestones (9).

It is estimated that 65 out of 10,000 people have autism globally (10). In the United States, 1 in 44 of the 8-year-old population was diagnosed with ASD in 2018 (11). In Hong Kong, approximately 9.5% of the population with ASD reported various levels of difficulties (12) and the newly diagnosed cases have increased 3-fold, from 755 in 2006, to 2,021 in 2015 (13). On top of the queuing time for diagnosis, the long waiting time ranging from 12 to 18 months (14) for special educational support puts caregivers in a stressful situation as support is minimal in meeting the children's and their own needs (15).

Parenting can become more stressful for parents of children with ASD. Children with developmental delays or disabilities may need additional support to acquire daily living skills and communicate with others (16). Parents can benefit from services designed to help caregivers learn strategies to support their children with special needs. However, engaging in these services can add burden (17) on top of a heavy load of daily chores

(18) as well as the challenges arising from the affected family functioning and their personal well-being (16).

With the restrictions of the social movements followed by the onset of COVID-19 in June 2019, children in Hong Kong have been unprecedentedly locked down. Due to the lockdown, children with developmental delays/disabilities could not access intervention because servicing centers were closed. The shutdown was meant to protect these vulnerable children. Nevertheless, staying at home, having online classes with plenty of distractions, and being unable to access adequate services did not only limit learning opportunities and energy outlets for children with higher needs, but led to the elevation of children's challenging behaviors and familial conflicts within the households. While the psychological impacts of being locked down were yet to be reported, the burden of parenting children with special needs was heightened with the sudden cancellation of school, unprepared daily nurturing roles, and confining children with special needs in an average living space of 13.5 square meters per person (19).

On the other hand, the pandemic also impacted the lives of working caregivers. Due to the lockdown situation, flexibility became even more essential as parents were spending time taking care of the stay-at-home school-aged children in the daytime while also working from home. With more time nurturing their children on their own than before, these caregivers experienced a sense of helplessness or inadequacy in their parenting (5). Wong and his colleagues (5) also found that caregivers indicated the need for a programme that could be more flexible in terms of time and place, to fit the office schedules of working parents. In addition, some practitioners revealed the challenges of having face-to-face home visits under the health and safety guidelines and regulations of their affiliated organizations. Thus, to address all these challenges, these voices called for alternative delivery modes of parenting programmes in Hong Kong, especially in this critical period.

There is growing evidence that caregiver-mediated programmes can empower caregivers to support children's communication and engagement (20), and help reduce challenging behaviors (21, 22), in turn, leading to better developmental, behavioral, and family functioning outcomes (21, 22). Furthermore, caregivers' stress can be alleviated by relevant interventions by encouraging caregivers to reach out for professional and informal support (23–25).

Following advocacy of high-quality development for children under the WHO Global Strategy for Women's, Children's and Adolescents' Health (26) and the growing needs of families of children with developmental disabilities (27), the World Health Organization (WHO) developed the Caregiver Skills Training Programme for Families of Children with Developmental Delays and Disabilities (CST). The CST programme is based on a common elements approach and informed by the findings of numerous meta-analyses (21, 22, 28) and was developed in consultation with experts and parents' associations from all WHO regions with financial support from Autism Speaks (29). The programme is a scalable, affordable parenting programme which is applicable to children with developmental delays and disorders in low and high resources settings by non-specialists (30). The programme targets caregivers of children aged 2 to 9 years with a developmental delay or disability, especially in the domains of social interaction and communication, although a diagnosis is not required. The aim is to teach caregivers basic strategies to promote their child's development and adaptive behaviors, to improve daily interactions between caregiver and child, as well as support caregivers' self-care (31). Under the framework of implementation science, CST is currently in the stage of pilot-testing in more than 30 countries (31) and several randomized controlled trials are underway and more evidence-based research is undergoing publication. Overall, the programme has shown excellent acceptability and feasibility in both high- and low-resource settings (32, 35) and indicators of clinical effectiveness (33, 34).

Different delivery modes

Financial constraints, commuting difficulties, lack of childcare, geographical distance, long waiting times, and time commitments are regarded as major barriers to conventional services (36–38). Although telehealth programmes provide a solution to the traditional servicing mode, the utilization of technology in the health-related field remained stagnant due to skepticism about the potential risks and unforeseen effects of their applications (39). However, the outbreak of COVID-19 has become the turning point, driving the field to give up conventional therapeutic approaches and embrace the potential benefits of delivering services through technologies (39–43).

The enactment of social distancing measures facilitates the escalating application of technology among mental health service providers, resulting in a rapid shift from in-person mode to telehealth services (43). Previous meta-analyses had shown that tele-mental health care could generally provide effective and adaptable solutions for the care of mental issues (44). Nevertheless, professionals also reported challenges in different areas, including technical problems (45–47), privacy protection concerns (48), and the need to adapt to an unfamiliar interaction pattern (5, 49) to foster a facilitative environment and supportive relationships with clients (41).

Delivery of social services using technology

While the efficacy of applying new technologies in social services is under investigation, there have been many telehealth interventions established in the community (50). The use of modern technology can accelerate different telehealth services in different manners, such as personal tele-psychotherapy, self-directed learning, and online group workshops, including those with high complexity and long duration, which can be supplemented by different delivery modalities and instructional design calibrated to meet the needs of the target clientele (51) such as eLearning, videoconferencing, and conventional in-person modes.

eLearning

ELearning can be regarded as the dissemination of knowledge through a technology-based learning system where learners can acquire knowledge at their own pace. A meta-analysis on the effectiveness of Internet-based eLearning across different beneficiaries suggested that eLearning was at least as effective as conventional learning methods (52). Some evidence illustrated that healthcare professional behaviors were improved better than without guidance at all (51, 53). Many studies on the adaptation of in-person training or therapies for online delivery revealed positive results in knowledge attainment, stress, and fidelity to treatment plan (54–58).

Evidence-based ASD interventions have also been adapted for delivery using technology. One three-month intervention programme for children with autism delivered by an app in Australia revealed that improvements in language and social communication skills were not only found in the posttest but prolonged at 12 months post-intervention assessment (59). Wainer and Ingersoll piloted a similar web-based self-directed telehealth programme, resulting in encouraging results that participants were able to learn about reciprocal imitation training (RIT) and increase the application of RIT techniques afterwards (57). Another Self-Directed Learning Programme showed significant differences between the treatment group and the control group on all dependent measures, including implementation fidelity on Pivotal Response Training (PRT) procedures, language opportunities, functional verbal utterances, and observed parent confidence (56).

Nevertheless, the findings of these studies indicated that participants suggest adding interactive remote coaching sessions to a self-directed eLearning parenting programme, which can be crucial and essential for online parent training programmes, including weekly telephone calls or email correspondence, peer groups, and regularly personalized coaching sessions (60). Given the limited but essential need for interaction with others, research has begun to explore how to use videoconferencing to facilitate remote feedback and support for parents of children with ASD (53, 55, 61–64).

Videoconferencing

Internet-based videoconferencing technology offers a structure like face-to-face meetings, providing real-time opportunities for therapists and patients to interact from different geographic locations. Videoconferencing can serve as a sole intervention medium or a supplementary component enhancing participation and interactions between therapists and participants. Studies revealed that parents find the adoption of virtual psychotherapy or training acceptable, easy to use, and effective (49, 58, 63, 65). Several empirical studies showed that Internet-based teaching or a combination of multiple strategies can provide promising results in applied-behavior-analysis (ABA) intervention (53, 55, 63, 64, 66–68).

Videoconferencing intervention, adding to self-directed programmes can be a solid combination for the programmes that last for several weeks or months. One combined-mode programme working with families of ASD children revealed encouraging outcomes in implementing behavioral management skills for both daily and play activities (69). Another study on combined modes, including an additional 13 hours of instructional guidance and 4 hours of group coaching for children with autism showed a significant improvement in intervention techniques (70), which shows that expert guidance and support, even virtual, can be helpful to achieve implementation fidelity.

In view of skills attaining programmes *via* online means, research shows that these programmes provide promising results for both parents and their children with ASD in different aspects. A three-week online training programme on a weekly basis with instructional content, group discussions, and problem-solving support is sufficient to demonstrate behavioral improvement in autistic children and reduction in parenting stress (53). Fisher and colleagues (55) (2020) conducted a comprehensive pre-post study on teaching applied-behavior-analysis skills to parents of children with ASD to evaluate the differences between the treatment group and the control group at individual and group levels, under structured and play contexts which demonstrated a significant improvement in the treatment group for the measures at posttest. These two studies on the efficacy of applying technologies to parenting programmes for ASD or DD families showed positive outcomes; however, comparing the outcomes with those through the conventional face-to-face programme or self-directed mode is needed.

Comparison of service delivery modalities: In-person and videoconferencing modes

There is a paucity of studies comparing the effectiveness of interventions facilitated by videoconferencing with those delivered in-person conventionally. Therefore, it is difficult to conclude whether videoconferencing or in-person delivery is more effective. Some studies show that videoconferencing interventions are as effective as in-person interventions. For instance, a qualitative study aimed at improving parental mental health with a mixed mode of three face-to-face sessions and five videoconferencing sessions was conducted to explore the acceptability and feasibility of this mixed service mode. Results indicated that parents valued videoconferencing as an acceptable delivery means of intervention programme while videoconferencing sessions promoted a higher attendance (49). In Luxton and colleagues' study (71), participants were allocated to either conventional in-person settings or home-based videoconferencing modes. Results suggested that participants in both groups showed reductions in hopelessness and depressive symptoms; however, there was no statistical significance between the groups. These suggest the effectiveness comparison of the two delivery modes is inconclusive.

The current study

As highlighted by Duan and Zhu (72), sufficiently dynamic and flexible psychotherapeutic interventions should be adapted quickly to different stages of the pandemic. Emphasis on flexibility and adaptability is a common theme of research in this special issue (42). Indeed, CST was developed to be adapted to the cultural, socioeconomic, geographic, and resource context in which it is used (30). From 2018 onwards, the University of Hong Kong has adopted and implemented CST in Hong Kong. It was thereafter tested in two phases: pre-pilot and pilot tests. The current study follows the effectiveness studies on the adapted CST materials of version 1.04 being conventionally delivered while further studying different feasible remote intervention modes in Hong Kong. In view of the aforementioned service gap and elevated parenting needs during this challenging time, this study aims to examine the acceptability and feasibility of an evidence-based parenting programme, the World Health Organization's Caregiver Skills Training Programme (WHO CST), to be delivered by eLearning (EL), videoconferencing (VC), and in-person hybrid (IP) modes of delivery in Hong Kong. The programme supports caregivers' skill attainment to strengthen children's communication and joint engagement while reducing challenging behavior. Together, these skills support children's adaptive behaviors and family functioning that ultimately lead to an improved caregiver-child interaction. The programme also helps strengthen caregivers' coping skills, psychological well-being, and quality of life, as well as mitigate the stigma against developmental disorders (30).

Methodology

Participants and randomization

Participants were recruited through a convenience sampling method by sending recruitment messages *via* the university bulk email services and *via* social media to the caregivers of children suspected of having developmental needs. They gave their consent for research participation and sharing of their demographic data along with an initial set of screening questions based on the inclusion and exclusion criteria, which was done *via* the encrypted platform along with an enquiry hotline supported by a research assistant. Participants were: (a) primary caregivers residing with a target child in Hong Kong, (b) able to communicate in Cantonese, and (c) literate in Chinese. The target children were: (a) between 2 and 6 years old, (b) with symptoms of autism spectrum disorder or other developmental disorders or delays, (c) with a score of 3 or above on the Modified Checklist for Autism in Toddlers (M-CHAT). The randomization was conducted using Microsoft Excel to allocate participants to eLearning (EL), in-person hybrid (IP), videoconferencing (VC), and wait-list control (WLC) groups, with 17, 18, 17, 18 participants respectively. This study was approved by The Human Research Ethics Committee of Hong Kong University (HKU) (EA200178) and all data collection was performed in accordance with HKU guidelines and regulations.

Settings and materials

The present study took place in both the virtual online setting and HKU campus. A standard Caregiver Child Interaction (CCI) toy kit was given to each participating family upon admission as a standardized tool to record the caregiver-child interaction for the three-time points. The CCI kit included wooden blocks and plastic balls, nesting cups, toy vegetables and fruits with velcro, a non-assembled stove with kitchen utensils, figurines, and a drawing set, which covers a developmental range of play from the earliest simple play skills (e.g., roll a ball, take apart Velcro fruit) through symbolic level pretend play (e.g., pretending to be a chef, pretending a nesting cup is an airplane). Also, Participant Booklets were sent to the IP and VC participants only.

The intervention

The intervention followed the CST Facilitator Guide version 1.00 (74, 75) and the WHO Parent Skills Training Programme for Caregivers of Children with Developmental Disorders:

Monitoring and Evaluation Framework provided by the World Health Organization (75). The intervention is an originally evidence-based programme package based on the principles derived from behavioral and social learning approaches, child development theories as well as positive parenting approaches focusing on training (coaching) parents or caregivers to play and home activities as daily routines and opportunities for learning and development (32, 33). It also emphasizes a task-shifting approach, in which paraprofessionals or non-specialists can be trained to deliver the programme to parents or caregivers to support their child's engagement in daily home and play activities. It also emphasizes a transdiagnostic approach such that children do not need to meet the diagnostic criteria for ASD or other pervasive developmental disabilities in order to access the programme. The programme consists of nine group sessions and three individual home visits. The content of the sessions includes (a) promoting joint engagement (sessions 1a, 1b, and 2), (b) promoting communication (sessions 3–4), (c) promoting positive child behavior and managing challenging behaviors (sessions 5–6), (d) learning new skills (session 7), and (e) empowering participants' troubleshooting and self-care capability (session 8). The programme strategies were explained through illustrated stories, role-plays, video demonstrations, group discussions, and guidance for home practice. The skills were further strengthened with the three home visits which were held before the first group session, after session four, and after the last group session.

The programme materials were adapted and translated from English to Chinese and back-translated following the CST Draft Adaptation and Implementation Guide Field Version 1.01. The adapted content was revised and endorsed through three adaptation meetings with various stakeholders, community experts, service providers, and caregivers, and the feedback from the need assessment study (5). Minor adaptations included agreements on the name of the programme and the proposed terms in Traditional Chinese (e.g., Joint Engagement), localized daily examples (e.g., irrelevant cause to developmental delays like "Sins of Family" was not emphasized in Hong Kong cultures and was reordered on the list), and additional support to participants to join the sessions (e.g., babysitting services).

With the programme materials remained constant, only the delivery modalities varied in this study. The three experimental groups were eLearning (EL), videoconferencing (VC), and in-person hybrid (IP) modes varying with the level of remote interventions and synchronicity. Detailed adaptations for each delivery mode are described in Appendix.

eLearning

This was an asynchronous, non-interfering mode with the programme materials including the digital versions of CST participant booklets and pre-recorded videos demonstrating CST skills and strategies provided through an encrypted online platform. This was to highlight the self-learning nature that the caregivers could learn CST skills online within the assigned weeks at their own pace, and free from location and time restrictions. Participants were given a unique credential to access the programme materials on a weekly basis and to submit their answers of the post-session quizzes and feedback forms after each session *via* the platform. The administrator would send out gentle reminders to the participants who had not sent in the answers and feedback on the fifth day of the assigned week. Data collected were encrypted and could be accessed by restricted researchers. The programme schedule together with the submission of the caregiver-child interaction videos over the standard CCI kit followed that of the other two experimental groups. No practice in pairs and no coaching was involved while wellness exercises and live demonstrations were replaced by pre-recorded demonstration videos.

Videoconferencing

This was a completely online mode where all home visits and weekly group sessions were conducted by a pair of qualified facilitators *via* Zoom videoconferencing software. Participants interacted with other parents with similar parenting experiences online despite their location. They received the Participant Booklet and the standard CCI kit by post before the intervention. Wellness exercises and live demonstrations were played by pre-recorded videos. Sharing and plans for home practices were discussed in the sessions and facilitators lectured on the programme key message and skills review. All components of the home visits were conducted online, including the observation of the parent-child interaction, facilitator verbal coaching, and goal setting.

In-person hybrid

The in-person delivery mode was originally planned as face-to-face group sessions at the HKU campus and individual home visits at the participants' homes. However, following the outbreak of COVID-19, a hybrid mode was employed to meet social distancing measures while the personal skills drilling components could be preserved. The changes included turning the nine face-to-face group sessions into online sessions with Participant Booklet received in advance while the three home visits remained in-person sessions with an addition of three 1-hour live skills practice sessions at the third, sixth and ninth sessions. In this way, experiential learning of the skills and peer support was made possible. Like VC, the didactic sessions were conducted online by a pair of qualified facilitators weekly at a fixed time slot with wellness exercises and live demonstrations through playing pre-recorded videos. Sharing on and plans for home practices were discussed in the group while facilitators lectured on the programme with key message and skills review. All the home visits were conserved as the original CST home visits and conducted in person, where parent-child interaction, facilitator coaching, and goal setting were made interactive and done face-to-face with the two facilitators (75).

Master trainers and facilitators

Two years prior to this study, a pre-pilot study and a pilot study were conducted in Hong Kong to test out the Hong Kong-adapted CST materials and the knowledge transfer from the CST trainers to master trainers in Hong Kong, and further to the facilitator level. Eight master trainers were trained by CST trainers in Hong Kong in early 2019. This study was conducted by the principal master trainer who had achieved over 98% at the implementation fidelity assessment. The principal master trainer engaged in the programme adaptation and implementation process, conducted several CST groups at the pre-pilot stage, as well as trained and supervised the facilitators at the pilot stage.

Four students attaining a master's degree in Social Science at the University of Hong Kong enrolled in the standardized 7-day CST facilitator training and were thereafter supervised by the in-charge Master Trainer, including didactic theories, role-play, demonstrations, and live practices. Following two months of supervised practice, the facilitators' fidelity of CST intervention strategy implementation was assessed. Each facilitator recorded a 10-min interaction with a child with a disability while applying the strategies in the context of play and home routines. All facilitators passed the 11-item Adult/Child Interaction Fidelity Rating of version 1.04 at 75% before the intervention commenced. Furthermore, the facilitators' group facilitation skills were evaluated through a live session demonstration with their peers engaged as caregiver participants. The intervention was conducted in pairs and continual supervision was secured throughout the programme. Group facilitation skills were evaluated using the 22-item ENhancing Assessment of Common Therapeutic Factors (ENACT) (73) during the intervention.

Assessments

Baseline measures

At baseline, participants' socio-demographic information along with a screening measure, the Modified Checklist for Autism in Toddlers (M-CHAT) (76), and a set of baseline measures, including the Caregivers Skill and Knowledge questionnaire, 12-item General Health Questionnaire (GHQ-12) (77), and 25-item Strengths and Difficulties Questionnaire (SDQ) (78) were collected.

Fidelity measures

Each group was run by a pair of facilitators who were assessed and supervised by a qualified Master Trainer. Before the programme, the facilitators had undergone an intensive 7-day Training of Trainer (CST ToT), and subsequently were qualified as facilitators by achieving at or over the fidelity of 75% using the 11-item Adult/Child Interaction Fidelity Rating of version 1.04, and over 2.5 (out of four) at the score of 22-item ENhancing Assessment of Common Therapeutic Factors (ENACT) (73) after 4 months of supervised practice.

Evaluation of acceptability

The acceptability of the programme in various implementation modes was evaluated by the feedback on the comprehensiveness and relevance, value conflict, duration of each session and on perceived usefulness of home visits and live coaching after each home visit by caregivers, and the post-session feedback concerning perceived acceptability of the programme, including contents of the sessions, level of engagement, acceptance of the caregivers, and level of involvement of caregivers by facilitators.

Post-session and Home Visit Feedback Form by Caregivers

Post-session Feedback Form by Caregiver and Home Visit Caregiver Feedback Form were completed by the caregivers after each CST group session and after each home visit accordingly. Six questions in the Post-session (PS) Feedback Form by Caregivers and three questions in Home Visit (HV) Caregiver Feedback Form were especially adopted to assess the acceptability of the programme being conducted in different modes.

Responses over the Post-session Feedback by Caregiver were selected to assess the comprehensiveness and relevance of each session. Comments of two other questions were evaluated to study the agreement with participants' and their family members' personal values. The appropriateness of time used in each group session and each home visit were evaluated. Lastly, the perceived usefulness of home visits and live coaching comments were asked in the Home Visit Feedback Form by Caregivers.

Facilitator's Feedback Form

Facilitator's Feedback Form concerning the perceived acceptability of the programme, including contents of the sessions, level of engagement, acceptance of the caregivers, and level of involvement of caregivers of each group session were collected after each session.

Evaluation of feasibility

Feasibility of CST conducted through different delivery modes were assessed in terms of participants' completion rate, attendance, adherence to home practice, observer's feedback on the involvement of the participants and implementation of the group sessions. These aspects were included in the following measures.

Caregiver diary

Caregivers' adherence to home practice was collected after home visit 2 (T1) and home visit 3 (T2). Caregivers reported their practice frequency on a weekly basis and the amount of time they practiced the "Skills and Strategies" each day.

Observer's Feedback Form

Group sessions' intervention fidelity was reported by observers after each group session, who was the facilitator not in-charge of the teaching that session, in rating the participants' degree of comfort, enthusiasm/interest, and level of involvement in planning home practice.

Facilitator's Feedback Form

Complexity, amount of the sessions' contents, and perceived preparation for the sessions were rated by the teaching facilitator to evaluate the implementation of the programme after each group session.

Clinical outcome measures

Quality of Life as parent outcome

The General Health Questionnaire (GHQ-12) is a 12 self-assessed instrument to reflect the mental wellness of respondents on a 4-point Likert-type scale from 0 (not at all) to 3 (always). Confirmatory factor analyses (CFA) study (77) showed that the general factor was strongly associated with symptoms of insomnia and mental health. The rating of the 12 questions (with those of questions 1, 3, 4, 7, 8, and 12 being reversed) were summed up to get the total score with a cut-off score of 12. The lower the score, the better the wellness. Participants were asked to complete the form at baseline (T0), after home visit 2 (T1), and after home visit 3 (T2) to investigate if there were changes in the caregivers' perception of their qualities of life along with the intervention, and to compare if there were any changes between learning modes.

Strengths and Difficulties Questionnaire as child outcome

The Strengths and Difficulties Questionnaire (SDQ) is designed to score children, aged 3 to 16, on their behaviors. Confirmatory factor analysis (79) demonstrated strong evidence for convergent and discriminant validity. There are 25 items in SDQ classified into 5 scales including one strength scale as prosocial skill and four difficulty scales in conduct problem, hyperactivity, emotional problem, and peer problem. These four scales are added together to generate a total difficulty score. Each scale consists of 5 items with a 3-point Likert-type scale, with "somewhat true" always scored as 1, but the scoring of "not true" and "certainly true" varies with the item scored as 2 or 0. The lower the score represents the better the condition. Participants were asked to complete the form at baseline (T0), after home visit 2 (T1), and after home visit 3 (T2).

Qualitative measures

Open-ended questions from Post-session and Home Visit Feedback Form by Caregivers

Comments from three open-ended questions in Post-session (after each group session) and Home Visit Feedback Form (after each home visit) by Caregivers were triangulated and supplemented with the quantitative data in this study. The questions were on the suggestions to improve each session, improvements to each home visit, and video-recording arrangement of each home visit. The feedback was investigated in detail and outstanding perspectives were reported.

Data analysis

This study employed the mixed-methods approach. Quantitative data were collected through questionnaires from CST protocols (75) and clinical outcome measures while qualitative data from the open-ended comments were gathered to corroborate and triangulate findings.

Quantitative data analysis

Data were input into SPSS 25 for cleaning, managing, and analysis. Central tendency and variance were used to characterize the general trend of the data. Descriptive analyses were conducted on all measures. In view of the small sample size, descriptive statistical analyses would be adequate to reflect the reality of implementing the CST in Hong Kong settings. Two-way ANOVAs were conducted to test if there were differences across the four conditions and between the T0 and T2 for GHQ-12 and SDQ.

Qualitative data analysis

A thematic analysis (80, 81) was done on qualitative data collected from each session and each home visit given by all attending participants across the three intervention groups while wait list control groups only gave comments on each home visit. The comments were recorded and collected in written form from the encrypted platform. The data were sent to two coders to read and generate the codes independently before they came to a consensus to identify a final set of codes. They continued to review the code set and their theme and sub-theme classification before the actual coding process on all the comments. The participants were coded with a group identifier as EL01, EL02, IP01, IP02, VC01, VC02, WLC01, WLC02 (...) to uphold privacy and confidentiality.

Results

Participants

Referring to Figure 1, a total of 82 participants showed interest in the programme, and 70 caregivers who fulfilled the criteria stated in 2.1.1 were considered eligible for the study.

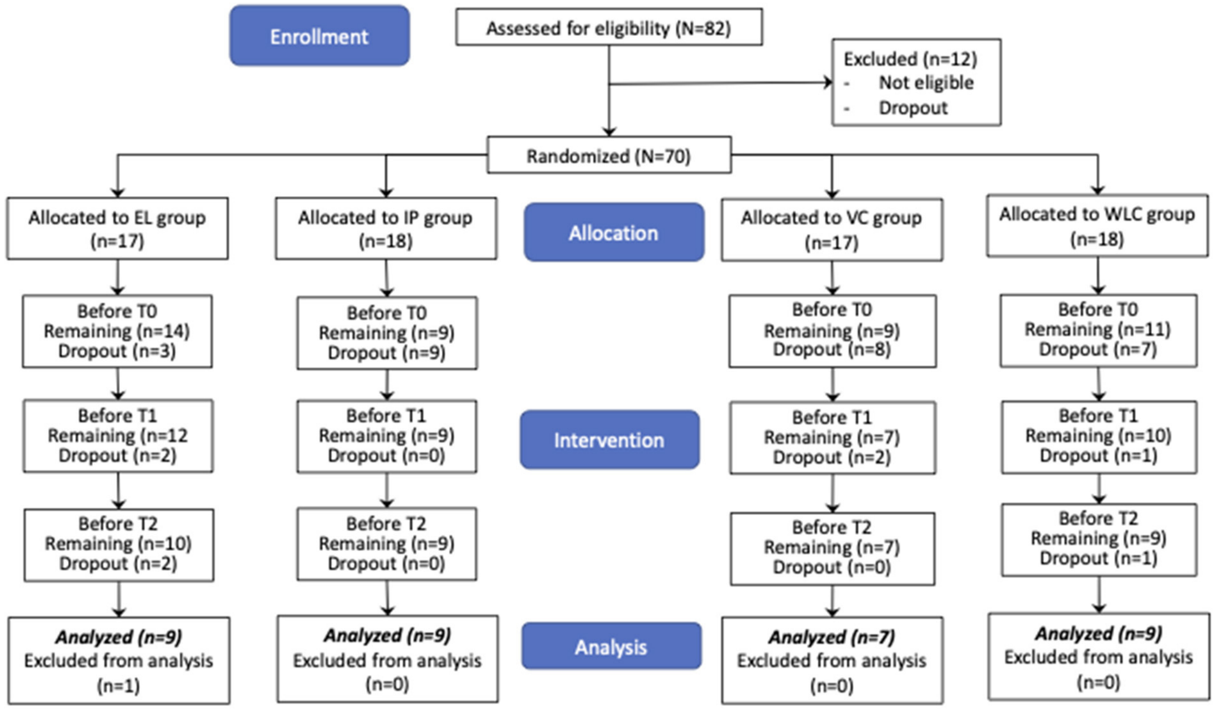


FIGURE 1
CONSORT 2010 flow chart for participant recruitment & processing.

The participants were randomized through excel functions and assigned into eLearning (EL), in-person hybrid (IP), videoconferencing (VC), and wait-list control (WLC) groups, with 17, 18, 17, 18 participants, respectively. They gave the Informed Consent Form. Three EL, nine IP participants, eight VC, and seven WLC participants withdrew before any intervention. A total of 8 participants (EL = 4, VC = 2, WLC = 2) withdrew from intervention due to the worsening pandemic situation. By T2, a total of 34 participants across EL ($n = 9$), IP ($n = 9$), VC ($n = 7$), and WLC ($n = 9$) participants completed the programme.

Eighty-eight percent (30 out of 34) of the caregivers were female, and their average age was 39 years (SD = 3.52; range = 32–47). Eighty-eight percent of them were born in Hong Kong, and the rest were born in Mainland China. All completed secondary school or above, of which sixty-two percent had completed tertiary education. Over 60% of the children had more than one caregiver. Sixty-eight percent of the participants had a full-time job, one of the 4 part-time parents worked at home, while others did not go to work. Eighty-two percent (28 out of 34) of the target children were male, and the average age of the children was 4.4 years (SD = 1.4; range = 2–7). All of them were born in Hong Kong. The demographic characteristics of the participants of all groups were summarized in Table 1. Except for "who is the main caregiver," there are no significant differences in other demographic characteristics between groups.

There were no significant differences in demographic attributes and baseline measurements comparison between those who dropped out and those who completed the study. Therefore, there were no obvious biases attributable to attrition.

Quantitative outcomes

Acceptability

Post-session and Home Visit Feedback Form by caregivers

Six questions in the Post-session (PS) Feedback Form-Caregiver and three questions in Home Visit (HV) Caregiver Feedback Form were selected to assess the comprehensiveness and relevance, and agreement with their personal values, duration, and usefulness. The results of comprehensiveness and relevance, and agreement with their personal values were summarized in Table 2, and the results for duration and usefulness were summarized in Table 3.

Results from the four questions in the Post-session (PS) Feedback Form-Caregiver indicated that except for one of the participants' ratings for sessions 1B and 2 in EL, all participants

TABLE 1 Demographic characteristics of the participants.

	EL ($n = 9$)	IP ($n = 9$)	VC ($n = 7$)	WLC ($n = 9$)	Overall ($n = 34$)
Caregiver					
Age (yr)	38.22 ± 2.64	39.56 ± 4.48	37.86 ± 2.80	40.22 ± 3.8	39.03 ± 3.52
Gender (%)					
Male	0 (0)	3 (33)	1 (14)	0 (0)	4 (12)
Female	9 (100)	6 (67)	6 (86)	9 (100)	30 (88)
Education (%)					
Secondary	1 (11)	4 (44)	1 (14)	3 (33)	9 (26)
Post-secondary	1 (11)	1 (11)	0 (0)	2 (22)	4 (12)
Tertiary or above	7 (77)	4 (44)	6 (86)	4 (44)	21 (62)
Employment (%)					
Full-time	4 (44)	5 (56)	6 (86)	8 (89)	23 (68)
Part-time	1 (11)	2 (22)	1 (14)	0 (0)	4 (12)
Unemployed	4 (11)	2 (22)	0 (0)	1 (11)	7 (20)
Caregiver (%)					
As the primary	2 (22)	1 (11)	0 (0)	0 (0)	3 (9)
One of the 2 main	2 (22)	5 (56)	0 (0)	1 (11)	8 (23)
Many caregivers	5 (56)	3 (33)	5 (71)	8 (89)	21 (62)
Other (babysitters)	0 (0)	0 (0)	2 (29)	0 (0)	2 (6)
Child					
Age (yr)	5.33 ± 1	4.22 ± 1.3	3.71 ± 1.5	4.22 ± 1.48	4.41 +/− 1.40
Gender (%)					
Male	6 (67)	6 (67)	7 (100)	9 (100)	28 (82)
Female	3 (33)	3 (33)	0 (0)	0 (0)	6 (18)
With sibling(s) (%)	5 (56)	4 (44)	3 (43)	2 (22)	14 (41)
Sibling(s) with ASD/DD	4 (80)	1 (25)	0 (0)	0 (0)	5 (36)

TABLE 2 Post-session Feedback Form-Caregiver (Comprehensiveness & relevance, agreement with personal values).

	Comprehensiveness & relevance #(Rate of dissatisfaction)			Conflicts with personal values ##(Rate of confliction)		
	EL ($n = 9$)	IP ($n = 9$)	VC ($n = 7$)	EL ($n = 9$)	IP ($n = 9$)	VC ($n = 7$)
S1A	0 (0%)	0 (0%)	0 (0%)	0 (0%)	4 (22%)	0 (0%)
S1B	1 (3%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)
S2	1 (3%)	0 (0%)	0 (0%)	0 (0%)	2 (11%)	2 (14%)
S3	0 (0%)	0 (0%)	0 (0%)	0 (0%)	4 (22%)	0 (0%)
S4	0 (0%)	0 (0%)	0 (0%)	0 (0%)	6 (33%)	0 (0%)
S5	0 (0%)	0 (0%)	0 (0%)	1 (6%)	2 (11%)	2 (14%)
S6	0 (0%)	0 (0%)	0 (0%)	2 (11%)	4 (22%)	3 (21%)
S7	0 (0%)	0 (0%)	0 (0%)	0 (0%)	4 (22%)	2 (14%)
S8	0 (0%)	0 (0%)	0 (0%)	0 (0%)	4 (22%)	2 (14%)

Value < 3 "Disagree, Strongly disagree."
Value > 3 "Agree, Strongly agree."

agreed with the comprehensiveness and relevance of the session contents across all groups.

Regarding the perceived conflicts with contents and their beliefs, IP & VC groups found the contents of sessions conflicting with their personal values, whereas the majority of the EL participants found the contents of sessions neither conflicted with their own or their families' values.

TABLE 3 Post-session and Home Visit Feedback Form by Caregivers (Duration, usefulness of home visits and skill coaching).

Rate of dissatisfaction		S1A	S1B	S2	S3	S4	S5	S6	S7	S8	HV1	HV2	HV3
Duration	EL ($n = 9$)	11%	11%	11%	11%	11%	0%	0%	11%	0%	22%	11%	11%
	IP ($n = 9$)	33%	7%	22%	0%	11%	19%	22%	0%	0%	11%	0%	0%
	VC ($n = 7$)	38%	14%	10%	5%	10%	14%	19%	10%	0%	14%	0%	0%
***Home visits*	EL ($n = 9$)										22%	22%	11%
	IP ($n = 9$)	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	0%	0%	0%
	VC ($n = 7$)										0%	0%	0%
***Skill coaching*	EL ($n = 9$)										22%	22%	22%
	IP ($n = 9$)	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	11%	0%	0%
	VC ($n = 7$)	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	0%	0%	0%

*Too long / too short.
**Value < 3 "Not very useful, Completely useless."

TABLE 4 Facilitator's Feedback Form (Contents of the sessions).

	IP	VC
Caregivers' degree of recognition of the concept of the program	4.7	4.8
Caregivers' sense of engagement and participation	4.8	4.9
Sessions' contents for perceived relevance to caregivers	4.8	4.7
Caregivers' acceptance of contents.	4.7	4.8

Since there was no session provided to WLC participants, comments on the duration of each session were collected only for EL, IP, and VC groups only, while all groups gave ratings on that of each home visit. Despite a few participants in both groups finding the first session too long, most participants found the duration of other sessions and home visits appropriate.

Participants were asked to comment on the usefulness of home visits and skill coaching. Caregivers, except for one EL, found the first home visit not very useful, and none of the other participants in IP and VC groups found the home visits and skill coaching not useful. On the contrary, some caregivers in EL and WLC groups rated the home visits and skill coaching as "not very useful" or "completely useless."

Facilitator's Feedback Form

Table 4 summarizes the average ratings of the nine sessions by facilitators over IP and VC groups regarding the content of the sessions, the participant's level of engagement, acceptance, and involvement in planning home practices after each session. Only IP and VC groups were facilitated, so only these two groups of data were collected *via* this measure.

The average rating of the four questions in both groups was higher than 4 (out of five), suggesting that caregivers were perceived to have a great recognition of the concept of the programme and a high level of engagement and participation. As for the perceived relevance and acceptance, the results showed inconceivably high too.

Feasibility

Feasibility of CST conducted through different delivery modes was evaluated by participants' completion rate, attendance, adherence to home practice, observer's feedback on the involvement of the participants, and implementation of the group sessions.

Attendance and completion rate

Both IP and VC groups achieved a high attendance (95% for IP and 100% for VC), which was recorded by facilitators during each session. Coupling with high attendance, the completion rate of these two groups was also high (100% for IP and 78%

TABLE 5 Attendance of EL, IP & VC groups in each session and completion rate of all groups.

	Attendance										Completion rate		
	S1A	**S1B**	**S2**	**S3**	**S4**	**S5**	**S6**	**S7**	**S8**	**Overall (%)**	**Start**	**End**	**%**
EL ($n = 9$)	9	8	4	6	6	7	8	6	4	72%	14	9	64%
IP ($n = 9$)	9	8	9	8	9	8	8	9	9	95%	9	9	100%
VC ($n = 7$)	7	7	7	7	7	7	7	7	7	100%	9	7	78%
WLC	/	/	/	/	/	/	/	/	/	/	11	9	82%

TABLE 6 Comparison of practice frequency and duration.

		EL ($n = 9$)		IP ($n = 9$)		VC ($n = 7$)	
		Mean	**SD**	**Mean**	**SD**	**Mean**	**SD**
[a]Frequency (Times/wk)	T1	3.1	4.2	5.1	4.0	7.4	6.2
	T2	3.2	4.1	6.9	5.5	4.6	4.5
	Difference	*0.1 (3%)*	*0.8*	*1.8 (35%)*	*4.6*	*−2.8 (38%)*	*4.1*
[b]Duration (Minutes/wk)	T1	50.6	67.1	78.6	94.4	139.3	137.1
	T2	47.0	63.7	83.8	91.3	65.6	56.9
	Difference	*−3.6 (−7%)*	*23.3*	*5.2 (7%)*	*27.5*	*−73.7 (−53%)*	*96.6*

[a] Takes 0.5 time per week for reporting less than one time a week, takes 3.5 times per week for reporting 3-4 times a week, takes 5.5 times per week for reporting 5-6 times a week, takes 2 times a day, i.e., 14 times per week for reporting more than one time a day.

[b] Takes 22.5 minutes each time for reporting 15-30 min each time, takes 30 min each time for reporting more than 30 min each time.

for VC) which were shown in Table 5. On the contrary, the low attendance of EL group resulted in the lowest completion rate (64%), suggesting that the asynchronous mode favors participants with more self-discipline to learn regularly and complete the programme.

Adherence to home practice

Caregivers' adherence to home practice was evaluated by the feedback in the Caregiver Diary on the frequency and the daily time the participants practiced "Skills and Strategy" in daily activities at T1 & T2.

Table 6 shows the average practice frequency and duration per week for each group. The average practice frequency practiced with the children per week for EL, IP, and VC groups were 3.1 (SD = 4.2), 5.1 (SD = 4.0), and 7.4 (SD = 6.2) at T1, and were 3.2 (SD = 4.1), 6.9 (SD = 5.5), and 4.6 (SD = 4.5) at T2, respectively.

The average minutes practiced with the children per week for EL, IP, and VC groups were 50.6 (SD = 67.1), 78.6 (SD = 94.4), and 139.3 (SD = 137.1) at T1, and were 47 (SD = 63.7), 83.8 (SD = 91.3), and 65.6 (SD = 56.9) at T2, respectively.

The duration of practice time per week dropped for both EL and VC groups. Although the percentage dropped in both practice frequency and duration per week for the VC group was substantially large (-38% and −53% respectively), the change was not significant due to widespread use of the data. This dramatic drop in VC was mainly because two caregivers reduced the practices from twice a day (14 times per week) to 5–6 times a week. However, it was found that there was an increase in both practice frequency and duration per week in the IP group. Figures also show that both the practice frequency and duration were lower in EL than that in IP and VC groups.

Intervention fidelity and feasibility of group sessions delivery

Group sessions' intervention fidelity was reported by observers in rating the participants' degree of comfort, enthusiasm/interest, and level of involvement in planning home practice. Table 7 summarizes the average ratings of the nine sessions on the intervention fidelity of the group sessions. The average rating of the four components in both groups was >4 (out of five), suggesting that the intervention fidelity of group sessions was relatively high for both delivery modes.

Complexity, amount of content, and perceived preparation for each session were rated by the teaching facilitator to evaluate the implementation of the programme. Ratings of the complexity of the sessions' contents and concepts, as well as the number of sessions in both groups, were approximate 3 (out of five), indicating that facilitators found these two components of most sessions appropriate. Regarding the perceived readiness for the sessions, facilitators reported very high ratings at 4.8 (out of

TABLE 7 Average ratings on the intervention fidelity and feasibility of group sessions' delivery.

	Components	IP	VC
Intervention fidelity	Caregivers' degree of comfort	4.7	4.5
	Caregivers' enthusiasm/interest	4.6	4.6
	Caregivers' level of confidence	4.5	4.4
	Review of home practice	4.4	4.4
Feasibility	*Complexity of the sessions' contents and concepts	3.3	3.1
	Appropriateness of the amount of the sessions	3.4	3.3
	Facilitators' perceived preparedness for the sessions	4.8	4.8

*Value = 3 "Appropriate"; < 3 "A bit simple, too simple"; > 3 A bit complex, too complex.

five) as shown in Table 7, indicating that facilitators were always well-prepared for the sessions.

Quality of Life as parent outcome

One-way ANOVA test revealed that there was no significant difference between groups at T0, $F < 1$. A two-way ANOVA was conducted with a within-participant factor of time points (2 levels: T0, T2) and a between-participant factor of condition (4 levels: EL, IP, VC, WLC). The main effects of both time points and condition were not significant, $F_{(1,30)} = 2.55$, $p = 0.12$ and $F < 1$. The interaction between time points and conditions was also not significant, $F < 1$.

Descriptive analyses showed that there was an overall improvement in EL, IP and VC groups (3, 13, and 16%, respectively), while WLC slightly worsened (−3%) from T0 to T2. The overall and individual item scores of the GHQ-12 in each group at T0, T2, and the differences between T0 and T2 are shown in Table 8.

At T2, there were 7 items improved in IP (with Q11 on self-worth yielding a significant change at $p < 0.05$), 6 items in both EL and VC groups, and 3 items in WLC. Whilst, one item worsened in IP, 2 in VC, 3 in EL, and 5 in WLC. Improvement in the IP condition was strongly evidenced as 6 out of these 7 items were with rating below one, meaning IP participants nearly always have positive feelings about those items. One item related to negative feelings, meaning they rarely or do not feel that way at all.

Strengths and Difficulties Questionnaire as child outcome

A two-way ANOVA was conducted for *SDQ total* (Table 8) with a within-participant factor of time points (2 levels: T0, T2) and a between-participant factor of condition (4 levels: EL, IP, VC, WLC), the main effect of time points was significant, with $F_{(1,30)} = 5.55$, $p < 0.05$, partial eta squared = 0.16. The main effect of conditions was also significant, $F_{(3,30)} = 3.21$, $p < 0.05$, partial eta squared = 0.24. The interaction between time points and condition was not significant, $F < 1$. Further investigation showed that the SDQ total of IP was significantly lower at T2 than at T0 ($p < 0.05$) and VC was significantly lower than that of EL at T2 ($p < 0.05$).

A two-way ANOVA was conducted for prosocial with a within-participant factor of time points (2 levels: T0, T2) and a between-participant factor of condition (4 levels: EL, IP, VC, WLC), the main effects of both time points [$F_{(1,30)} = 3.65$, $p = 0.07$, partial eta squared = 0.11] and condition [$F_{(3,30)} = 1.54$, $p = 0.23$, partial eta squared = 0.13] were not significant. The interaction between time points and condition was also not significant, $F < 1$.

The average scores of the Difficulties-total for EL, IP, VC, and WLC were 21.33 (SD = 5.39), 21.56 (SD = 4.64), 15.72 (SD = 3.64), and 18.00 (SD = 5.48) at T0, and were 20.00 (SD = 5.61), 18.78 (SD = 4.41), 13.29 (SD = 2.56), and 18.00 (SD = 5.05) at T2, respectively.

The average scores of the Prosocial Scale for EL, IP, VC, and WLC were 3.78 (SD = 2.11), 2.44 (SD = 2.65), 2.00 (SD = 2.16), and 4.56 (SD = 2.60) at T0, and were 4.00 (SD = 1.94), 3.33 (SD = 3.16), 2.71 (SD = 1.25), and 4.67 (SD = 2.83) at T2, respectively.

Descriptive analyses showed that there was an overall improvement (decrease in problematic behaviors and increase in prosocial behaviors) in all experimental groups, while the total difficulties score of the WLC group remained unchanged. In particular, the two experimental groups with facilitators (IP and VC) gained greater improvements (with 13% and 15% decrease in Difficulties-total, and 36.5% and 35.5% increase in Prosocial Scale for IP and VC groups, respectively) than the group without a facilitator (EL group), with only 6% decrease in Difficulties-total and 5.9% increase in Prosocial Scale. This suggests that the presence of facilitators can be a factor in the decrease in problematic behaviors and increase in prosocial behaviors among children.

Qualitative outcomes

Open-ended feedback on each session and each home visit that was received across the groups underwent pattern coding and major themes were established. Three underlying themes including "Delivery Format," "Programme Materials" and "Interaction" were identified and further broken down into various sub-themes under the domain of acceptability and feasibility with both positive and negative valence. Table 9 shows the descriptions of the comments reported vastly in the four groups, in which heavyweight subthemes are described

TABLE 8 GHQ-12 item and SDQ scores at T0 and T2.

		EL ($n = 9$)	IP ($n = 9$)	VC ($n = 7$)	WLC ($n = 9$)
[a]GHQ-12	T0	17.33	15.56	13.99	13.45
	T2	16.77	13.57	11.71	13.90
	Diff	*−0.56 (−3%)*	*−1.99 (−13%)*	*−2.28 (−16%)*	*0.45 (3%)*
[a]SDQ- Difficulties-total	T0	21.33	21.56	15.72	18.00
	T2	20.00	18.78	13.29	18.00
	Diff	*−1.33 (−6%)*	*−2.78 (−13%)*	*−2.43[#] (−15%)*	*0 (0%)*
[b]SDQ- Prosocial	T0	3.78	2.44	2.00	4.56
	T2	4.00	3.33	2.71	4.67
	Diff	*0.22 (5.9%)*	*0.89[#] (36.5%)*	*0.71 (35.5%)*	*0.11 (2.4%)*

[a] Differences between T2 & T0: -ve - improved, +ve – worsened.
[b] Differences between T2 & T0: +ve - improved, -ve – worsened.
*$p < 0.05$; [#] Marginal $0.05 < p < 0.1$.

qualitatively hereafter and noteworthy subthemes are evidenced with group identifiers. As the video recording arrangement is a standalone question in the Post Home Visit Feedback Form, the finding is separately described in a distinct section below.

Findings show that there was a high level of acceptability and feasibility of the adapted programme in the aspects of delivery formats and programme materials. As for interaction, it was the most popular theme among IP and VC participants while it became the deficient factor to be accepted and implemented in EL and WLC groups where interactions were limited.

Delivery formats

Perceived convenience and increased privacy raised the acceptability, and effective use of multimedia boosted the feasibility of the adapted delivery formats. Convenience was viewed differently in both group sessions and home visits by different groups. EL enjoyed the time and location flexibility, as well as the room to learn at their own pace. Some revealed that the self-learning modes could fit the busy working parents while privacy could be preserved.

> EL11: "The self-learning sessions were comprehensive and suited busy working mothers."

Group sessions were considered to be convenient among IP & VC as they strictly followed the social distancing measures where in-person facilitation was not allowed. Besides, IP participants also appreciated the privacy of one-on-one home visits too.

> IP01: "The advantage of doing it via recording is that it is less intrusive. The family and the child may not notice strangers around and therefore help create a more home-like environment."

Nevertheless, poor time management and a large number of participants in the groups lowered the feasibility of IP and VC. Unstructured learning decreased the feasibility of EL. Whilst barriers to taking videos and length of videos were the negative factors in implementing the programme across all groups.

Programme materials

Comprehensive session content and efficacious skill and strategies were the acceptable and feasible elements of the programme irrespective of the intervention modes.

> EL12: "This session (session 2 on joint engagement) changed my mindset. It made me understand that I was too instructive while playing with my kid, and from now on, I have to prepare two sets of toys to imitate his way of playing."

However, some responses showed that there were insufficient contents or examples in the materials, for sessions 5, 6, and 7 (behavioral management and teaching skills). This can be improved accordingly to increase the feasibility of the programme materials.

> EL05: "How to deal with self-harming behaviors?"
>
> VC11: "This topic is very complicated, and I recommend having in-depth discussions, for example, through the case study."

Interactions

Interactions of any kind were deemed as an important factor in acceptability and feasibility across. Subthemes identified were supportive peers' sharing, interactive professional facilitation on skills application with more examples, interactive professional coaching during home visits for the acceptability domain, and

TABLE 9 Themes derived from comments of each Post-session & Home Visit.

	Theme	*Valence*	*Subthemes grouped over feedbacks on sessions and home visits*	EL	IP	VC
Acceptability	***Delivery format***	+ve	Perceived convenience (PS/HV)	✓	✓	✓
			Elevated privacy (PS/HV)	✓	✓	✓
			Support to video-recording (HV)	✓	✓	✓
		-ve	Barriers to video-recordings (HV)	✓	✓	✓
	Programme materials	+ve	Comprehensible content (PS)	✓	✓	✓
	Interaction	+ve	Supportive peers' sharing (PS)		✓	✓
			Interactive professional facilitation on skill application and examples (PS)	✓	✓	✓
			Instant professional coaching at home visits (HV)	✓	✓	✓
		-ve	No interactions with peers (PS)	✓	✓	✓
			Limited professional guidance or feedback (PS/HV)	✓	✓	✓
Feasibility	***Delivery format***	+ve	Effective use of multimedia materials (PS)	✓	✓	✓
		-ve	Time control (PS)	✓	✓	✓
			Large number of participants (PS)			✓
			Challenges to complete the sessions without a fixed time session schedule (PS/HV)	✓	✓	✓
			Videos taken being too lengthy (HV)	✓	✓	✓
	Programme materials	+ve	Effective skills & strategies (PS/HV)	✓	✓	✓
		-ve	Insufficient content or illustration of examples in some of the sessions (PS)	✓	✓	✓
	Interaction	+ve	Face-to-face skill practices (PS/HV)		✓	
			In-person demonstration with children (HV)	✓	✓	✓
		-ve	Lack of in-person skill practices/in-person home visits (PS/HV)	✓	✓	✓

face-to-face skill practices and in-person skill demonstrations with children for the feasibility domain. All IP participants reported that they enjoyed and appreciated much with all the listed delivery elements at group sessions and home visits, and thus they showed a high level of acceptability and feasibility. Some of them even gave "perfect" in their comment. Responses revealed there were perceived positive changes in better understanding of children's communication, better handling of children's behaviors, improvement in parent-child relationship, and harmony in the family which led to better family functioning.

> IP09: "I learnt at the skill coaching section from [CST interventionist] and the facilitators which were practical and applicable to deal with SEN children. I could better understand my child and better handle her behaviors. There are fewer conflicts in the family and my hubby appreciated a lot of the changes. We no longer argued as much as before and it's magical!"

As for VC, they also had a high level of acceptability as they were offered the interactive elements as IP. They appreciated the professional facilitation at group sessions and the instant professional coaching at home visits online. Yet, they would suggest the addition of face-to-face skill practices and in-person skill demonstrations with children for VC delivery modes. Meanwhile, the majority of EL participants complained about lacking professional guidance or feedback and interactions with peers in addition to face-to-face skill practices and in-person skill demonstrations with children. They were frustrated as they perceived no improvements in their parenting skills. Also, they found the home visits not helpful as no instant feedback was provided. A few participants desperately requested interaction as:

> EL07: "I am so lonely and cannot improve my skills without interactions and feedback."

Arrangement of video recordings at the home visits

All participants in three interventions accepted video recording as a necessary part of the first home visit. This acceptability remained across the rest of the two home visits in IP and VC while the acceptability in EL had been dropping since the second home visit. Participants reported that they understood the arrangement as essential and as an alternative under the pandemic situation while a majority understood and expected that the videos taken would receive professional feedback on their practice, serve as a self-learning opportunity to improve their skills and be a part of sustaining the programme in the long run.

> EL09: "Both my child and I enjoyed the video-recording very much."
>
> IP11: "The videos can be a reference and review the child's development in the future."
>
> VC09: "Routines are important in training the children with SEN. Video taking is crucial in assisting the assessment of their abilities and in developing them."

Despite the acceptability, some of them complained about the "barriers to video-recordings," which included the distractions from other family members at home and technical problems of using electronic equipment to capture the videos and uploading the videos to the platform.

Discussions

This study sought to investigate the acceptability and feasibility of various implementation methods of CST among families with children with developmental disabilities in Hong Kong under lockdown.

High quantitative ratings for comprehensiveness and relevance, in line with their personal values, duration, and usefulness at post-session and post home visit among the participants indicate high acceptance and satisfaction with CST across various delivery modes.

As per the qualitative findings, the IP delivery mode received the highest level of acceptability and feasibility, and VC came second with the satisfied comments received in general. Then, EL followed VC, and WLC showed the lowest acceptance and feasibility. Items with negative valence under IP and VC groups are controllable factors, such as better time management and smaller group size. Furthermore, the suggestions of additional content and examples in behavior management, echoed the caregivers' needs in an Italian study (34), and provided more alternatives to mitigate challenging behaviors such as self-harming behaviors are suggested.

Comments from EL also provided insights and suggestions for delivering the programme with improved acceptability and feasibility. This is aligned with the key elements of interactions - two-way communication - which can be online or in-person, and with facilitators or with peers. Caregivers of children with developmental delays or disabilities in Hong Kong are special caregivers with out-of-proportional parenting stress (16–18). Although they enroll in the programme to acquire knowledge on parenting, they also wish for more guidance, support, encouragement, and recognition. EL caregivers in the study reported being frustrated by the lack of feedback and experiencing loneliness that was not mitigated by the intervention. Besides, World Health Organization's Caregiver Skills Training Programme (WHO CST) is a skill transferring programme that skill practices with interactive feedback would be more beneficial for the skill-attaining process. In addition, the demand for interactive activities increased with the number of sessions and home visits delivered and the demands were more desperate with time.

In view of EL feasibility, the more flexible self-learning mode appears more suitable for caregivers who are less busy and have better time management skills. Caregivers reported that they were not able to complete the sessions within a week because of work commitments, which is reflected in their attendance. Regular weekly reminders were given to those who had not completed the previous session.

With the clinical outcomes, IP and VC yielded greater improvements in SDQ and GHQ-12 than EL, including the well-being of caregivers, perceived children's difficulties, and prosocial behaviors, which are triangulated with the acceptability and feasibility levels of delivery modes. Results of SDQ show an overall improvement (decrease in problematic behaviors and increase in prosocial behaviors) in all experimental groups, while the total difficulties score of the WLC group remained unchanged. In particular, the two experimental groups with facilitators (IP and VC) gained greater improvements (with 13% and 15% decrease in Difficulties-total, and 36.5% and 35.5% increase in Prosocial Scale for IP and VC groups, respectively) than the group without a facilitator (EL group), with only 6% decrease in Difficulties-total and 5.8% increase in Prosocial Scale. This suggests that the presence of facilitators can be a factor in the development of caregiver skills for decreasing challenging behaviors and increasing prosocial behaviors.

From GHQ-12 results, all groups involved in CST showed positive results, while the group (EL) without a facilitator showed improvement, but to a lesser extent. A decreased GHQ-12 score in the control group participants may be due to the challenges and frustrations in engaging their children during the video recording. These results also echoed qualitative findings that the programme helped improve caregivers' well-being, child's communication and behaviors, and thus better family functioning. Concerning the value conflict, the quantitative result was mixed. Thereafter, further analysis revealed that individual participants showing a conflict in personal values did have positive changes in the GHQ-12 results. This suggests that conflicting information does not necessarily obstruct the caregivers from learning new concepts. The intervention may prompt caregivers to revisit their prior beliefs that result in positive improvements in their mental health and quality of life.

In other words, participants perceived both in-person and videoconferencing modalities favor skill attainment among parents strengthening the communication and behavioral management with their children. These were supported by the results of GHQ and SDQ that VC and IP promoted caregivers' well-being and child's adaptive behaviors and reduced challenging behaviors among the children. All groups, except the wait-list control group, showed some positive changes in SDQ, which suggest the video recordings as data collection may increase their awareness of the interaction with their child, resulting in certain improvements. Overall, only IP gained significant improvements while other groups did not show any advantage. This indicates the in-person live practice sessions, and skill coaching during the home visits played a role in the improvement of children's behaviors.

Limitations

There were several limitations to this study. First, the initial dropout rate (51%) resulted in a dramatic decrease in sample size from 70 to 34 due to the sudden pandemic spike. Some participants became more concerned about social distancing in face-to-face mode while others who expected in-person sessions dropped out when they were assigned to non-interacting modes. This reduced the power to detect differences between the groups, in particular, IP and VC, but not EL groups. A more accurate understanding of the effect of the experimental conditions could only be obtained when the study is conducted under normal circumstances. The current results however would be informative to the understanding of the experimental effect under a pandemic situation with disrupted social interaction. Second, the outbreak of COVID-19 disrupted the original research for the IP group, leading to swapping the face-to-face sessions to online sessions, like VC, which lowered the difference between the two groups. It might have affected the research outcomes. Lastly, although the results promote confidence in further expansion of implementation, the study has less experimental control over confounding factors, which may affect the persuasiveness of the results.

All in all, this study showed supportive evidence of the acceptability and feasibility of the World Health Organization's Caregiver Skills Training Programme (WHO CST) conducted by different delivery modes, including in-person, virtual, and self-learning, among caregivers of children with developmental disabilities. This study only reported two measures related to caregivers' quality of life and children's difficulties and adaptive behaviors, so future studies are needed to examine other related variables, such as caregivers' coping skills, joint engagement, and children's behaviors in different domains with larger sample size. To address comments from caregivers, future research on effectiveness can be carried out with further adaptations to the current delivery modes with more interactive elements incorporated.

Conclusions

Current findings indicate that three adapted CST delivery modes were found highly acceptable and feasible. Participants indicated that the programme was comprehensive, reliable, and useful despite the different ways the programme was delivered. To conclude, the programme was determined to have high acceptability and feasibility, regardless of the mode of delivery. When comparing the degree of effectiveness, all three modes of delivery yielded positive impacts on caregivers and children. In-person delivery mode was found to have the greatest improvement in mitigating perceived child's challenging behaviors, whereas the interactive modes, IP and VC, showed the larger positive change in improving caregivers' well-being, which was followed by EL mode. In-person skill practices can significantly improve children's behaviors whereas personal support, either face-to-face or virtual, appears to be key in maintaining caregivers' mental well-being. In addition, caregivers highlighted that having facilitators' face-to-face coaching and interactive peer support contributed to their persistence and momentum to complete the programme. The findings can be linked to how the programme nature can be highlighted to satisfy the objectives of the programme.

On top of this, the heterogeneous findings suggest that it is difficult to conclude which service delivery mode, eLearning, videoconferencing, or in-person, is better. Self-directed programmes usually provide a convenient means for users to learn at their own pace, without being restricted by time, location, or other tangible conditions (82). It appears to be most suited to caregivers who have strong independent learning and self-management skills. However, for some intervention programmes based on a more complex theoretical design, self-learning alone may not give desirable outcomes. On the other hand, programmes requiring a high degree of interaction to demonstrate ways to teach new skills and manage behaviors, or those interventions requiring meticulous observations of subtle expressions or body language, and immediate reciprocity in psychotherapy sessions may not have a good fit with the existing technological level. Under such circumstances, the traditional face-to-face mode still has advantages (83). Furthermore, service receivers are different populations, so the acceptance and capability of technology applications can vary considerably (84), which goes against a "one-size-fits-all" approach. Offering caregiver-mediated interventions in a variety of delivery modes may best meet the needs of a diverse group of caregivers and maximize the reduction in the treatment gap for children with developmental disabilities and delays and their families. Therefore, future research should be focused on the advantages of each service delivery mode and how they can fit into the existing community settings and fit the target beneficiaries. It is also necessary to investigate the factors that affect the efficacy of the intervention, so as to exclude the possibility of erroneously attributing the results of the intervention to the service delivery modes.

Different CST modes of delivery were found acceptable and feasible whereas the statistical GHQ-12 and SDQ findings gave different degrees of positive changes among the delivery modes. In this study, the in-person mode received the highest acceptability and feasibility, and the most desirable clinical outcomes. Findings also can be interpreted that the degree of interaction was a positive factor in its effectiveness. Various interactive facilitation components can be considered and incorporated to fit best into the needs of the caregivers as well as the practitioners. This serves as a direction for how World Health Organization's Caregiver Skills Training Programme (WHO CST) can be implemented in Hong Kong, or a reference to other countries implementing the programme in their community, when balancing the effectiveness and constraints in different settings.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving human participants were reviewed and approved by the Human Research Ethics Committee of Hong Kong University (EA200178). All data collection was performed in accordance with HKU guidelines and regulations. The patients/participants provided their written informed consent to participate in this study.

WHO CST team

Chiara Servili[1], Erica Salomone[1,2], Laura Pacione[1,3], Stephanie Shire[4] and Felicity L. Brown[5,6]

[1]Department of Mental Health and Substance Use, World Health Organization, Geneva, Switzerland
[2]The University of Milano-Bicocca, Milan, Italy
[3]Department of Psychiatry, Division of Child and Youth Mental Health, University of Toronto, Toronto, ON, Canada
[4]The University of Oregon, Eugene, OR, United States
[5]Research and Development Department, War Child Holland, Amsterdam, Netherlands
[6]Amsterdam Institute of Social Science Research, University of Amsterdam, Amsterdam, Netherlands.

Author contributions

Material preparation, data collection and analysis were performed by JL, SL, and FI. The first draft of the manuscript was written by JL and FI. All authors contributed to the study conception and design, commented on previous versions of the manuscript, and read and approved the final manuscript.

Acknowledgments

This work was the piece of work in collaboration with the Master of Social Sciences (Counseling) Programme at The University of Hong Kong. We would like to thank the participants of the study and the students for their facilitation. We sincerely wish to thank the WHO CST research team for guiding us throughout the research process. We also want to thank CS, ES, LP, SS, Dr. Andy Shih and Dr. Pamela Dixon, for their never falling support and kind hearts to walk the baby steps with us throughout the years.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyt.2022.915263/full#supplementary-material

References

1. Faber J, King J. *How to Talk so Little Kids Will Listen*. New York, NY: Scribner Book Company. (2017).

2. Jones K. *Early Childhood Brain Development Has Lifelong Impact*. Arizona, AZ: Arizona PBS. (2017).

3. Chudler, E. *Neuroscience For Kids - Brain Development*. (2015). Available online at: http://faculty.washington.edu/chudler/dev.html (accessed June 8, 2021).

4. Gale CR, O'Callaghan FJ, Godfrey KM, Law CM, Martyn CN. Critical periods of brain growth and cognitive function in children. *Brain*. (2004) 127:321–9. doi: 10.1093/brain/awh034

5. Wong PW, Lam Y, Lau JS, Fok H. The resilience of social service providers and families of children with autism or development delays during the COVID-19 pandemic-a community case study in Hong Kong. *Front Psychiatry*. (2020) 11:561657. doi: 10.3389/fpsyt.2020.561657

6. Brown SM, Doom JR, Lechuga-Peña S, Watamura SE, Koppels T. Stress and parenting during the global COVID-19 pandemic. *Child Abuse Negl*. (2020) 110:104699. doi: 10.1016/j.chiabu.2020.104699

7. Lawson M, Piel MH, Simon M. Child maltreatment during the COVID-19 pandemic: consequences of parental job loss on psychological and physical abuse towards children. *Child Abuse Negl*. (2020) 110:104709. doi: 10.1016/j.chiabu.2020.104709

8. Unicef.org. *COVID-19: Protecting Children From Violence, Abuse and Neglect in the Home*. (2020). Available online at: https://www.unicef.org/media/68711/file/COVID-19-Protecting-children-from-violence-abuse-and-neglect-in-home-2020.pdf (accessed June 8, 2021).

9. World Health Organization. *mhGAP Intervention Guide for Mental, Neurological and Substance Use Disorders in Non-Specialized Health Settings*. (2020). Available online at: http://apps.who.int/iris/bitstream/handle/10665/250239/9789241549790-eng.pdf?sequence=1 (accessed June 8, 2021).

10. Zeidan J, Fombonne E, Scorah J, Ibrahim A, Durkin MS, Saxena S, et al. Global prevalence of autism: a systematic review update. *Autism Res*. (2022) 15:778–90. doi: 10.1002/aur.2696

11. CDC. *Community Report on Autism 2021*. CDC (2021).

12. Census and Statistics Department. *Social Data Collected Via the General Household Survey - Special Topics Report No. 63: Persons With Disabilities and Chronic Diseases*. HKSAR (2021).

13. Food and Health Bureau. *Mental Health Review Report*. HKSAR (2017).

14. Hong Kong Special Administrative Region. *LCQ18: Support for Children With Special Needs*. GovHK (2020).

15. Tsang E. *Charity Offers Low-Income Hong Kong Children Mental Health Care as City Waiting Times Hit 3 Years*. Hong Kong: South China Morning Post. (2017).

16. Montes G, Halterman JS. Psychological functioning and coping among mothers of children with autism: a population-based study. *Pediatrics*. (2007) 119:e1040–6. doi: 10.1542/peds.2006-2819

17. Johnson CP, Myers SM, American Academy of Pediatrics Council on Children With Disabilities. Identification and evaluation of children with autism spectrum disorders. *Pediatrics*. (2007) 120:1183–215. doi: 10.1542/peds.2007-2361

18. Crnic KA, Greenberg MT. Minor parenting stresses with young children. *Child Dev*. (1990) 61:1628. doi: 10.2307/1130770

19. Statista. *Average Living Space of PRH Tenants Hong Kong 2007–2021*. (2021). Available online at: https://www.statista.com/statistics/630746/hong-kong-public-rental-housing-average-living-space-per-person/ (accessed December 20, 2021).

20. Kasari C, Lawton K, Shih W, Barker TV, Landa R, Lord C, et al. Caregiver-mediated intervention for low-resourced preschoolers with autism: an RCT. *Pediatrics*. (2014) 134:e72–9. doi: 10.1542/peds.2013-3229

21. Oono IP, Honey EJ, McConachie H. Parent-mediated early intervention for young children with autism spectrum disorders (ASD). *Evid Based Child Health*. (2013) 8:2380–479. doi: 10.1002/ebch.1952

22. Reichow B, Servili C, Yasamy MT, Barbui C, Saxena S. Non-specialist psychosocial interventions for children and adolescents with intellectual disability or lower-functioning autism spectrum disorders: a systematic review. *PLoS Med*. (2013) 10:e1001572. doi: 10.1371/journal.pmed.1001572

23. Ferraioli SJ, Harris SL. Comparative effects of mindfulness and skills-based parent training programs for parents of children with autism: feasibility and preliminary outcome data. *Mindfulness (NY)*. (2013) 4:89–101. doi: 10.1007/s12671-012-0099-0

24. Koegel LK, Koegel RL, Ashbaugh K, Bradshaw J. The importance of early identification and intervention for children with or at risk for autism spectrum disorders. *Int J Speech Lang Pathol*. (2014) 16:50–6. doi: 10.3109/17549507.2013.861511

25. Leung C, Fan A, Sanders MR. The effectiveness of a Group Triple P with Chinese parents who have a child with developmental disabilities: a randomized controlled trial. *Res Dev Disabil*. (2013) 34:976–84. doi: 10.1016/j.ridd.2012.11.023

26. World Health Organization. *mhGAP : Mental Health Gap Action Programme : Scaling Up Care for Mental, Neurological and Substance Use Disorders*. Geneva: World Health Organization (2008).

27. WHA. *Comprehensive and Coordinated Efforts for the Management of Autism Spectrum Disorders*. (2014). Available online at: https://www.who.int/mental_health/maternal-child/WHA67.8_resolution_autism.pdf (accessed June 8, 2021).

28. Reichow B, Kogan C, Barbui C, Smith I, Yasamy MT, Servili C. Parent skills training for parents of children or adults with developmental disorders: systematic review and meta-analysis protocol. *BMJ Open*. (2014) 4:e005799. doi: 10.1136/bmjopen-2014-005799

29. World Health Organization. *Ninth Meeting of the Mental Health Gap Action Programme (mhGAP): mhGAP Forum "Mental Health Capacity Building Within Countries"*. (2017). Available online at: http://www.who.int/mental_health/mhgap/forum_report_2017/en/ (accessed June 8, 2021).

30. Salomone E, Pacione L, Shire S, Brown FL, Reichow B, Servili C. Development of the WHO caregiver skills training program for developmental disorders or delays. *Front Psychiatry*. (2019) 10:769. doi: 10.3389/fpsyt.2019.00769

31. World Health Organization. *Mental Health - Training Parents To Transform Children's Lives*. (2018). Available online at: http://www.who.int/mental_health/maternal-child/PST/en/ (accessed June 8, 2021).

32. Tekola B, Girma F, Kinfe M, Abdurahman R, Tesfaye M, Yenus Z, et al. Adapting and pre-testing the world health organization's caregiver skills training programme for autism and other developmental disorders in a very low-resource setting: findings from Ethiopia. *Autism*. (2020) 24:51–63. doi: 10.1177/1362361319848532

33. Salomone E, Ferrante C, Salandin A, Ferrara F, Torchio E, Foletti G, et al. Acceptability and feasibility of the world health organization's caregiver skills training implemented in the Italian national health system. *Autism*. (2021) 26:859–74. doi: 10.1177/13623613211035228

34. Salomone E, Settanni M, McConachie H, Suma K, Ferrara F, Foletti G, et al. Pilot randomized controlled trial of the WHO caregiver skills training in public health services in Italy. *J Autism Dev Disord*. (2021). doi: 10.1007/s10803-021-05297-x

35. Sengupta K, Shah H, Ghosh S, Sanghvi D, Mahadik S, Dani A, et al. World health organisation-Caregiver Skills Training (CST) program: feasibility of delivery by non-specialist providers in real-world urban settings in India. *J Autism Dev Disord*. (2021 D) 1:1–8. doi: 10.1007/s10803-021-05367-0

36. Backhaus A, Agha Z, Maglione ML, Repp A, Ross B, Zuest D, et al. Videoconferencing psychotherapy: a systematic review. *Psychol Serv*. (2012) 9:111–31. doi: 10.1037/a0027924

37. Stahmer AC, Gist K. The effects of an accelerated parent education program on technique mastery and child outcome. *J Posit Behav Interv*. (2001) 3:75–82. doi: 10.1177/109830070100300203

38. Symon JB. Parent education for autism: issues in providing services at a distance. *J Posit Behav Interv*. (2001) 3:160–74. doi: 10.1177/109830070100300304

39. Chen CK, Nehrig N, Wash L, Schneider JA, Ashkenazi S, Cairo E, et al. When distance brings us closer: leveraging tele-psychotherapy to build deeper connection. *Couns Psychol Q*. (2021) 34:554–67. doi: 10.1080/09515070.2020.1779031

40. Battistin T, Mercuriali E, Zanardo V, Gregori D, Lorenzoni G, Nasato L, et al. Distance support and online intervention to blind and visually impaired children during the pandemic COVID-19. *Res Dev Disabil*. (2021) 108:103816. doi: 10.1016/j.ridd.2020.103816

41. Geller S. Cultivating online therapeutic presence: strengthening therapeutic relationships in teletherapy sessions. *Couns Psychol Q*. (2021) 34:687–703. doi: 10.1080/09515070.2020.1787348

42. Inchausti F, MacBeth A, Hasson-Ohayon I, Dimaggio G. Telepsychotherapy in the age of COVID-19: a commentary. *J Psychother Integr*. (2020) 30:394–405. doi: 10.1037/int0000222

43. Watts S, Marchand A, Bouchard S, Gosselin P, Langlois F, Belleville G, et al. Telepsychotherapy for generalized anxiety disorder: Impact on the working alliance. *J Psychother Integr.* (2020) 30:208–25. doi: 10.1037/int0000223

44. Langarizadeh M, Tabatabaei M, Tavakol K, Naghipour M, Moghbeli F. Telemental health care, an effective alternative to conventional mental care: a systematic review. *Acta Inform Med.* (2017) 25:240. doi: 10.5455/aim.2017.25.240-246

45. Ashburner J, Vickerstaff S, Beetge J, Copley J. Remote versus face-to-face delivery of early intervention programs for children with autism spectrum disorders: perceptions of rural families and service providers. *Res Autism Spectr Disord.* (2016) 23:1–14. doi: 10.1016/j.rasd.2015.11.011

46. Etzelmueller A, Radkovsky A, Hannig W, Berking M, Ebert DD. Patient's experience with blended video- and internet based cognitive behavioural therapy service in routine care. *Internet Interv.* (2018) 12:165–75. doi: 10.1016/j.invent.2018.01.003

47. Nelson E-L, Barnard M, Cain S. Treating childhood depression over videoconferencing. *Telemed J E Health.* (2003) 9:49–55. doi: 10.1089/153056203763317648

48. Geller SM. Therapeutic presence and polyvagal theory: Principles and practices for cultivating effective therapeutic relationships. In: *Clinical Applications of the Polyvagal Theory*. New York, NY: Norton Professional Books (2018). p. 106–26.

49. Lodder A, Papadopoulos C, Randhawa G. Using a blended format (videoconference and face to face) to deliver a group psychosocial intervention to parents of autistic children. *Internet Interv.* (2020) 21:100336. doi: 10.1016/j.invent.2020.100336

50. Gros DF, Morland LA, Greene CJ, Acierno R, Strachan M, Egede LE, et al. Delivery of evidence-based psychotherapy *via* video telehealth. *J Psychopathol Behav Assess.* (2013) 35:506–21. doi: 10.1007/s10862-013-9363-4

51. Sinclair PM, Kable A, Levett-Jones T, Booth D. The effectiveness of Internet-based e-learning on clinician behaviour and patient outcomes: a systematic review. *Int J Nurs Stud.* (2016) 57:70–81. doi: 10.1016/j.ijnurstu.2016.01.011

52. Florean IS, Dobrean A, Păsărelu CR, Georgescu RD, Milea I. The efficacy of internet-based parenting programs for children and adolescents with behavior problems: a meta-analysis of randomized clinical trials. *Clin Child Fam Psychol Rev.* (2020) 23:510–28. doi: 10.1007/s10567-020-00326-0

53. Pennefather J, Hieneman M, Raulston TJ, Caraway N. Evaluation of an online training program to improve family routines, parental well-being, and the behavior of children with autism. *Res Autism Spectr Disord.* (2018) 54:21–6. doi: 10.1016/j.rasd.2018.06.006

54. Catarino A, Bateup S, Tablan V, Innes K, Freer S, Richards A, et al. Demographic and clinical predictors of response to internet-enabled cognitive-behavioural therapy for depression and anxiety. *BJPsych Open.* (2018) 4:411–8. doi: 10.1192/bjo.2018.57

55. Fisher WW, Luczynski KC, Blowers AP, Vosters ME, Pisman MD, Craig AR, et al. A randomized clinical trial of a virtual-training program for teaching applied-behavior-analysis skills to parents of children with autism spectrum disorder. *J Appl Behav Anal.* (2020) 53:1856–75. doi: 10.1002/jaba.778

56. Nefdt N, Koegel R, Singer G, Gerber M. The use of a self-directed learning program to provide introductory training in pivotal response treatment to parents of children with autism. *J Posit Behav Interv.* (2010) 12:23–32. doi: 10.1177/1098300709334796

57. Wainer AL, Ingersoll BR. Disseminating ASD interventions: a pilot study of a distance learning program for parents and professionals. *J Autism Dev Disord.* (2013) 43:11–24. doi: 10.1007/s10803-012-1538-4

58. Wainer AL, Ingersoll BR. Increasing access to an ASD imitation intervention via a telehealth parent training program. *J Autism Dev Disord.* (2015) 45:3877–90. doi: 10.1007/s10803-014-2186-7

59. Parsons D, Vaz S, Lee H, Robinson C, Cordier R. A twelve-month follow-up of an information communication technology delivered intervention for children with autism spectrum disorder living in regional Australia. *Res Dev Disabil.* (2020) 106:103743. doi: 10.1016/j.ridd.2020.103743

60. Hinton S, Sheffield J, Sanders MR, Sofronoff K. A randomized controlled trial of a telehealth parenting intervention: a mixed-disability trial. *Res Dev Disabil.* (2017) 65:74–85. doi: 10.1016/j.ridd.2017.04.005

61. Baharav E, Reiser C. Using telepractice in parent training in early autism. *Telemed J E Health.* (2010) 16:727–31. doi: 10.1089/tmj.2010.0029

62. Suess AN, Romani PW, Wacker DP, Dyson SM, Kuhle JL, Lee JF, et al. Evaluating the treatment fidelity of parents who conduct in-home functional communication training with coaching via telehealth. *J Behav Educ.* (2014) 23:34–59. doi: 10.1007/s10864-013-9183-3

63. Vismara LA, Young GS, Rogers SJ. Telehealth for expanding the reach of early autism training to parents. *Autism Res Treat.* (2012) 2012:121878. doi: 10.1155/2012/121878

64. Vismara LA, McCormick C, Young GS, Nadhan A, Monlux K. Preliminary findings of a telehealth approach to parent training in autism. *J Autism Dev Disord.* (2013) 43:2953–69. doi: 10.1007/s10803-013-1841-8

65. Ingersoll B, Berger NI. Parent engagement with a telehealth-based parent-mediated intervention program for children with autism spectrum disorders: predictors of program use and parent outcomes. *J Med Internet Res.* (2015) 17:e227. doi: 10.2196/jmir.4913

66. Ferguson J, Craig EA, Dounavi K. Telehealth as a model for providing behaviour analytic interventions to individuals with autism spectrum disorder: a systematic review. *J Autism Dev Disord.* (2019) 49:582–616. doi: 10.1007/s10803-018-3724-5

67. Ingersoll B, Wainer AL, Berger NI, Pickard KE, Bonter N. Comparison of a self-directed and therapist-assisted telehealth parent-mediated intervention for children with ASD: a pilot RCT. *J Autism Dev Disord.* (2016) 46:2275–84. doi: 10.1007/s10803-016-2755-z

68. Vismara LA, Colombi C, Rogers SJ. Can one hour per week of therapy lead to lasting changes in young children with autism? *Autism.* (2009) 13:93–115. doi: 10.1177/1362361307098516

69. Fisher WW, Luczynski KC, Hood SA, Lesser AD, Machado MA, Piazza CC. Preliminary findings of a randomized clinical trial of a virtual training program for applied behavior analysis technicians. *Res Autism Spectr Disord.* (2014) 8:1044–54. doi: 10.1016/j.rasd.2014.05.002

70. Vismara LA, Young GS, Stahmer AC, Griffith EM, Rogers SJ. Dissemination of evidence-based practice: can we train therapists from a distance? *J Autism Dev Disord.* (2009) 39:1636–51. doi: 10.1007/s10803-009-0796-2

71. Luxton DD, Pruitt LD, Wagner A, Smolenski DJ, Jenkins-Guarnieri MA. Gahm G. Home-based telebehavioral health for US military personnel and veterans with depression: a randomized controlled trial. *J Consult Clin Psychol.* (2016) 84:923–34. doi: 10.1037/ccp0000135

72. Duan L, Zhu G. Psychological interventions for people affected by the COVID-19 epidemic. *Lancet Psychiatry.* (2020) 7:300–2. doi: 10.1016/S2215-0366(20)30073-0

73. Kohrt BA, Jordans MJD, Rai S, Shrestha P, Luitel NP, Ramaiya MK, et al. Therapist competence in global mental health: Development of the ENhancing Assessment of Common Therapeutic factors (ENACT) rating scale. *Behav Res Ther.* (2015) 69:11–21. doi: 10.1016/j.brat.2015.03.009

74. World Health Organization. *Ninth Meeting of the Mental Health Gap Action Programme (mhGAP) mhGAP Forum "Mental Health Capacity Building Within Countries".* (2017). Available online at: http://www.who.int/mental_health/mhgap/forum_report_2017/en/ (accessed June 8, 2021).

75. World Health Organization. *Caregiver Skills Training Package for Caregivers of Children With Developmental Delays or Disorders (Field-Test Version).* (2017). Available online at: https://ezcollab.who.int/pst/ (accessed June 8, 2021).

76. Robins DL. *Chat*^{TM} *- MCHAT R/F Translations*. (2022). Available online at: https://mchatscreen.com/mchat-rf/translations/ (accessed July 1, 2022).

77. Hystad SW, Johnsen BH. The dimensionality of the 12-item General Health Questionnaire (GHQ-12): comparisons of factor structures and invariance across samples and time. *Front Psychol.* (2020) 11:1300. doi: 10.3389/fpsyg.2020.01300

78. The Centre for Clinical Trials and Epidemiological Research at the Chinese University of Hong Kong. *YouthinMind. "Strengths and Difficulties Questionnaire (SDQ) – Chinese".* (2020). Available online at: https://www.sdqinfo.org/py/sdqinfo/b3.py?language=Chinese (accessed November 1, 2020).

79. Bull R, Lee K, Koh IH, Poon KK. Confirmatory factor analysis of the strengths and difficulties questionnaire in Singaporean kindergartners. *Child Care Health Dev.* (2015) 42:109–16. doi: 10.1111/cch.12288

80. Braun V, Clarke V. Using thematic analysis in psychology. *Qual Res Psychol.* (2006) 3:77–101. doi: 10.1191/1478088706qp063oa

81. Rabiee F. Focus-group interview and data analysis. *Proc Nutr Soc.* (2004) 63:655–60. doi: 10.1079/PNS2004399

82. Baggett KM, Davis B, Feil EG, Sheeber LB, Landry SH, Carta JJ, et al. Technologies for expanding the reach of evidence-based interventions: Preliminary results for promoting social-emotional development in early childhood. *Topics Early Child Spec Educ.* (2010) 29:226–38. doi: 10.1177/0271121409354782

83. Békés V. Aafjes-van Doorn K. Psychotherapists' attitudes toward online therapy during the COVID-19 pandemic. *J Psychother Integr.* (2020) 30:238–47. doi: 10.1037/int0000214

84. Poletti B, Tagini S, Brugnera A, Parolin L, Pievani L, Ferrucci R, et al. Telepsychotherapy: a leaflet for psychotherapists in the age of COVID-19. A review of the evidence. *Couns Psychol Q.* (2021) 34:352–67. doi: 10.1080/09515070.2020.1769557

frontiers | Frontiers in Psychiatry

TYPE Policy and Practice Reviews
PUBLISHED 15 September 2022
DOI 10.3389/fpsyt.2022.916256

OPEN ACCESS

EDITED BY
Andy Shih,
Autism Speaks, United States

REVIEWED BY
Rachel Grove,
University of Technology
Sydney, Australia
Hanne De Jaegher,
University of the Basque
Country, Spain

*CORRESPONDENCE
Jonathan Lai
jlai@autismalliance.ca

SPECIALTY SECTION
This article was submitted to
Autism,
a section of the journal
Frontiers in Psychiatry

RECEIVED 09 April 2022
ACCEPTED 22 August 2022
PUBLISHED 15 September 2022

CITATION
Antony C, Campbell M, Côté S,
Bruno G, Tinglin C and Lai J (2022)
Informing care pathways and policies
for children and youth with Indigenous
perspectives to advance Canada's
National Autism Strategy.
Front. Psychiatry 13:916256.
doi: 10.3389/fpsyt.2022.916256

Informing care pathways and policies for children and youth with Indigenous perspectives to advance Canada's National Autism Strategy

Celina Antony[1], Madison Campbell[1], Stephanie Côté[2], Grant Bruno[3,4], Carolyn Tinglin[5] and Jonathan Lai[2,6]*

[1]Department of Health Sciences, McMaster University, Hamilton, ON, Canada, [2]Autism Alliance of Canada, Toronto, ON, Canada, [3]Department of Pediatrics, University of Alberta, Edmonton, AB, Canada, [4]Samson Cree Nation, Maskwacis, AB, Canada, [5]Department of Education, Law, and Society, Simon Fraser University, Burnaby, BC, Canada, [6]Institute of Health Policy Management and Evaluation, Dalla Lana School of Public Health, University of Toronto, Toronto, ON, Canada

In Canada, most services for Autistic people are provided by provincial and territorial governments. However, support for Indigenous Peoples (First Nations, Inuit, and Métis) are under federal responsibility and are outlined by a set of treaties and agreements with the Crown and a few regional governments. This patchwork results in barriers in service access and navigation challenges in many under-resourced communities, including under-diagnosis and potentially life-threatening outcomes. Designing equitable policy structures and processes would reduce harms and meaningfully interface with Indigenous and other racialized communities. The objective of this Policy Practice Review is to provide a framework for the discovery of appropriate care strategies addressing the conceptualization of autism in Indigenous Peoples and to understand the interactions between racialized Autistic peoples and the Criminal Justice System. First, we conducted environmental scans of publicly-accessible government services available in Canada pertaining to autism in Indigenous communities and the justice system, and explored the dissonance with beliefs and perceptions of autism in Northern Indigenous communities. Second, we focused on the interactions of Indigenous and other racialized populations, with an emphasis on Autistic children and youth with the justice system, an interaction that is often life-altering, downstream, and detrimental to health and wellbeing. The implications of this work include identifying the need for Indigenous-led knowledge and policy recommendations for Canada's upcoming National Autism Strategy, informing the need for culturally appropriate multidisciplinary care and facilitating the coordination between health and social services for these communities.

KEYWORDS

autism, policy, Indigenous, services, culturally safe care, Canada, BIPOC

This Policy and Practice Review presents the systems and structures that challenge inclusive policy design and lead to barriers for Autistic people in health, social services, and justice. As part of the development of a National Autism Strategy (NAS) in Canada, there is the opportunity to acknowledge, recognize, and integrate the unique populations that are affected by the systems and structures in place. The current review explores issues encountered by Indigenous Peoples and other racialized groups with the concept of autism and recommendations for the upcoming Canadian strategy. The interactions examined were in relation to health, social services, and justice systems in Canada. When accessing the various programs and services in these systems, Indigenous Peoples in Canada are required to meet the government-established criteria for Indian status. The Canadian Constitution recognizes three distinct groups of Indigenous Peoples: First Nations, Métis, and Inuit (1). Indian status is governed by the Indian Act of 1876, a Canadian federal legislation that authorized the federal government to regulate the lives of First Nations Peoples registered under the Act. This legislation involves the political management of governance structures used within First Nations communities, rights to practicing and teaching Indigenous cultural traditions, and the regulation of Indigenous resources and lands in the form of reserves (2). Indian status is often required for eligibility to varying federal or territorial government benefits, services, and programs (3). The Indian Act resulted in many jurisdictional challenges, a well-known challenge being access to healthcare. Whereas, health services are under the responsibility of Canada's 13 provinces and territories, funding and delivery of health services to Indigenous Peoples are the responsibility of the federal government according to the Indian Act (4). The current structure continues to cultivate mistrust among Indigenous Peoples with regards to existing healthcare services, and this is especially prevalent for developmental disabilities such as autism (5).

With Canada's current work to develop an NAS that adequately serves the needs of Autistic people in Canada, the systematic barriers and inequities that affect racialized communities require critical reflection in the development of appropriate and culturally safe services. Although there are more than 630 First Nations communities situated across Canada, there continues to be an underrepresentation of Indigenous Peoples diagnosed with autism in reports of autism prevalence (6, 7). For instance, a prevalence study found significant under-representation of autism among Indigenous children in Manitoba and Prince Edward Island (8). According to Lindblom, this may stem from the lack of culturally safe diagnostic services and research available within these communities (7). Research has shown that engagement in traditional cultural practices can help to reduce personal challenges and improve social development in Autistic First Nations children (7). Hence, there are evident cultural influences affecting the unique experiences of Autistic Indigenous Peoples, which require additional focus in research to develop culturally appropriate and safe services for Indigenous Peoples.

Community members who use traditional knowledge and healing practices often experience stronger Indigenous identity and improved spiritual health compared to those who did not use traditional methods (9). Despite the value that traditional practices hold for Indigenous Peoples, the diagnostic supports and services available within Indigenous communities often take a Western approach to screening and detection. This dissonance between Indigenous traditional values and the services available for Autistic Indigenous Peoples emphasizes the need for policy and practice which critically consider the historical contexts that shape the interactions of racialized peoples with social systems, such as health, school, and the CJS.

The unique context in which Indigenous and Black Autistic communities exist and interact within Canada

Indigenous Peoples in Canada have faced a prolonged history of colonialism and systemic oppression that have affected their interactions in seeking health and social services. This has taken several forms in addition to the Indian Act, which include but are not limited to: the Indian Residential School system, a system separating Indigenous Peoples from their cultural identities and traditions, assimilating Indigenous students to colonialist societies; the sixties scoop, the mass removal of Indigenous children from their families in the 1960s into the child welfare system; the continual neglect of issues depriving Indigenous communities of basic living needs, such as access to clean water supply on reserves; as well as the implementation of Indian hospitals during the tuberculosis epidemic, which advanced the notion that Indigenous Peoples posed a potential threat to non-Indigenous populations (10–12). These experiences, among countless others, have left a deep imprint in Indigenous cultural conceptions of health and illness, and continue to propagate the mistrust felt by Indigenous Peoples in the Western-oriented health and social systems. Acknowledging the experiences of racialized Autistic individuals who have been forced to participate in Western defined services such as health care and justice systems would be a step toward building trust, and reducing the continued structural violence that shapes Canada's current health programs and services.

Along with a clear disconnect between Western and Indigenous approaches to mental health and wellbeing, the Western approaches adopted within government-funded programs embed in a history of colonialism, institutional racism, systemic barriers, and inequitable policies and practices that continue to affect Indigenous communities (13, 14). To our knowledge, wellness within Indigenous communities can only be adequately evaluated using a holistic model, which seeks balance between all physical, emotional, social, and spiritual aspects of life (13). Specifically, supports and services available to Indigenous communities do not address Indigenous Ways of Knowing, the process of observing, discussing, and making sense of new information traditionally used among Indigenous Peoples (14, 15). This misalignment prevents access to meaningful supports for Indigenous Peoples. For instance, under Canada's Jordan's principle, Indigenous children and youth with autism have the right to care whether located on or off-reserve; however, this principle does not consider youth transitioning into adulthood that may still require funding to access service (16, 17), and the criteria for eligibility for support for autism remain inconsistent across cases as well as unclear (16).

The importance of understanding how Autistic people interact with the CJS is beginning to emerge. The National Health Service (NHS) in the United Kingdom (UK) has released a national strategy for Autistic children, young people, and adults and contains a section dedicated to the interactions of Autistic people and the CJS (18). The intention is to promote research about these interactions and improve the support for Autistic people within the CJS (18). Racialized Autistic peoples in Canada have unique interactions with the Canadian CJS (C-CJS), due to the historical context and legacy of colonialism and racism (19–22). Although CJS interactions with each identity have been studied, to varying extents, studies of the interactions of these intersecting identities are non-existent. Currently, there is an emerging acknowledgment of this lack of research, however, substantial, peer-reviewed articles on the topic continue to be lacking. In reviewing the literature, studies can be found on (i) Black and Indigenous peoples and the C-CJS, (ii) Autistic individuals and the C-CJS, and (iii) the intersection between Autistic and racialized identities.

The Canadian Charter of Rights and Freedoms established the freedoms and rights of every Canadian, regardless of race, gender, age, or background (23). However, the C-CJS has been reported to not uphold this unbiased testament (24), operating with the legacy of colonialism, and the context of racism (19, 20, 22). Black and Indigenous people have higher rates of interactions throughout the CJS such as homicide victimization and offending, police interactions, and incarceration rates (19). Rates of homicide victimization are much higher in Black and Indigenous populations, compared to the overall Canadian population (19). The Indigenous murder rate in Canada is almost seven times higher than the murder rate for non-Indigenous individuals (25). Indigenous adults account for 5% of the Canadian adult population yet represent 30% of the federally incarcerated inmates (26).

Similarly, people with neurodevelopmental dis/abilities, such as autism, are more likely to come into contact with the CJS when compared to their neurotypical counterparts (27). By the age of 21, approximately 20% of Autistic youth will have interacted with law enforcement officers (28). This can be in the form of offender, victim, suspect, or witness (27, 29), though it is important to note that Autistic people are more likely to be victims of violent crime instead of offenders (29). Characteristic behaviors of autism, such as being non-responsive or lack of eye contact, can increase the risks during interactions with the CJS as these behaviors can be perceived as intentional (28). Misinterpretation of these characteristics can result in the Autistic person being identified as dangerous, suspicious, or unreliable (28, 30).

The scarcity of research on racialized Autistic people is an important gap to address when we are discussing how Autistic and Black or Indigenous identities interact with the CJS. Autistic people are more likely than their neurotypical peers to have police interactions, and Black and Indigenous people are more likely to have negative outcomes of interactions with the CJS, in which they are overrepresented (19, 27). If the identities of racialized Autistic people continue to be separated when researching interactions with the CJS there can be no future planning, interventions, or policies that are culturally competent and effective for helping this unique community.

Canada's National Autism Strategy

In Canada, most recent estimates have 1 in 50 children and youth in Canada diagnosed with autism, with more than 50% of those being diagnosed by the age of 6 and over 90% of children diagnosed by the age of 12 (31). It is recognized that these data may not be representative of Indigenous children with autism living in Canada (7, 32). In 2019, the Canadian Government committed to developing its very first NAS (33, 34). Autism strategies can be found in countries such as New Zealand, Malta, Australia, Spain, Scotland, Hungary, the USA, England, Wales, and Northern Ireland (35). In Canada, the NAS falls under the responsibility of the federal government (36). It is critical that Indigenous nations and the Canadian government develop a distinct approach for autism care, addressing the risks of jurisdictional disputes as well as creating an Indigenized approach. This is especially important when highlighting Article 19 of the Calls for Actions by the Commission of Canada's Truth and Reconciliation Commission (TRC) (37) as well as Articles 19, 21, and 23 of the United Nations Declaration on the Rights of Indigenous Peoples (38). For the elaboration of a NAS that is inclusive, it is imperative that a rights-based approach be used. This approach is recognized internationally; for instance, the United Nations Convention on the Rights of Persons with Disabilities and associated United Nations Committee on the Rights of Persons with Disabilities issued Concluding Observations on Canada's initial report (38) include specific recommendations to adopt cross-sectorial strategies to combat inequities and discrimination faced by persons with dis/abilities (39).

These highlight the importance of increasing our understanding of the experiences and needs of Autistic people who are Indigenous, especially in policy and practice. Unfortunately, research focused on intersectionality between race and autism is sparse, but critical to develop, as people with autism cannot separate their identities. The experience of a racialized Autistic person is unique and distinct in and of itself, especially when it involves the CJS.

Our aim, objectives and intended uses

The goal of this Policy Practice Review is to provide a framework for the discovery of appropriate care strategies addressing the conceptualization of autism in Indigenous Peoples and to understand the interactions between racialized Autistic peoples and the Criminal Justice System. We scanned for publicly available autism supports and services with the purpose of highlighting issues and recommendations regarding the silos of support for Autistic people and families across Canada, with a focus on issues experienced by Indigenous children and youth on the spectrum, including jurisdictional disputes for care.

We used the Dis/ability Critical Race Theory (DisCrit) framework when presenting the support, issues, and recommendations highlighted in this scan, as this framework aids in understanding how the identities of racialized Autistic communities influence their interactions with Canadian systems. This worldview informed us to use identity-first language, such as Autistic person, rather than person-first language such as person with autism. Identity-first language has been indicated as the preferred language by most Autistic self-advocates (40). As one of the DisCrit tenets is that it privileges the voices of marginalized populations, traditionally not acknowledged within research, it was essential that we listened to the voices of Autistic self-advocates on their choice for preferred language (41). However, we acknowledge that Indigenous identity is complex and contextual. Meaning, the preference on language pertaining to the colonial definition of autism is context dependent and may differ among Indigenous communities.

DisCrit Theory informed the terminology choice in our discussions about Black and Indigenous peoples' interactions with the CJS. Within our research methodology for investigating the CJS the term BIPOC (Black, Indigenous, People of color) was utilized as a search term, as this term has become popular in some activism spaces, and greatly increased our likelihood of finding information on our population of focus (42, 43). However, BIPOC is not the preferred term of these communities, as it clusters communities and puts Black and Indigenous people at the center of any issues involving people of color (42, 43). This can result in distraction from the systemic issues, or from the accurate representation of how these communities interact with systems (42, 43). The term racialized allows us to focus on the systems and structures that create race and racialized people (those who deviate from the accepted norm, whiteness), and how the multiple systems and services affect these communities (44, 45).

Finally, this tenet informed our decision to not restrict our systematic literature reviews to peer reviewed articles. Racialized and Autistic voices have not had a dominant role in research, and their voices could be neglected in this search had we included this criterion. Therefore, to gain a broader understanding of the current situation for Indigenous and racialized Autistic peoples, it is important to listen to these voices in the alternative formats in which they have been able to speak, including magazine articles, news reports, interviews, and other forms of information sharing that are meaningful to these communities can allow a greater opportunity for these voices to be accurately conveyed, than if our systematic literature review was restricted to peer-reviewed articles.

Using this approach, we set out to answer two questions. First, we aimed to understand Indigenous people's perceptions and conceptualizations of Autism. Second, we aimed to understand the interactions between racialized Autistics and the CJS. Both questions had the purpose of informing policy and systems. To answer these questions, we facilitated two environmental scans, specified to each research question. Environmental Scan 1 focused on the perspectives and experiences of Indigenous Peoples in Canada with autism and dis/ability. Environmental Scan 2 focused on how Indigenous and Black communities uniquely interact with Canadian systems and supports.

Methods

Dis/ability critical race theory framework

To lessen the separation of intersectional identities that affect interactions with health, social systems and the justice system; the DisCrit framework is instructive in understanding this intersection. This framework can be used to understand why these identities cannot continue to be evaluated separately and frame how we can collect and assess data. DisCrit theories state that "racism and ableism are normalizing processes that are interconnected and collusive" [(41) p. 6]. Racism and ableism are grounded in the belief that deviations from a social norm, such as Black, Indigenous, or neurodivergent people (e.g., Autistic), are less valued (25). DisCrit emphasizes that these identities, and their correlated deviations from ideologies, must not be evaluated separately, as the systems themselves are interconnected (41). This highlights the importance of exploring the dearth of research on these interconnected identities, otherwise understood as the concept of intersectionality.

TABLE 1 Inclusion and exclusion criteria for scholarly literature review on the beliefs and perceptions of autism within indigenous communities in Canada.

Inclusion criteria	Exclusion criteria
Literature on ideas, perceptions, values, beliefs, feelings, attitudes, practices, experiences and/or understandings of Autism and disability, through an Indigenous lens	Literature relating to Indigenous communities outside of Canada
Available in the English or French languages	Secondary research sources
	Duplications

Intersectionality is a theoretical framework encompassing unique identities such as race and ethnicity, gender, socio-economic status, and sexuality, and how they interact to affect a person's experiences in society (46). Historically, social endeavors such as slavery, segregation and employment justified these ideologies and current social structures for education, health, and justice may continue to perpetuate these ideologies (25). DisCrit Theory enables the examination of the intersection of identities for an Indigenous Autistic person, further, in the specified example of how racialized Autistic persons uniquely interact with the Canadian CJS (C-CJS). This information will enable the creation of policy and practices that improve interactions and outcomes of interactions with social systems, such as health, school, and the CJS, for Indigenous Autistic people.

Environmental scans

Each environmental scan was composed of three phases: (1) we defined the context using a brief systematic literature review of scholarly articles for both the perceptions of autism in Indigenous communities, and the interactions of Indigenous and other racialized youth with autism with the justice system; (2) we scanned government websites for current programs and services targeting Autistic individuals and their families for both areas; and (3) we co-authored this review with two stakeholders. One was Grant Bruno, a PhD in Medical Sciences student at the University of Alberta. He is also a registered member of Samson Cree Nation, one of the four reserves that makes up the nehiyawak (Plains Cree) community of Maskwacis. He is also the father to an autistic son and currently chairs the Indigenous Relations Circle for the Autism Society of Alberta. The other, Carolyn Tinglin is a PhD candidate and emerging scholar in anti-Black ableism studies. Tinglin's work examines how race, dis/ability, and other identity-based social constructs intersect and impact racialized people. Both had relevant lived experience regarding the current policy environment. Here, stakeholders are defined as those with a vested interest in creating positive change for both the autism community and Indigenous Peoples, and contribute to the cause through experience, knowledge, and expertise.

A Strengths, Weaknesses, Opportunities, and Threats (SWOT) analysis was used to assess how political change, such as elections and varying legislation among provinces, impacted our policy scan (38). This has been previously used by Namugenyi et al. to identify and analyze the internal and external factors that have an impact on the viability of a project, product, place or person entities (47).

Environmental scan of the perspectives and experiences of Indigenous peoples in Canada with autism and dis/ability

Scholarly literature review

Procedure

A literature review was conducted to identify scholarly literature directly relevant to the beliefs and perceptions of autism and dis/ability within Indigenous communities in Canada. The search strategy included the key search terms "Indigenous," "First Nations," "autism," "dis/ability," and "Canada." Indigenous Peoples search filters established by the University of Alberta and the University of Prince Edward Island were used to develop the search strategy for this review (48, 49). Systematic literature searches were performed in the following eight databases: Medline, Cumulative index to Nursing and Allied Health Literature (CINAHL), Web of Science, Scopus, PsychInfo, Arctic Health Publications Database, Circumpolar Health Bibliographic Database, iPortal, and Native Health Database (50). The titles and abstracts of all returned articles were screened thoroughly by the researcher for relevance to the research question. Included articles then underwent full-text review to ensure fulfillment of inclusion criteria (see Table 1). Themes related to the beliefs and perceptions of autism and dis/ability within Indigenous communities in Canada were extracted and gathered for further analysis.

Results

There was a total of 445 returned publications. The titles and abstracts of all returned publications were thoroughly screened, which led to the exclusion of 381 articles. Of these excluded articles, 86 were duplicates and 295 were on topics unrelated to the research question. The remaining 64 publications then underwent full-text review, after which there remained a total of five articles included for review (see Figure 1). The five articles in this review exhibited the following themes:

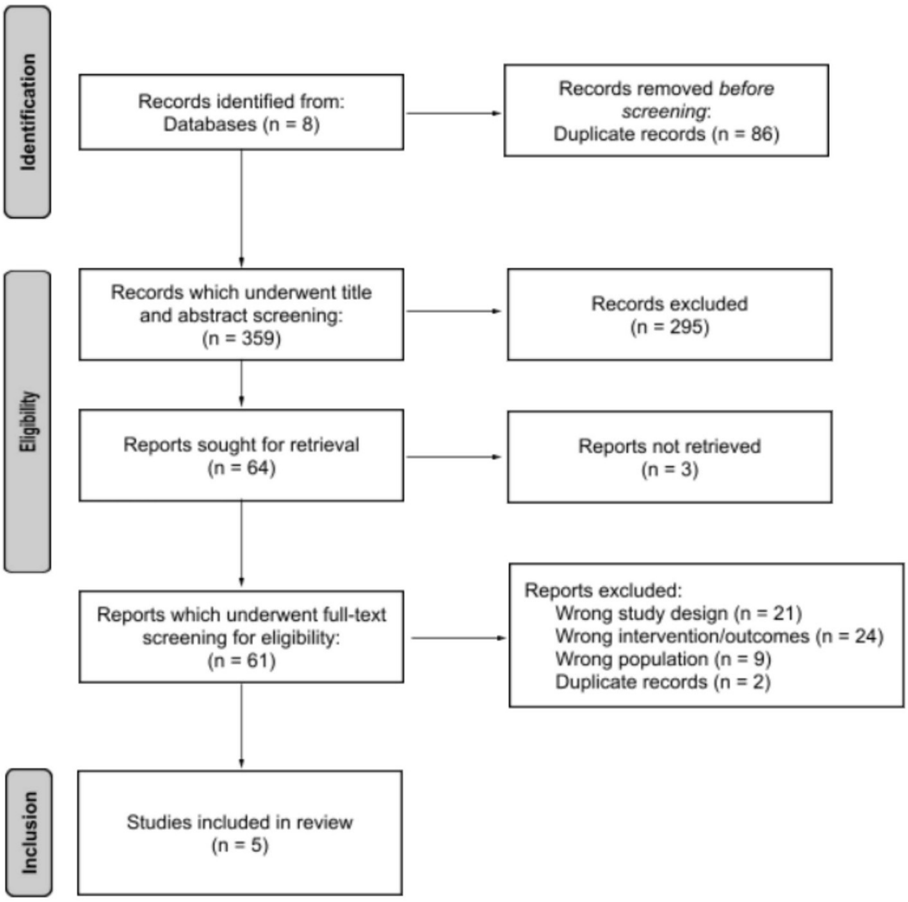


FIGURE 1
Systematic literature eligibility results for the perspectives and experiences of Indigenous peoples in Canada with autism and dis/ability. Adapted from: Page (51).

Theme 1: Inclusion and acceptance

The theme of inclusion and acceptance demonstrates the warmth with which Indigenous Peoples embrace autism and dis/ability. In some Indigenous cultures, autism is not conceptualized as a form of deficit or shortcoming (52). Rather, it is acknowledged and accepted by community members, with the intent to support the individual to their strongest capacity (52–54). This contrasts with the medicalized, deficit-model employed within Western societies, which labels and treats the individual's condition as a medical issue requiring treatment intervention. To our knowledge, Indigenous Peoples often do not label or categorize community members based on their abilities (52). Labeling may evoke a sense of division among community members, which can cause strain in community relationships. Alternatively, many Indigenous communities treat Autistic or dis/abled people as any other valued member of the community, and they receive the support of their family and community members in their daily lives (52). In sum, Indigenous communities in Canada foster a climate of inclusion and acceptance of the diverse contributions of all community members, despite their unique abilities.

Theme 2: Supportive network of family and community members

The theme of a well-supported network of family and community is articulated across all five of the articles included in this review. Indigenous ways of living often emphasize the interconnectedness of family and community, promoting a sense of inclusion and acceptance of people of all abilities (53, 55). When defining dis/ability, it was demonstrated that some Indigenous communities place emphasis on the community context (55). Dis/ability can be considered to impact not merely the individual themselves, but the family and community with which they interact (55). Once a dis/ability is recognized, it is embraced by community members and can become the shared responsibility of the individual's family and community to help provide a vital source of support (52–54). This close-knit network of family and community can provide a powerful sense of inclusion and empowerment to the Autistic or dis/abled person.

Furthermore, within some Indigenous communities, dis/ability can be viewed as a result of disharmony or imbalances in aspects of the mind, body, and spirit (55, 56). Dis/ability can also be understood to manifest due to imbalances within various environmental contexts, such as family, school, and the greater community (55). This reflects the holistic approach that is habitually employed within Indigenous communities to assess health and wellbeing. Thus, strengthening one's connections with family, school, and community can help to restore a sense of harmony and balance in people with dis/abilities, optimizing their overall health and wellbeing (55).

Theme 3: Engaging in Indigenous cultural identity

The theme of engaging in Indigenous cultural identity to optimize wellbeing conveys the need for culturally safe supports and services for Indigenous Peoples with autism and dis/ability. To our knowledge, the implementation of Indigenous ways of knowing, being, and doing within services can help to support the desire of Indigenous Peoples with dis/abilities to partake in traditional culture in a meaningful way (56). Practicing one's Indigenous cultural identity, through the teaching and learning of cultural traditions, can serve as a protective factor in conserving the wellbeing of Indigenous Peoples, both individually and collectively (56).

In Lindblom's case study involving five Autistic First Nations children in British Columbia, the meaning of music to the children was examined (52). The study findings indicated that there was a lack of cultural sensitivity in the music interventions allocated for Autistic First Nations children in British Columbia (52). Lindbolm's following article highlighting two of these cases explored the intersections of dis/ability, gender, ethnicity, and culturally safe traditional music interventions in the improvement of communication and social inclusion among Autistic First Nations children (53). Lindbolm's findings suggested that meaningful engagement of Autistic First Nations children in traditional practices, such as Indigenous music interventions, can help to ease personal challenges and enhance communication and social development (53). Hence, the implementation of supports and services that are culturally safe may help to strengthen Indigenous cultural identity and support the development of Indigenous children with autism.

Gray literature scan

A gray literature scan was performed to assess all of the existing policies and programs being delivered within Northern Indigenous communities in Canada, specifically in the territories of Nunavut, Yukon, and the Northwest Territories (NWT). Following this gray literature search, a S.W.O.T. analysis was performed to critically comment on the programs and services available in the North, in terms of their strengths, gaps, and areas for improvement (see Table 2).

TABLE 2 Summary of S.W.O.T analysis for gray literature search on the perspectives and experiences of indigenous peoples in Canada with autism and dis/ability.

S.W.O.T components	Indigenous perspectives on autism
Strengths	Programs and services incorporate autism within the broader lens of "disability." This use of terminology can minimize the likelihood of labeling, which can be detrimental to Indigenous communities.
Weaknesses	There is a lack of programming specified toward Autistic people or Indigenous Autistic people. There are only two disability-focused programs in Yukon, the Home Care Program, and the Home Repair Program, which include First Nations residents in their eligibility, but do not have services specified for their needs. Further, the language used in program information reflects person-first language, which portrays disability as a deficit and may misalign with Indigenous cultural views.
Opportunities	There is an evident need for culturally safe programs, developed in collaboration between the Canadian government and Indigenous Peoples within the context of the NAS. Additionally, changing to identity-first language can reduce the stigma of disability and better reflect Indigenous cultural views for those seeking support.
Threats	There is a dearth of research focused on autism in Indigenous communities in Canada. Existing studies have found significant underrepresentation of Indigenous Peoples diagnosed with autism in reports of autism prevalence. This lack of research could result in a lowering of prioritization for the Canadian government, resulting in a continued lack of culturally safe support for Indigenous Autistic people. Moreover, limited internet access in the North may reduce online availability of pertinent information for the advancement of policies and programs.

Recommendations for decision-makers in developing autism supports for Indigenous peoples

Actionable recommendations

Although Indigenous Autistic people in Canada experience some of the same challenges as non-Indigenous Autistic individuals, it is important to recognize and understand the complex layers and intersections colonialism has created. The following recommendations are meant to be approached with a distinction-based focus, meaning that there is a need to recognize the diversity and uniqueness of each group including differences for First Nations, Métis, and Inuit, as well as geographic differences such as urban, rural, and remote as well as differences in culture and language. It should also be noted that these recommendations are not comprehensive and as the NAS is implemented new priorities may arise. One more thing to consider is some Indigenous communities' challenges need to be addressed upstream. For example, some communities may have challenges around poverty, chronic housing shortages, boil water advisories, etc. that may need to be addressed at the same time as addressing the challenges around autism. Using an approach that allows the community to lead the project in full partnership is now considered best practice. The following are actionable recommendations to address the aforementioned issues and themes:

1. Develop an Indigenous autism engagement framework that recognizes and honors the diversity of Indigenous peoples across Canada. Build relationships with key Indigenous individuals and groups using a distinction-based approach by creating national autism advisory groups for First Nations, Métis, and Inuit, respectively.
2. Assess the barriers to healthcare including diagnostic assessments, services provision, socio economic considerations, and overall mistrust of the health care system. Areas to focus on include racism, ableism, poverty, and current and historical injustices. An actionable step would be to, in partnership with Indigenous groups, develop a report that focuses on the barriers and solutions to accessing healthcare.
3. Partner with Indigenous communities and organizations to collect data and evidence to understand autism prevalence and the lived experience of autism in Indigenous communities. Listen to Indigenous Autistic people in the ways that they wish to be heard, to ensure the collection of data and evidence that is both meaningful and accurate to Indigenous communities. Provide support for Indigenous-led research in partnership with communities and organizations to create empirical evidence that reflects the wants and needs of Indigenous Peoples in Canada.
4. Explore what Indigenous led and culturally safe services, assessments, and interventions would look like. In full partnership with Elders, traditional knowledge keepers, community members, Indigenous autistic individuals, and Indigenous service providers, create and pilot culturally-informed autism services.
5. Provide equitable funding across Canada for Indigenous communities and organizations to provide culturally safe autism services, appropriate autism assessments, autism awareness and education. Research and develop a report that addresses jurisdictional disputes between federal, provincial, municipal, and band councils that are specific to autism in Indigenous communities and explore opportunities to address these challenges.

TABLE 3 Search terms for racialized autistic individuals and the CJS.

Domain:	**Autism**	**Justice system**	**Black and Indigenous peoples**
Search terms:	Autis*	• Crim* • Police • Arrest • Charge • Justice • Court • Law	• Black • Indigenous • Aboriginal • First Nations • Métis • Inuit • Rac* • Minority • Intersect*

* Refers to the truncation of a search term, which is the shortening of a search term in a literature search to attract words with the same root word, but different endings.

Environmental scan of how Indigenous and Black communities uniquely interact with Canadian systems and supports

Scholarly literature review

Procedure

Four Databases were searched: ProQuest, Web of Science, PubMed, and EBSCOhost. Truncations were implemented to retrieve articles that contained multiple applications of the word stem, in order to broaden the search and generate as many relevant articles as possible. The search terms pertained to three domains to explore the research pertaining to racialized Autistic individuals and the CJS (see Table 3). The domains included autism, Black and Indigenous peoples, and the Justice system. Each domain contained several possible search terms that could attempt to address the domain, connected by the Boolean operator "OR." Each of the three domains was connected with the Boolean operator "AND." The final search terms were applied to each database: (autis*) AND (crim* OR police OR arrest OR charge OR justice OR court OR law) AND (black OR indigenous OR aboriginal OR first nations OR métis OR inuit OR rac* OR minority OR intersect*).

Results

In total 859 publication records were collected from the four databases. The publications were then scanned based on

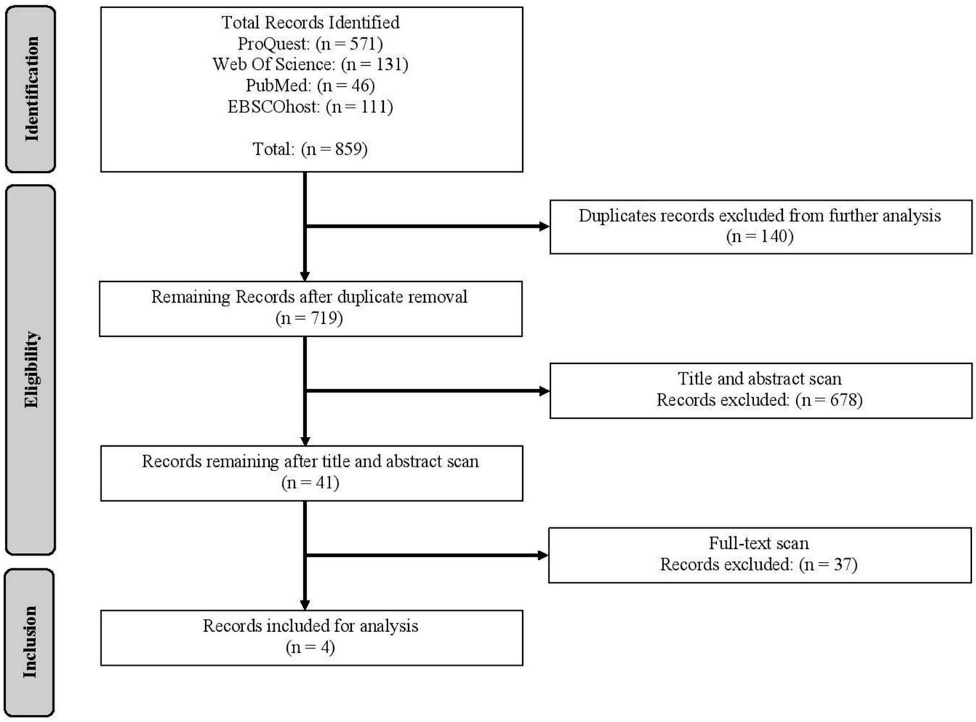


FIGURE 2
Systematic literature eligibility results for how Indigenous and Black communities uniquely interact with Canadian systems and supports.

inclusion and exclusion criteria (depicted in Figure 2). The inclusion and exclusion criteria (depicted in Table 4) focused on ensuring that relevant, recent publications addressing the intersectionality of identities for racialized Autistic individuals interacting with the CJS were included for final assessment. The publications were first scanned for duplicates; 140 publications were excluded from further analysis. A title and abstract search were then conducted using the inclusion and exclusion criteria, after which 678 articles were excluded. Lastly, based on this same criterion, a full text scan was conducted; 37 articles were excluded from the qualitative analysis. In conclusion, after applying the inclusion and exclusion criteria 855 publications were excluded, and four publications were included in the final qualitative analysis of the systematic literature review.

All articles included were based in the United States of America (USA) and focused on Black Autistic people's interactions with the CJS. Articles geographically based outside of the USA or focusing on Indigenous Peoples were not found during this literature search or were not applicable to the inclusion and exclusion criteria. The four articles examined had three main themes:

Theme 1: Increased risk of interactions for racialized Autistics with the CJS

The theme of increased risk of interactions for racialized Autistics with the CJS explores how intersecting identities can lead to and prolong interaction with the CJS. For example, having racialized identity can prompt interaction with police, such as the situation explored by Vargas, where the characteristics and presentation of autism (such as aversion to touch) can escalate an interaction with police and lead to negative outcomes (57). This trend of one identity initiating the interaction, and one identity increasing the risk and negative outcomes continues throughout this literature search. Carley, as well as Solomon and Lawlor explored how the characteristic behaviors (such as elopement) and fixations of autism can lead to police interactions, during which being a racialized Autistic person increases negative outcomes (58, 59). Meaning, further prejudice toward the racialized Autistic person, misunderstanding of characteristic behaviors, and continued negative outcomes with a lack of support for the individual (58, 59). Davenport et al. documented a program to train Autistic Black adolescents on how to interact with police (60). Davenport et al. recognized the increased risk of interactions for both identities (without research specified to

TABLE 4 Inclusion and exclusion criteria for scholarly literature review on racialized autistic individuals and the CJS.

Inclusion	Exclusion
Researcher has access to the full article	Researcher does not have access to the full article
Published in the English or French language	Published in languages other than English and French
Unique publication	Duplicate of another publication
Article's subject centers on how each of the three domains of the research question intersect and affect each other. Articles will do this through discussing at least one facet of each of the three domains: (1) BIPOC (2) autism (3) criminal justice system In order to assure that the main research question is able to be answered: What are the outcomes of Autistic Black and Indigenous youth's interactions with the Justice System	Article's subject does not center on the intersection of each domain, that needs to be considered while aiming to answer the question of how the intersecting identities affect interactions with the criminal justice system.
Article focuses on Autistic people	Article does not make clear whether Autism qualifies in their definition of a developmental disability; the article discusses dis/abilities or other neurodevelopmental disorders that are not autism, e.g., FASD instead of ASD.
The subject of the article is focused on youth; the publication depicts multiple life stages which includes youth	The subject of the article excludes youth, focusing on adults or elderly people.
The subject of the article is non-fictional	The subject of the article is fictional

the intersection of identities), and negative outcomes from individuals within the CJS's notions and conceptions of either identity (60).

Theme 2: Lack of culturally competent supports and interventions

The theme of lacking culturally competent support and interventions focuses on how systems fail to adequately support racialized Autistic youth at multiple stages of interactions with the CJS. Meaning, that systems lack cultural competence in understanding how racialized Autistic persons uniquely interact with the CJS in terms of prevention of crime, arrest, charging, court appearance, and incarceration. This could be due to a lack of understanding on how characteristic behaviors of autism (such as fixation, and elopement) can lead to interactions with the CJS, and/or the lack implementation of supports which could act as preventative measures such as demonstrated by Carley as well as Solomon and Lawlor (58, 59). Carley then further explored how Darius McCollum was treated differently in the courts due to his intersecting identities of being Black and Autistic, which led to harsher sentencing and misunderstanding of the locus of control (58). As the judge perceived Darius McCollum's actions to be due to solely his own volition and control, rather than a continued lack of support, and systemic misunderstanding of Autism, this led to continuing the sentencing of jail time, and the continued lack of support for coping with this fixation which led to criminal behavior (58). At the initial interaction police are inept to interact with Autistic people and the manifestation of autism characteristics and behaviors, leading to escalating situations. Especially when considering the prejudices that police enter the situation with if the Autistic person is Black or Indigenous, as explored by Vargas (57). The study and program created by Davenport et al. was a response to this lack of culturally competent support and interventions (60). Their goal was to not only recognize the unique interactions Black Autistic persons have with police, but also to train these adolescents on how to navigate this interaction (60).

Theme 3: Uninformed and biased systems

The final theme of uninformed and biased systems describes how the CJS fails racialized Autistic youth at every step, due to a lack of knowledge, prejudice, misinformation, racism, and ableism. At initial interaction police are ill equipped to interact with Autistic people and their manifestation of characteristics, leading to escalating situations. This is especially true when considering the prejudices, the police begin the situation with if the Autistic person is Black, as the results of this literature review focused on. This was explored by Vargas as the scenario was initiated when the police were called because a Black man appeared suspicious while sitting down waiting outside the library (57). The interaction is believed to have escalated further due to both the characteristics of autism, and the Black identity of the man (57). Furthermore, Carley explores how the combined identities of being Black and Autistic led to harsher, inappropriate sentencing, and a continued lack of support, due to prejudice and racism (58). For example, the judge demonstrated a lack of understanding and prejudice of autism as the judge was quoted as saying that Asperger's syndrome (a previous term used for diagnosis of autism) was a "dangerous, mental disorder" (58). This prejudice is not restricted to the CJS, as explored by Solomon and Lawlor where, when Black mothers would reach out to healthcare teams for support with their children's elopement, they were often dismissed for their concerns (59). This is a trend not seen in their white counterparts (59). This dismissal provides no support for the behavior, which in turn can lead to CJS interaction and involvement, potentially putting children at risk of further negative outcomes (59). Finally, these inefficient and biased systems can lead to communities creating support systems in response, as was explored in the creation and implementation of the program explored by Davenport et al. (60). Davenport et al. recognized that Black Autistic adolescents are too much at risk to ignore how these intersecting identities disproportionately result in the risk of negative outcomes (60).

Gray literature scan

Each Province and Territory's government website, as well as the federal government websites, were scanned for resources that Autistic people and their support systems could access when interacting with, or having interacted with, the C-CJS. Following this gray literature search, a S.W.O.T analysis was performed to critically comment on the programs and services available in Canada, in terms of their strengths, weaknesses, opportunities, and threats (see Table 5). Two provinces and two territories, Ontario, Alberta, Yukon, and Northwest Territories indicated some support available for people who are involved with the C-CJS in some capacity. In analyzing the descriptions of available support on ministry websites, the depth of information was restricted to short descriptive texts, ranging from indicating access to support through this program, assurance of continued human rights, or indicating availability of community programs. For example, Ontario indicates that Children's and Young Person's rights remain recognized, even when the youth is in the justice system. Alberta promises specialized support from experts to help if someone with developmental dis/abilities has additional needs because of mental illness, behavioral issues, addictions and/or involvement with the law. The Yukon has defined two separate programs. The first is for offenders and is based on changing behavior by targeting antisocial thoughts and cognitive skills deficits. The second collaborates with Yukon First Nations to incorporate Indigenous cultural heritage into the correction processes. The NWT has created programs designed to address the underlying issues that may contribute to reoffending.

TABLE 5 Summary of S.W.O.T analysis for gray literature search on how indigenous and black communities uniquely interact with Canadian systems and supports.

S.W.O.T components	Racialized Autistic's interactions with the CJS
Strengths	The need for specialized support for Autistic people, relative to the CJS, has begun to emerge. Meaning, some provinces and territories have begun to implement programs for CJS support for Autistic people, or statements regarding continued human rights.
Weaknesses	Not every province and territory has acknowledged this need, meaning, inconsistency in support available across Canada. Acknowledgment of intersectionality necessitating specified care was not found.
Opportunities	The NAS provides an opportunity for incorporation of specified supports for intersectional identities that are consistent across Canada. Further, the Specialized Court: Wellness Court in the Northwest Territories provides an example for future development.
Threats	As research on racialized Autistics, such Black and Indigenous people, is greatly lacking, this could result in programs specified to this population not being prioritized by the Canadian government when developing a NAS. Meaning, the Canadian government might choose to focus on populations and issues that have a greater wealth of information on needs, resulting in programs' continued failure in supporting racialized Autistics in interacting with the C-CJS.

Recommendations for decision-makers in supporting racialized autistic people with the criminal justice system

Actionable recommendations

The following are actionable recommendations to address the issues and themes discussed relative to racialized Autistic people and the C-CJS:

1. Commit resources including funding, technology, and human resources to develop research examining the ways in which Canadian policing policies and practices impact racialized youth with autism. Creating this research opportunity is imperative as there are negligible current research studies that investigate how justice system policies and practices impact racialized and Autistic people.
2. Ensure racialized Autistic people are central to research focused on race, dis/ability, and policing. Furthermore, ensure equitable opportunities for racialized people with autism to meaningfully engage in this area of research, recognizing the importance of community-based research for and by community members.
3. Review and/or develop policing policies which address anti-Black racism, as well as racialized ableism, while supporting equitable care of racialized Autistic people in carceral settings. In other words, create policies and practices grounded in anti-racism and anti-ableism, ethics of care, and culturally competent protocols which guide encounters with the justice system.
4. Establish a mandatory training program for law enforcement, incorporating the experiences and knowledges of racialized Autistic people, grounded in anti-racist, anti-ableist approaches to policing.

5. Be proactive in identifying opportunities for justice system stakeholder engagement being mindful of the historical, political, and social contexts and implications of policing minoritized Canadians.
6. Create accessible services for racialized Autistic people who are incarcerated, based on individual and community needs. This might be accomplished through community round tables, stakeholder forums, and justice system policy reviews.
7. Implement an anonymous *police encounters reporting system/hotline* to collect important data about law enforcement encounters with racialized Autistic peoples across Canada.
8. Provide additional resources and funding to agencies which serve racialized youth with autism, to research and address the psychosocial, physical, and emotional wellbeing of intersectionally-positioned youth who are incarcerated.

Discussion

In this paper, we synthesize findings from both the perspectives and experiences of Indigenous Peoples in Canada with Autism and Dis/ability and how Indigenous and Black Communities uniquely interact with Canadian systems and supports in order to clearly present commonalities and develop policy and practice recommendations. To understand the data and recommendations presented in this article, it is essential to maintain understanding of the historical, and current context within which Indigenous Peoples in Canada live. Indigenous Peoples in Canada live within the historical context, and continual legacy, of colonialism and systemic oppression. This has taken forms such as the Indigenous Residential School system, the sixties scoop, the implementation of the Indian hospitals, and continual neglect of basic needs, such as clean water on reservations (10–12, 61). This historical, and current, context and events have had detrimental effects on Indigenous people's perceptions and trust of Canadian systems and supports and thus lead to negative health outcomes (5).

This context further extends into understanding how Indigenous Peoples, specifically Indigenous Autistic people, uniquely interact with Canadian systems and supports. The two main systems explored in this article, diagnostic services and the CJS, both have roots in colonialism and institutional racism which continue to affect Indigenous communities, and other racialized groups such as Black communities (13, 19). This can be seen in terms of a misalignment of conceptualization of autism, underdiagnosis, and supports and systems that fail to acknowledge and implement Indigenous ways of knowing (14). Furthermore, this can also be seen in the overrepresentation of Indigenous Peoples in the CJS (19, 26). This historical, and current context allows for better understanding on why these systems are currently ineffective, and inadequate at supporting Indigenous communities within Canada, specifically related to the topic of autism.

To ensure that Indigenous Peoples with autism have access to meaningful supports and services across the lifespan, it is crucial to implement culturally safe health and social services within Indigenous communities. Indigenous Peoples identify with distinct cultural definitions of wellness and experience unique interactions with the criminal justice system. Nonetheless, Indigenous Peoples are currently required to adopt Western perspectives on disability to receive adequate access to services and funding through the diagnostic process, which demonstrates the continued colonialism, institutional racism, systemic barriers with which the health and criminal justice systems are embedded. Government programs and policies often adopt a Western, deficit-model approach to assessment tools and services, which misaligns with Indigenous cultural values and practices. Appropriate care and social service pathways for Indigenous Peoples with autism can be developed through strategies that acknowledge the conceptualization of autism within Indigenous communities and the unique challenges that Indigenous Peoples face across the lifespan.

The findings from the included systematic literature reviews reveal the growing gaps in research regarding autism in Indigenous communities in Canada and how racialized Autistic Peoples interact with the CJS. With a total of five articles included for the former area of study and four articles identified for the latter, several common themes surfaced across the literature with regards to the perceptions of autism in relation to Indigenous Peoples.

In Indigenous communities in Canada, autism is acknowledged with acceptance and is received with inclusion and empowerment by community members. Indigenous cultures are distinctly welcoming of diversity and embrace the unique abilities and contributions of all community members. When an individual conveys specific needs, community members can take up the instinctive responsibility to help them perform in their roles to the best of their capacities (52–54). This lies in direct contrast to the perceptions of autism demonstrated by individuals in the CJS. The Judge trying Darius McCollum stated that Asperger's syndrome (a previous term used for diagnosis of Autism) was a "dangerous, mental disorder" (58). Solomon and Lawlor continued this discussion of how perceptions can impede gaining adequate support and fair treatment relative to the CJS (59). When Black mothers would seek help in addressing elopement (a behavior that can present in Autistic people), they found their concerns were often dismissed and support in altering this behavior which could lead to CJS involvement was absent (59).

To our knowledge, dis/ability in some Indigenous communities is not an isolated experience. Rather, it has extensive impacts on the family and community with which the individual interacts (55). Indigenous Peoples hold a strong interconnectedness with community, as it serves as a critical source of support for Indigenous Peoples, especially those with autism and dis/ability (52–54). Thus, a disharmony in one's connections with family and the greater community may substantially influence Indigenous experiences with autism and dis/ability, especially when individuals try to seek support from systems outside of the local community.

When reviewing the system's cultural competency, relative to the CJS, there is a need for improvement particularly around a lack of understanding the characteristics of autism, and the potential to lead to interactions with the CJS (58, 59). Or, the lack of support which could prevent interaction with the CJS (58, 59). This is essential to recognize as being a racialized Autistic person can have risks when interacting with the CJS (57). This has led to communities being the ones to develop supports and programs in order to not only recognize that this is an issue, but also take steps toward creating solutions and supports (60).

Indigenous Peoples with autism and dis/ability often desire to engage in Indigenous cultural traditions and practices (56). However, there is a lack of culturally safe interventions and services available for Indigenous Peoples with autism (7, 52). This can lead Indigenous Peoples to feel disconnected from their cultural identity, which can pose detrimental impacts on the mental health and wellbeing of Indigenous communities (62). According to Lindblom, it was determined that the interactions of Autistic First Nations children with traditional practices, such as Indigenous music interventions, can help to ease personal difficulties and improve communication and social interactions (53). Hence, the incorporation of interventions and services that are culturally safe may help to strengthen Indigenous cultural identity and, as a result, enhance the development of Autistic Indigenous children.

Within our results, two key issues emerge. First, one of the main weaknesses found in the environmental scans was a lack of acknowledging how intersectional identities inform the need for specified programs and supports. Meaning, that there was scant mention of how Indigenous ways of knowing are incorporated into current supports and programs, nor how Indigenous conceptualizations of autism are taken into consideration. Furthermore, the implications of how the intersectional identities of racialized Autistics would affect interactions with the CJS, and resulting necessitated programs, were not found. Second, the threats portion of the S.W.O.T analysis provided further insight into the challenges that could arise while aiming to reduce this deficiency. Both the field of Indigenous perspectives on autism, and racialized Autistic interactions with the CJS are undeveloped. This could create a barrier when the Canadian government is deciding on what new programs to create, and how to create them. Meaning, they might be more likely to gather political support and gain funding if they choose to implement programs with a wealth of evidence on the likelihood of success, and dire need of implementation. Thus, this lack of research not only is an issue in and of itself, but it further creates a barrier when trying to develop and implement programs related to these fields.

To create programs that can appropriately support Indigenous autistic people, and address relevant issues the actionable recommendations are as follows:

1. The current barriers and systems need to be assessed with a historical context in mind. These areas include racism, ableism, poverty, current and historical injustices, as well as the current policies and practices related to policing. This is essential to future research and planning of policies and practices as without a context on the current situation, and what has led to its culmination, these new implementations could continue to be ineffective at addressing the needs and issues of Indigenous Autistic and other racialized communities.
2. It is critical that commitment is made to the development of strength-based Indigenous-led research partnerships and support provided for the synthesis of research that reflects the health and social needs of Indigenous Peoples living in Canada. Within research partnerships, it is imperative that opportunities for meaningful engagement are present and accessible to Indigenous communities throughout the research process. Ensuring that the entire process is Indigenous-led can allow for the performance of research that is both meaningful and applicable to Indigenous communities.
3. New culturally informed, and culturally competent policies and practices must be curated. These new policies must be grounded in anti-racism, anti-ableism, and ethics of care to adequately serve the communities they are being curated for. To do so, these interventions must be created and developed in partnership, in meaningful and sustainable ways.
4. Equitable funding and resources are required to adequately invest in the development of culturally safe and competent services, assessment tools, and programs for Autistic Indigenous Peoples. These services and programs will aim to address the psychosocial, physical, and emotional wellbeing and needs of intersectionality-positioned populations with Autism across the lifespan.

Limitations

A critical gap exists in the literature with regards to Autism in Indigenous communities in Canada and how Autistic Indigenous Peoples interact with the CJS. The lack of Indigenous perspective of autism in Canada may derive from the over-emphasis of research on fetal alcohol spectrum disorder within Indigenous populations (63). It can be argued that the focus of FASD research further reinforces negative stereotypes of Indigenous people and is a form of victim blaming. In the absence of a comprehensive understanding of the various factors that shape Indigenous experiences with Autism and their interactions in seeking health and social services, the needs of Autistic Indigenous Peoples may emerge to be misrepresented in developing policies and programs. This emphasizes the need for further research in this area of study, to inform the development of culturally safe and competent services for Autistic Indigenous Peoples across the lifespan.

Another critical gap in data, pertaining to how racialized peoples interact with the criminal justice system, outside of the USA, affected our ability to create a holistic picture of how racialized Autistic people interact with the C-CJS. Although recommendations were formulated based on the literature reviewer and environmental scan, this limitation in the applicability of data is important to note.

We also recognize the limitation of using environmental scans of publicly accessible government websites to search for current programs and supports. This could have restricted the programs with which we were able to discover, as some programs might be in development, yet have not been listed publicly at the time of the search. Furthermore, we did not have access to in-depth descriptions of programs and were restricted to what was available to the public.

Additionally, it is of consideration that lack of critical infrastructure such as internet access may be limited in some of the Northern provinces and territories in Canada. This would have limited the information that we were able to retrieve, as other pertinent information relating to the current programs and services may have been publicized on alternative platforms beyond the online network. Some government websites were devoid of information on their last update. This could have also impacted the recency and relevance of the programs found, due to this missing information.

Conclusion and future directions

There is still much work to be done to modify Canadian supports and systems to adequately care for Indigenous Autistic peoples. This Policy and Practice Review identified pertinent issues relative to these systems and their ability to provide culturally safe care, and prevent harm, specifically diving into the example of racialized Autistic peoples and the CJS. However, the fields of research related to autism within Indigenous communities in Canada, and how Autistic Indigenous individuals interact with Canadian systems are still in their infancy. Research should be based on continuous and meaningful engagement with Indigenous communities to assure that all changes made, and innovations proposed, are based in culturally safe and competent practices. Mutual relationship and collaboration with Indigenous Elders, traditional knowledge keepers, community members, Indigenous Autistic individuals, Indigenous service providers, and other relevant stakeholders is pivotal throughout the entirety of the research process, from the development of methodologies to the analyses of results. Partnerships with Indigenous communities in the collection of data and evidence that affects Indigenous Peoples should be Indigenous-led, to ensure that data collection and research is meaningful to Indigenous communities. Further, within this engagement there should be capacity building and training for Indigenous researchers, services providers, and policy makers, to assure continued culturally safe and competent practices extending beyond this initiative. This is essential, as with the described historical and current context within which Indigenous Peoples live, self-determined and Indigenous-led research and policy development through Canada's upcoming NAS can act as a method to overcome barriers due to historical neglect and abuse. Should the above recommendations be implemented, there could be a significant impact on how Indigenous Peoples in Canada define autism, what success looks like for supports, and mechanisms to focus resources and innovation.

There remains extensive work to be done in the development of adequate and meaningful supports that serve the needs of Autistic Indigenous Peoples and racialized communities in Canada. However, there are clear opportunities for developments in research and policy through the development and implementation of a NAS, and actionable steps that will allow for the achievement of equitable service access and culturally safe and competent care for such communities.

Author contributions

CA and MC wrote the initial draft equally as shared first authors. GB and CT contributed academic and lived experience insight. JL and SC conceived of the presented idea and supervised the work. All authors discussed contributed and approved the final manuscript.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

References

1. Indigenous and Northern Affairs Canada. *Indigenous Peoples and Communities*. Government of Canada (2009). Available online at: https://www.rcaanc-cirnac.gc.ca/eng/1100100013785/1529102490303

2. Minister of Justice. *Indian Act*. (2019). Available online at: https://laws-lois.justice.gc.ca/eng/acts/i-5/fulltext.html

3. Indigenous Services Canada. *About Indian Status*. Government of Canada (2008). Available online at: https://www.sac-isc.gc.ca/eng/1100100032463/1572459644986

4. Health Canada. *Canada's Health Care System*. Government of Canada (2011). Available online at: https://www.canada.ca/en/health-canada/services/health-care-system/reports-publications/health-care-system/canada.html

5. Browne AJ, Varcoe C, Lavoie J, Smye V, Wong ST, Krause M, et al. Enhancing health care equity with Indigenous populations: evidence-based strategies from an ethnographic study. *BMC Health Serv Res.* (2016) 16:544. doi: 10.1186/s12913-016-1707-9

6. Government of Canada. *First Nations*. (2008). Available online at: https://www.rcaanc-cirnac.gc.ca/eng/1100100013791/1535470872302

7. Lindblom A. Under-detection of autism among First Nations children in British Columbia, Canada. *Disabil Soc.* (2014) 29:1248–59. doi: 10.1080/09687599.2014.923750

8. Ouellette-Kuntz H, Coo H, Yu CT, Chudley AE, Noonan A, Breitenbach M, et al. Prevalence of pervasive developmental disorders in two Canadian provinces. *J Policy Pract Intellect Disabil.* (2006) 3:164–72. doi: 10.1111/j.1741-1130.2006.00076.x

9. George J, MacLeod M, Graham K, Plain S, Bernards S, Wells S. Use of traditional healing practices in two Ontario first nations. *J Commun Health.* (2018) 43:227–37. doi: 10.1007/s10900-017-0409-5

10. Eric Hanson, Daniel P. *Games, Alexa Manuel. The Residential School System*. Indigenous Foundations (2020). Available online at: https://indigenousfoundations.arts.ubc.ca/residential-school-system-2020/

11. Eric Hanson. *Sixties Scoop*. Indigenous Foundations. Available online at: https://indigenousfoundations.arts.ubc.ca/sixties_scoop/

12. McGregor D. Traditional knowledge and water governance: the ethic of responsibility. *AlterNative Int J Indig Peoples*. (2014) 10:493–507.

13. Hill DM. Traditional medicine and restoration of wellness strategies. *Int J Indig Health.* (2009) 5:26–42. doi: 10.3138/ijih.v5i1.28976

14. Redvers N, Blondin B. Traditional indigenous medicine in North America: a scoping review. *PLoS ONE.* (2020) 15:e0237531. doi: 10.1371/journal.pone.0237531

15. Berkes F. Indigenous ways of knowing and the study of environmental change. *J R Soc N Z.* (2009) 39:151–6. doi: 10.1080/03014220909510568

16. Blackstock C. Jordan's principle: Canada's broken promise to First Nations children? *Paediatr Child Health.* (2012) 17:368–70. doi: 10.1093/pch/17.7.368

17. Sinha V, Wong S. Ensuring First Nations children's access to equitable services through Jordan's principle: the time to act is now. *Paediatr Child Health.* (2015) 20:62–4. doi: 10.1093/pch/20.2.62

18. Government of United Kingdom. *The National Strategy for Autistic Children, Young People and Adults: 2021 to 2026.* (2021). p. 43. Available online at: https://assets.publishing.service.gov.uk/government/uploads/system/uploads/attachment_data/file/1004528/the-national-strategy-for-autistic-children-young-people-and-adults-2021-to-2026.pdf

19. Reasons CE, Hassan S, Bige M, Paras C, Arora S. Race and criminal justice in Canada. *Int. J. Criminal Justice Sci.* (2016) 11:26. Available online at: https://digitalcommons.cwu.edu/cgi/viewcontent.cgi?article=1720&context=cotsfac

20. Government of Canada. *Understanding the Overrepresentation of Indigenous People in the Criminal Justice System*. Available online at: https://www.justice.gc.ca/socjs-esjp/en/ind-aut/uo-cs

21. Owusu-Bempah A. Race and policing in historical context: dehumanization and the policing of Black people in the 21st century. *Theor Criminol.* (2017) 21:23–34. doi: 10.1177/1362480616677493

22. Corrado RR, Kuehn S, Margaritescu I. Policy issues regarding the overrepresentation of incarcerated aboriginal young offenders in a Canadian context. *Youth Justice.* (2014) 14:40–62. doi: 10.1177/1473225413520361

23. Government of Canada. *The Constitution Act, 1982. Schedule B to the Canada Act 1982.* (1982). c 11. Available online at: https://www.canlii.org/en/ca/laws/stat/schedule-b-to-the-canada-act-1982-uk-1982-c-11/latest/schedule-b-to-the-canada-act-1982-uk-1982-c-11.html

24. Cotter A. Experiences of discrimination among the Black and Indigenous populations in Canada, 2019. (2019) p. 14.

25. Armstrong A, Jaffray B. *Homicide in Canada, 2020.* (2020). p. 36.

26. Kingsley MF. *Indigenous People in Federal Custody Surpasses 30% Correctional Investigator Issues Statement and Challenge*. Office of the Correctional Investigator (2020). Available online at: https://www.oci-bec.gc.ca/cnt/comm/press/press20200121-eng.aspx

27. Salerno AC, Schuller RA. A mixed-methods study of police experiences of adults with autism spectrum disorder in Canada. *Int J Law Psychiatry.* (2019) 64:18–25. doi: 10.1016/j.ijlp.2019.01.002

28. Tint A, Palucka AM, Bradley E, Weiss JA, Lunsky Y. Correlates of police involvement among adolescents and adults with autism spectrum disorder. *J Autism Dev Disord.* (2017) 47:2639–47. doi: 10.1007/s10803-017-3182-5

29. Copenhaver A, Tewksbury R. Interactions between autistic individuals and law enforcement: a mixed-methods exploratory study. *Am J Crim Justice.* (2019) 44:309–33. doi: 10.1007/s12103-018-9452-8

30. Diamond LL, Hogue LB. Preparing students with disabilities and police for successful interactions. *Interv Sch Clin.* (2021) 57:3–14. doi: 10.1177/1053451221994804

31. Government of Canada. *Autism Spectrum Disorder Among Children and Youth in Canada 2018.* (2018). Available online at: https://www.canada.ca/en/public-health/services/publications/diseases-conditions/autism-spectrum-disorder-children-youth-canada-2018.html

32. Pietro N, Illes J. Closing gaps: strength-based approaches to research with aboriginal children with neurodevelopmental disorders. *Neuroethics.* (2016) 9:1–10. doi: 10.1007/s12152-016-9281-8

33. Canadian Autism Spectrum Disorder Alliance. Working together to advance a National Autism Strategy. *CASDA in the News*. (2021). Available online at: https://www.casda.ca/working-together-to-advance-a-nas/

34. *Canada PHA of. Autism: National Autism Strategy.* (2020). Available online at: https://www.canada.ca/en/public-health/services/diseases/autism-spectrum-disorder-asd/national-strategy.html

35. CASDA, KBHN. *Informing Canada's Autism Strategy: Lessons From Across the Globe*. Canadian Autism Spectrum Disorder Alliance (2020). Available online at: https://www.casda.ca/wp-content/uploads/2020/07/CASDA-KBHN-InternationalStategies-June2020.pdf

36. Government of Canada. *Minister of Health Mandate Letter*. Prime Minister of Canada (2021). Available online at: https://pm.gc.ca/en/mandate-letters/2021/12/16/minister-health-mandate-letter

37. Truth and Reconciliation Commission of Canada. *Calls to Action.* (2015). Available online at: http://www.trc.ca/

38. Committee on the Rights of Persons with Disabilities. *Concluding Observations on the Initial Report of Canada*. United Nations (2017). Available online at: http://docstore.ohchr.org/SelfServices/FilesHandler.ashx?enc=6QkG1d%2FPPRiCAqhKb7yhshFUYvCoX405cFaiGbrIbL87R7e4hNB%2FgZKnTAU8BqK7FKCyFSQGUzS4dKwSRSD%2FCPUoSzW7oP9OI5lweGr%2Br%2B7wpRzQbCN1rv%2B%2BwMd4F0fZ

39. United Nations. *Shared Framework on Leaving No One Behind: Equality and Non-Discrimination at the Heart of Sustainable Development*. Asia-Pacific Disaster Report 2015. New York, NY: UN (2016). Available online at: https://www.un-ilibrary.org/content/books/9789210576086c010

40. Botha M, Hanlon J, Williams GL. Does language matter? Identity-first versus person-first language use in autism research: a response to vivanti. *J Autism Dev Disord [Internet]*. (2021). doi: 10.1007/s10803-020-04858-w. [Epub ahead of print].

41. Annamma SA, Connor D, Ferri B. Dis/ability critical race studies (DisCrit): theorizing at the intersections of race and dis/ability. *Race Ethn Educ.* (2013) 16:1–31. doi: 10.1080/13613324.2012.730511

42. Lane J. *Black Peoples, Indigenous Peoples, People(s) of Colour (BIPOC): Inclusive and Antiracist Writing*. Simon Fraser University (2021). Available online at: https://www.lib.sfu.ca/about/branches-depts/slc/writing/inclusive-antiracist-writing/bipoc

43. Deo ME. *Why BIPOC Fails*. Va Law Rev (2021). p. 107. Available online at: https://www.virginialawreview.org/articles/why-bipoc-fails/

44. City of Ottawa. *Equity and Inclusion Lens Snapshot - Racialized People.* (2016). p. 20. Available online at: https://documents.ottawa.ca/sites/documents/files/racializd_ss_en.pdf

45. Nicol J, Osazuwa B. *Race and Ethnicity: Evolving Terminology*. Library of Parliament. Social Affairs and Population (2022). Available online at: https://hillnotes.ca/2022/01/31/race-and-ethnicity-evolving-terminology/

46. Else-Quest NM, Hyde JS. Intersectionality in Quantitative Psychological Research: I. Theoretical and epistemological issues. *Psychol Women Q.* (2016) 40:155–70. doi: 10.1177/0361684316629797

47. Namugenyi C, Nimmagadda SL, Reiners T. Design of a SWOT analysis model and its evaluation in diverse digital business ecosystem contexts. *Procedia Comput Sci.* (2019) 159:1145–54. doi: 10.1016/j.procs.2019.09.283

48. University of Alberta Library. *Health Sciences Search Filters: Indigenous Peoples*. (2022). Available online at: https://guides.library.ualberta.ca/health-sciences-search-filters/indigenous-peoples

49. University of Prince Edward Island Robertson Library. *Indigenous Peoples Search Terms*. Available online at: https://library.upei.ca/indigenous-search-terms

50. University of Toronto Libraries. *Indigenous Health Resources: Databases*. (2022). Available online at: https://guides.library.utoronto.ca/c.php?g=250466&p=5037464

51. Page MJ, McKenzie JE, Bossuyt PM, Boutron I, Hoffmann TC, Mulrow CD, et al. The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. *BMJ.* (2021) 372:n71. doi: 10.1136/bmj.n71

52. Lindblom A. Exploring autism and music interventions through a First Nations lens. *Altern Int J Indig Peoples.* (2017) 13:202–9. doi: 10.1177/1177180117729854

53. Lindblom A. 'It gives them a place to be proud' - Music and social inclusion. Two diverse cases of young First Nations people diagnosed with autism in British Columbia, Canada. *Psychol Music.* (2017) 45:268–82. doi: 10.1177/0305735616659553

54. Gerlach A. "Circle of caring": a first nations worldview of child rearing. *Can J Occup Ther*. (2008) 75:18–25. doi: 10.1177/000841740807500107

55. Simmons. Words have power: (re)-defining serious emotional disturbance for American Indian and Alaska native children and their families. *Am Indian Alsk Native Ment Health Res.* (2004) 11:59–64. doi: 10.5820/aian.1102.2004.59

56. Rossow-Kimball B, Lavis M, Blackhurst M. "I can find my own Elder!" Cultural engagement as serious leisure for Aboriginal adults living in non-Aboriginal group homes. *Leis Stud.* (2017) 36:244–55. doi: 10.1080/02614367.2015.1085592

57. Vargas T. I have to do what I have to do. *The Washington Post.* (2010). Available online at: https://web-s-ebscohost-com.libaccess.lib.mcmaster.ca/ehost/detail/detail?vid=0&sid=666bebf4-d25d-4796-8357-bb92cac77971%40redis&bdata=JnNpdGU9ZWhvc3QtbGl2ZSZzY29wZT1zaXRl#AN=WPT207297757210&db=bwh

58. Carley M. Autism-Shmautism: in the end, Darius McCollum was Poor and Black. *TCA EP World LLC.* (2018) 48:34–8. Available online at: https://www.linkedin.com/pulse/autism-schmautism-end-darius-mccollum-poor-black-michael-john-carley

59. Solomon O, Lawlor MC. "And I look down and he is gone": narrating autism, elopement and wandering in Los Angeles. *Soc Sci Med.* (2013) 94:106–14. doi: 10.1016/j.socscimed.2013.06.034

60. Davenport MA, Romero ME, Lewis CD, Lawson T, Ferguson B, Stichter J, et al. An initial development and evaluation of a culturally responsive police interactions training for black adolescents with autism spectrum disorder. *J Autism Dev Disord.* (2021). doi: 10.1007/s10803-021-05181-8. [Epub ahead of print].

61. Lisa Stevenson. *Life Beside Itself: Imagining Care in the Canadian Arctic*. University of California Press (2014). Available online at: https://www.ucpress.edu/book.php?isbn=9780520282940

62. O'Gorman M. Mental and physical health impacts of water/sanitation infrastructure in First Nations communities in Canada: an analysis of the Regional Health Survey. *World Dev.* (2021) 145:105517. doi: 10.1016/j.worlddev.2021.105517

63. Inman CE. Absence and epidemic: autism and fetal alcohol spectrum disorder in indigenous populations in Canada. *Can J Disabil Stud.* (2019) 8:227–61. doi: 10.15353/cjds.v8i4.531

frontiers | Frontiers in Psychiatry

TYPE Original Research
PUBLISHED 02 November 2022
DOI 10.3389/fpsyt.2022.929833

OPEN ACCESS

EDITED BY
Diana Schendel,
Drexel University, United States

REVIEWED BY
Caihong Sun,
Harbin Medical University, China
Xiaohua Liang,
Children's Hospital of Chongqing
Medical University, China
Michele Roccella,
University of Palermo, Italy

*CORRESPONDENCE
Hao Zhou
haoye320@163.com
Xiao Hu
18096105119@163.com

†These authors have contributed equally to this work

SPECIALTY SECTION
This article was submitted to Autism,
a section of the journal Frontiers in Psychiatry

RECEIVED 27 April 2022
ACCEPTED 10 October 2022
PUBLISHED 02 November 2022

CITATION
Liu Y, Zhou F, Qin J, Lin Y, Li T, Zhu C, Long F, Wang X, Hu X and Zhou H (2022) The intervention dilemma and high burden of children with autism in Guizhou province, Southwest China.
Front. Psychiatry 13:929833.
doi: 10.3389/fpsyt.2022.929833

The intervention dilemma and high burden of children with autism in Guizhou province, Southwest China

Ye Liu[1]†, Fang Zhou[2]†, Jixuan Qin[3], Yong Lin[4], Tonghuan Li[5], Chengyong Zhu[6], Fang Long[7], Xike Wang[2], Xiao Hu[8]* and Hao Zhou[2]*

[1]Department of Otolaryngology, Guizhou Provincial People's Hospital, Medical College of Guizhou University, Guiyang, China, [2]Department of Developmental Behavioral Pediatrics, Guizhou Provincial People's Hospital, Medical College of Guizhou University, Guiyang, China, [3]Department of Pediatric Rehabilitation, Anshun Women's & Children's Hospital, Anshun, China, [4]Department of Pediatric Rehabilitation, The First People's Hospital of Zunyi, Zunyi, China, [5]Department of Pediatric Rehabilitation, Affiliated Hospital of Zunyi Medical University, Zunyi, China, [6]Department of Pediatric Rehabilitation, The Third Affiliated Hospital of Guizhou Medical University, Duyun, China, [7]Department of Healthcare, Liupanshui Women & Children Hospital, Liupanshui, China, [8]Department of Neurology, Guizhou Provincial People's Hospital, Guiyang, China

Background: Autism spectrum disorder (ASD) is a highly disabling neurodevelopmental disorder, and the burden is high. Data on the burden of ASD are limited in China, especially in the southwest. Therefore, the aims of this study were to investigate the intervention status and burden of children with ASD in Southwest China.

Materials and methods: Families of children with ASD were recruited from hospitals, special education schools, and private rehabilitation centers; they participated in the survey and completed the questionnaire. Descriptive analysis was conducted on the questionnaire results, which included basic demographic characteristics, rehabilitation status, and burden. Multivariate analysis was used to analyze the association of basic family demographic characteristics, rehabilitation status, and costs of ASD.

Results: A total of 231 families of children with ASD participated in this survey, and 78.35% (181/231) of the children with ASD were male. The mean age was 4.34 ± 2.09 years. A total of 55.84% (129/231) of the children with ASD had an intellectual disability. Only 46.32% (107/231) started receiving intervention within 1 month after diagnosis. The institutions for rehabilitation interventions for children with ASD were mainly tertiary hospitals (39.39%), special education schools (29.87%) and private rehabilitation institutions (21.64%). For a total of 42.86% (99/231) of the children with ASD, the duration of the intervention was less than 10 h per week. A total of 74.89% (173/231) of the children with ASD received a rehabilitation intervention at home. A total of 66.67% of the parents were satisfied with the treatment. The monthly cost of medical intervention for the patients of children with autism was 7,225 ± 474 RMB ($1,134 ± 74), and the non-medical intervention cost was 2,133 ± 107 RMB ($334 ± 17). The annual burden of patients with autism was 86,700 ± 5,688

RMB ($13,596 ± 892). The estimated total annual burden of ASD was 5.548 billion RMB ($870 million) in Guizhou province.

Conclusion: The results revealed that rehabilitation resources are limited and that the burden of ASD is high in Guizhou province; therefore, improving the rehabilitation status and easing the burden of children with ASD is urgent in these regions.

KEYWORDS

autism, rehabilitation status, disease burden, questionnaire, China

Introduction

Autism spectrum disorder (ASD) is a group of highly heterogeneous neurodevelopmental disorders (1). ASD has been associated with significant social functional impairments and lasts a lifetime, and it can severely impact the quality of life of patients with ASD and their families (2). The number of individuals diagnosed with ASD has obviously increased in recent decades. The prevalence of ASD was 1/44 in the U.S. and our previous national epidemiological study showed that the prevalence of ASD was 0.7% in the Chinese child population (3, 4). According to preliminary estimates, there are at least millions of patients with ASD, and an epidemiological survey revealed that ASD is the main cause of disability in children under the age of 6 in China (5, 6). Accurate estimates of the economic effect of ASD are very important and urgently needed to illustrate the responsibilities of the families of children with ASD and society.

The burden of ASD is very high, and the total burden includes direct medical costs (medical checkup costs, rehabilitation costs, etc.), and indirect costs (family care, education, living expenses, etc.) (7); however, the overall and economic effects of ASD are not well-established worldwide. A previous study indicated that the lifespan costs of individuals with ASD with or without intellectual disability were $2.4 and $1.4 million in the United States and $2.2 and $1.4 million in the United Kingdom, respectively (8). Hong et al. reported that the economic burden of ASD was estimated to be $2,700,596 in 2008 and $9,645,503 in 2015 in South Korea (9). Another study showed that ASD placed a higher burden on caregivers compared with other mental illnesses, such as schizophrenia (10). Currently, the data on ASD burden mainly come from developed countries, while such data are rare in developing countries. The limited data demonstrated that the burden of children with autism was 19,582.4 RMB ($3,704) per year in Beijing City, China (11). Another study showed that the average loss of annual income associated with having a child with ASD was 44,077 RMB ($7,226) in Changsha City, China (12). Medical resources and economic levels are uneven in China (13), and Beijing and Changsha are developed cities. Our previous research found low levels of awareness and knowledge of ASD among child health care workers in Guizhou province in China (14), which is an underdeveloped region in terms of medical resources and economic level. Therefore, investigating the rehabilitation status and burden of children with ASD is urgent in these regions, and the aim of this study was to focus on this issue.

Materials and methods

Study sites and participants

The study was conducted from March 2022 to April 2022 in Guizhou province, China, which is located at 24°37'–29°13'N, 103°36'–109°35'E. The resident population of Guizhou in 2021 was 38,562,100 according to the 7th National Census,[1] and the GDP was ranked 22 among the 31 provinces in mainland China, which is a relatively low level within China. Medical resources, especially rehabilitation resources for ASD, are insufficient in Guizhou province, and only a few hospitals have rehabilitation centers for children or developmental behavioral pediatrics units. Thus, families of children with ASD were recruited from six rehabilitation centers for children (Guizhou Provincial People's Hospital, Anshun Women's & Children's Hospital, the First People's Hospital of Zunyi, the Affiliated Hospital of Zunyi Medical University, and Liupanshui Women's & Children's Hospital), two special educational institutions (Shanchen and Huanlechuan Special Educational Institutions, which are representative special educational institutions for children with ASD in Guizhou province), and a community hospital (Shuikoushi Community Hospital, which is the only community hospital with a training center for ASD in Guizhou province).

---

[1] http://www.stats.gov.cn/tjsj/pcsj/rkpc/d7c/202111/P020211126523667366751.pdf

Questionnaire

The information was collected by using our self-designed questionnaire. The multidisciplinary team for questionnaire development was composed of clinical experts with rich experience in ASD, epidemiologists, special education teachers, and public health experts. All items of the questionnaire were original, and the questions were arranged in the questionnaire according to the expert group's recommendations. After designing the questionnaire, we conducted a pilot study to evaluate the content validity of the questionnaire and invited 30 families of children with ASD from Guizhou Provincial People's Hospital to complete the questionnaire. All feedback indicated that the questions were easy to understand in the Chinese cultural context.

The questionnaire consisted of three parts. The first part collected the basic demographic characteristics of the families of children with ASD, including the child's age at the time of the study and sex; parents' education levels, occupations, and economic income; parents' understanding of their child's developmental age according to his or her abnormal developmental trajectory; age of the child at the time of ASD diagnosis; and whether the child had an intellectual disability (note: the cognitive level was based on the results of cognitive tests such as the Gesell assessment). The second part contained nine items to assess rehabilitation training status, including the interval from diagnosis to receiving rehabilitation training, where a patient received rehabilitation training, whether the parents received family intervention training, whether the patient received a family intervention, and the employment situation of the parents. The third part contained six questions that evaluated the burden of ASD, including monthly out-of-pocket payments for rehabilitation training, disability benefits, other non-medical expenses (the costs of accommodations, meals, transportation), the share of rehabilitation training costs in the household income, and whether rehabilitation training had to be given up due to high health care costs. All questions were multiple choice and included options for "Yes" and "No."

Data collection

WeChat (Tencent Corp) is the most popular social software used in China. The questionnaires were developed and sent through the WeChat network by doctors and special education teachers using Sojump (Changsha ran Xing InfoTech Ltd., Changsha, China), which is a professional online survey, evaluation and polling platform that provides personalized services, including questionnaire design and data collection. The families of children with ASD used a mobile phone to scan a QR code, and there was no time limit to complete the questionnaire. If participants had any questions, professional staff answered them. All participants voluntarily and anonymously completed the questionnaires and provided oral informed consent. Questionnaires with incomplete basic information and unchecked questions were excluded.

Data analysis

Data analysis was performed by utilizing SPSS 26.0 (SPSS Inc., Chicago, IL). Enumeration data are described as the mean ± SD, and categorical variables are presented as percentages. The basic information, the status of training, and the costs were analyzed by descriptive analysis. The total annual burden of ASD was estimated based on the total number of children with ASD and the average cost per person every year in Guizhou province. The total number of children with ASD in Guizhou province was calculated according to an ASD prevalence of 0.7% in the Chinese population and a total of 9,200,000 children aged 0–14 years (contents refer to the 7th National Census (see text footnote 1) in Guizhou province. A total of approximately 64,000 (0.007 × 9.2 million) children had been diagnosed with ASD. The effects of the different variables on the therapeutic effect and costs were compared by the chi-square test, and the Friedman test was conducted to explore the contributions of the variables to the therapeutic effect and costs. All tests were two-tailed, and a P-value of less than 0.05 was considered statistically significant.

Results

Basic information of participants

A total of 231 families of children with ASD participated in this survey; 78.35% (181/231) of the children with ASD were male, and 21.65% (50/231) were female. The mean age of the children with ASD was 4.34 ± 2.09 years. The parents reported that the mean developmental age of their children with abnormal developmental trajectories was 2.11 ± 1.09, and the mean age at the time of ASD diagnosis was 2.58 ± 1.35. A total of 51.51% (119/231) of the families earned less than 5,000 RMB ($786) per month. The main caregivers in rehabilitation training for the children were their mothers. A total of 55.84% (129/231) of the parents reported that their child with ASD had an intellectual disability (**Table 1**).

Intervention status of children with autism spectrum disorder

Among the children with ASD who participated in the questionnaire, only 46.32% (107/231) started an intervention within 1 month after diagnosis. The institutions for rehabilitation interventions for the children with ASD were

TABLE 1 Basic information of respondents (*n*, %).

Characteristics	Total (*n*)	Ratio (%)
Number	231	
Age (years, mean ± mean)		
Present age	4.34 ± 2.09	
Children with abnormal developmental trajectories	2.11 ± 1.09	
At diagnosis with ASD	2.58 ± 1.35	
Sex		
Male	181	78.35
Female	50	21.65
Father's career		
Professional	53	22.94
Civil servant	19	8.22
Farmer	42	18.18
Freelancer	117	50.66
Mother's career		
Professional	50	21.65
Civil servant	10	4.32
Farmer	50	21.65
Freelancer	121	52.38
Father's level of education		
Junior	156	67.53
Undergraduate	64	27.71
Graduate	11	4.76
Mother's level of education		
Junior	152	65.80
Undergraduate	73	31.60
Graduate	6	2.60
Monthly income		
<5,000 RMB ($786)	119	51.51
5,000–10,000 RMB ($786–1,572)	94	40.69
>10,000 RMB ($1,572)	18	7.80
Caregiver in rehabilitation training		
Father	25	10.82
Mother	145	62.77
Grandparent	59	25.54
Nanny	2	0.87
Developmental delay		
Yes	129	55.84
No	102	44.16

mainly tertiary hospitals (39.39%), special education schools (29.87%) and private rehabilitation institutions (21.64%). A total of 42.86% (99/231) of the children with ASD had an intervention duration of less than 10 h per week, and only 9.53% (22/231) had an intervention duration of more than 30 h per week. More than 50% of the children with ASD received family intervention training, and 74.89% (173/231) received rehabilitation intervention at home. A total of 22.08% of the children with ASD received other non-behavioral intervention therapies, such as acupuncture, mouse nerve growth factor treatment, and hyperbaric oxygen treatment. A total of 66.67% of the parents were satisfied with the treatment, and almost two-thirds of the families of children with ASD chose to resign from their jobs after their children were diagnosed with ASD (Table 2).

Costs for families of children with autism spectrum disorder

The monthly cost of medical intervention for patients with autism was 7,225 ± 474 RMB ($1,134 ± 74), of which the self-funded portion was 3,679 ± 466 RMB ($577 ± 73), and the subsidy of the China Disabled Persons' Federation (CDPF) was

TABLE 2 The rehabilitation intervention status of children with autism spectrum disorder (ASD) in Guizhou province (*n*, %).

Characteristics	Total	Ratio (%)
Number	231	
Interval from diagnosis to receiving intervention		
<1 month	107	46.32
1–3 months	52	22.51
3–6 months	30	12.99
>6 months	42	18.18
Intervention site		
Tertiary hospital	91	39.39
Special education school	69	29.87
Private rehabilitation institution	50	21.64
Community hospital	21	9.09
Total intervention time per week		
<10 h	99	42.86
10–19 h	75	32.46
20–29 h	35	15.15
>30 h	22	9.53
Receiving family intervention training		
Yes	126	54.55
No	105	45.45
Family intervention program		
Yes	173	74.89
No	58	25.11
Treatment beyond intervention		
Yes	51	22.08
No	180	77.92
Treatment effect		
Satisfied	154	66.67
Unsatisfied	77	33.33
Parent resignation from employment		
Father	17	7.36
Mother	104	45.02
Both	12	5.20
None	98	42.42

1,321 ± 82 RMB ($207 ± 13). Notably, more than 70% of the families were aware of the CDPF subsidy policy and received funding. The non-medical intervention cost was 2,133 ± 107 RMB ($334 ± 17). The annual burden of patients with autism was 86,700 ± 5,688 RMB ($13,596 ± 892). Approximately 30% of the parents thought that the burden accounted for 70% of their family income, and 37.66% of them had planned to give up treatment because of the heavy burden.

The total annual burden of ASD was estimated based on the total number of children with ASD and the average cost per person every year in Guizhou province. The total expenses were 5.548 billion RMB (64,000 × 86,700 RMB, equivalent to US $870 million), the total annual medical expenses were 3.9 billion RMB (64,000× 7,225 × 12 RMB, equivalent to US $610 million), and the total annual non-medical expenses were 1.657 billion RMB (64,000 × 2,133 × 12 RMB, equivalent to US $260 million) in Guizhou province.

Impacts of the basic demographic characteristics on training and costs of autism spectrum disorder

There were significant differences in the choice of rehabilitation institutions among parents of different occupations; for example, farming families preferred to choose special education schools for their children's ASD training, and professional and civil servants always preferred tertiary hospitals or private rehabilitation institutions (**Figures 1A,B**). There was no significant difference among children with ASD in the interval from diagnosis to receiving intervention, the total duration of the intervention per week, or the satisfaction with the intervention effect by parents' occupation (**Figures 1C–H**).

From **Figures 2A–C** we can see that the intervention effect of children with ASD was associated with the total intervention duration per week, and families with 10–19 h of intervention had the highest satisfaction with the treatment effect ($P < 0.05$). There were significant differences in satisfaction with the intervention treatment for children with ASD among mothers with different educational backgrounds. Mothers with a junior college degree or below had the highest satisfaction ($P < 0.05$). Other factors, such as paternal educational background, parents' occupations, monthly family income, whether there was a developmental lag, whether children received treatment other than the rehabilitation intervention, and whether family intervention was carried out, had no significant impact on satisfaction with the intervention effect for children with ASD.

The weekly intervention duration for children with ASD was significantly different among the different rehabilitation sites ($P < 0.05$), with durations of more than 30 h for private rehabilitation institutions and special schools and less than 10 h for public hospitals (**Figure 3**).

The DPF subsidy for children with ASD varied significantly among different rehabilitation treatment sites. The highest subsidy was 1,863 ± 110 RMB and 1,825 ± 245 RMB per month in tertiary hospitals and special education schools, respectively, and the lowest was in community rehabilitation institutions (**Figure 4A**). There was no significant difference between the costs and parents' occupations, the total duration of the intervention per week, the place where intervention treatment was received and whether children received other treatments (**Figures 4B–D**).

Discussion

Autism spectrum disorder is the most common highly disabling neurodevelopmental disorder in childhood and has a high burden. The exact costs of ASD worldwide are still unclear. In this study, we first described the intervention status and burden of children with ASD in Guizhou province, Southwest China. The cross-sectional survey revealed that only 46.32% of the children with ASD started an intervention within 1 month after diagnosis. The institutions for rehabilitation interventions for children with ASD were mainly tertiary hospitals, special education schools and private rehabilitation institutions. For almost half of the children with ASD, the duration of intervention was less than 10 h per week. A total of 74.89% of the children with ASD received a rehabilitation intervention at home. Only two-thirds of the parents were satisfied with the treatment. In families of children with ASD with a high burden, the annual burden of patients with autism could reach as high as 86,700 ± 5,688 RMB ($13,596 ± 892).

Intervention status of autism spectrum disorder

The results of this survey showed that the parents realized that the mean developmental age of their children with abnormal developmental trajectories was 2 years, and the mean age at the time of ASD diagnosis was 2.5 years. Only half of the children with ASD started an intervention within 1 month after diagnosis, indicating that many children with ASD received interventions around the age of 3 years. This seems to be a challenge for ASD prognosis, and a previous study demonstrated that early screening and intervention were extremely important for ASD prognosis (15). Early ASD screening networks have been established in many developed cities in China, such as Shanghai, Tianjin, and Guangzhou (16–18). Medical resources and economic levels are unevenly distributed in China (13). Guizhou province is located in Southwest China, which is an underdeveloped region in terms of medical resources and economic level. Early screening of ASD is limited, and our previous research found low levels of awareness

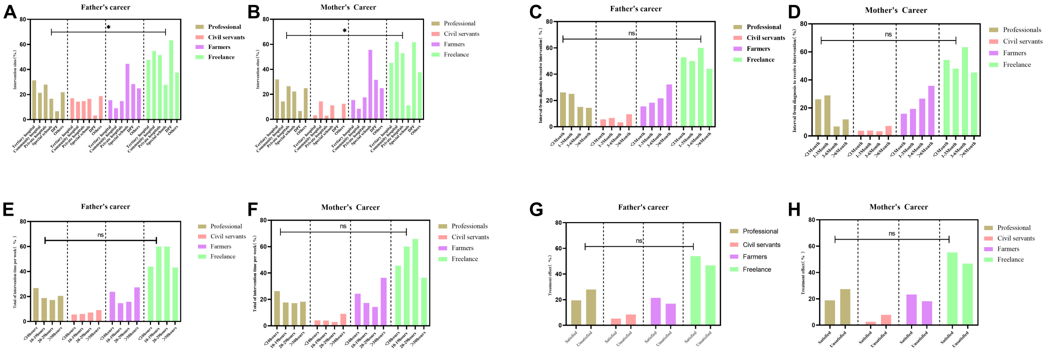


FIGURE 1
Influence of parental occupation on the intervention status of children with autism spectrum disorder (ASD), **(A,B)** for the choice of rehabilitation institutions, **(C,D)** for the interval from diagnosis to receiving intervention, **(E,F)** for the total duration of the intervention per week, **(G,H)** for satisfaction with the intervention effect. *$p < 0.05$; ns, $p > 0.05$.

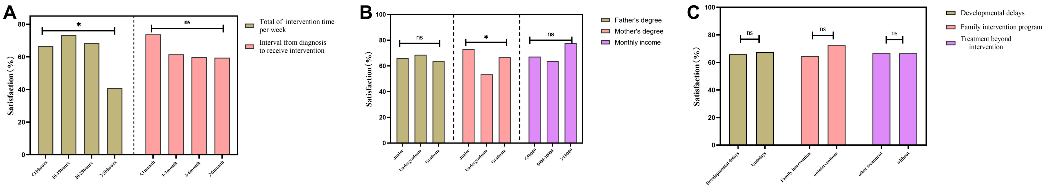


FIGURE 2
Analysis of influencing factors of satisfaction with intervention treatment for children with Autism spectrum disorder (ASD). **(A)** For total intervention duration per week and interval from diagnosis to receiving intervention, **(B)** for parents' degree and monthly income, and **(C)** for developmental delays, family intervention program, and treatment beyond intervention. *$p < 0.05$; ns, $p > 0.05$.

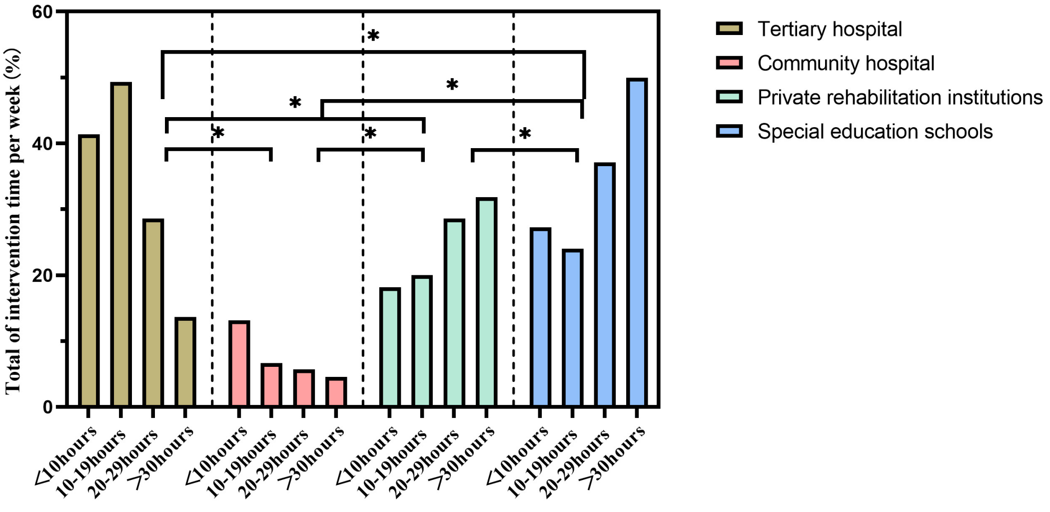


FIGURE 3
Influence of rehabilitation location on the total weekly intervention duration for children with Autism spectrum disorder (ASD). *$p < 0.05$; ns, $p > 0.05$.

and knowledge of ASD among child health care workers in Guizhou province (14). Thus, there is an urgent need to establish the early screening network of ASD in these less developed areas, in China.

In addition to the delay in the age of ASD screening and intervention, the results of this study revealed that ASD public medical resources are insufficient in Guizhou province. Half of the families of children with ASD chose special

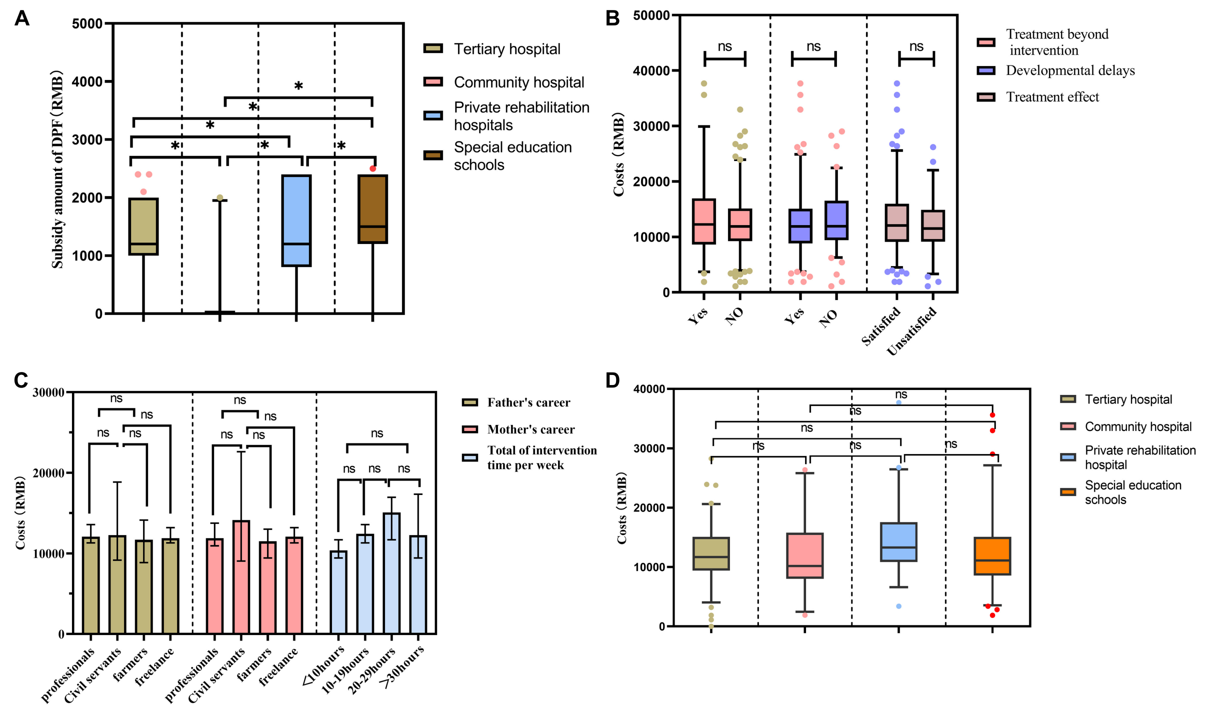


FIGURE 4
Impacts of different factors on rehabilitation costs. *$p < 0.05$; ns, $p > 0.05$.

education schools and private rehabilitation institutions to conduct interventions. A previous investigation showed that ASD interventions mainly occur in private institutions in China (19). It is interesting to note that parents' occupations impact the choice of rehabilitation institutions, and the main reason is that family income may affect the choice of rehabilitation institutions and family income varies across different occupations. To address this dilemma of a medically underserved population, in recent years, experts have called for enhanced family intervention for children with ASD, and the family may play a key role in ASD intervention and provide a clear benefit for ASD prognosis (20–22). In this study, a total of 74.89% of the children with ASD received rehabilitation interventions at home, which revealed that family intervention has become increasingly valued. However, while many children with ASD received family intervention, for almost half of the children with ASD, the duration of the intervention was less than 10 h per week, especially for those receiving interventions in public hospitals. This may be related to medical students unwillingly to be occupied in pediatrics in China (23, 24) and hospitals being short-staffed for the provision of ASD interventions. Of concern, 22.08% of the children with ASD received other non-behavioral intervention therapies, such as acupuncture, mouse nerve growth factor treatment, and hyperbaric oxygen treatment; however, these therapies lack evidence-based recommendations.

Costs of autism spectrum disorder

The results of this study showed that the annual burden of patients with autism could be as high as 86,700 ± 5,688 RMB ($13,596 ± 892) in Guizhou province, China. In 2011, Xiong et al. reported that the burden of children with autism was 19,582.4 RMB ($3,704) per year in Beijing City, China (11). In 2015, Ou et al. conducted a survey and revealed that the average loss of annual income associated with having a child with ASD was 44,077 RMB ($7,226) in Changsha City, China (12). We can see from the current study's data that the cost of ASD may have increased in China over time, which is similar to a previous study showing that the economic burden of ASD in 2015 was quadruple that in 2008 in South Korea (9). Seventy percent of the families of children with ASD received DPF funding in our study; however, ASD costs were mainly self-funded. The burden accounted for 70% of the families' income, and approximately one-third of the families planned to give up treatment because of the heavy burden. The results indicated that the burden of ASD is very high for families of children with ASD and the government in China and that the insurance system for ASD needs to be further improved. The results illustrated that the highest DPF subsidy was for special education schools, which can explain why farming families preferred to choose special education schools for their children's ASD training. It is worth noting that the burden of ASD comorbid with other diseases was obviously increased in a previous study (8); however, whether the children had developmental delays was not related to the costs of ASD in our study. It could be that the respondents were younger in this study, and other costs, such as educational expenses, had not been incurred.

Limitations

This is the first study to describe the intervention status and burden of children with ASD in Guizhou province, Southwest China. However, there are two limitations in this survey. First, the sample size was relatively small, and the survey was conducted mainly in cities, making some of the results prone to bias. Second, the respondents were younger in this study, and other costs had not been incurred, so the actual burden may have been underestimated here. Therefore, a multicenter, multidimensional survey is urgently needed in future studies.

Conclusion

In conclusion, this is the first study to describe the intervention status and burden of children with ASD in an underdeveloped city in China. The results revealed that the rehabilitation resources were limited and that the burden of ASD was high in Guizhou province; therefore, improving the rehabilitation status and easing the burden of children with ASD is urgent in these regions.

Data availability statement

The original contributions presented in this study are included in the article/supplementary material, further inquiries can be directed to the corresponding authors.

Ethics statement

The studies involving human participants were reviewed and approved by the Guizhou Provincial People's Hospital. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

Author contributions

HZ and XH conceived the study. YeL and FZ contributed to the analysis, synthesis, and interpretation of the results and wrote the manuscript. JQ, YoL, TL, CZ, and FL contributed to the data collection. All authors contributed to the preparation of the manuscript.

Funding

This project was supported by the Foundation of Guizhou Science and Technology Agency (LC[2021]018).

Acknowledgments

We thank all the families of children with ASD who participated in this study, as well as Jing Li (Shanchen Special Educational Institution), Yuanmei Zhang (Huanlechuan Special Educational Institution), and Minhao Wang, Chongnan Zhang, and Xinyu Zhang (Shuikoushi Community Hospital), who contributed to the collection of data.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

References

1. Lord C, Elsabbagh M, Baird G, Veenstra-Vanderweele J. Autism spectrum disorder. *Lancet.* (2018) 392:508–20. doi: 10.1016/S0140-6736(18)31129-2

2. Vohra R, Madhavan S, Sambamoorthi U, St PC. Access to services, quality of care, and family impact for children with autism, other developmental disabilities, and other mental health conditions. *Autism.* (2014) 18:815–26. doi: 10.1177/1362361313512902

3. Xu G, Strathearn L, Liu B, Bao W. corrected prevalence of autism spectrum disorder among US children and adolescents. *JAMA.* (2018) 319:505.

4. Zhou H, Xu X, Yan W, Zou X, Wu L, Luo X, et al. Prevalence of autism spectrum disorder in China: A nationwide multi-center population-based study among children aged 6 to 12 years. *Neurosci Bull.* (2020) 36:961–71. doi: 10.1007/s12264-020-00530-6

5. Li N, Chen G, Song X, Du W, Zheng X. Prevalence of autism-caused disability among Chinese children: A national population-based survey. *Epilepsy Behav.* (2011) 22:786–9. doi: 10.1016/j.yebeh.2011.10.002

6. Wang F, Lu L, Wang SB, Zhang L, Ng CH, Ungvari GS, et al. The prevalence of autism spectrum disorders in China: A comprehensive meta-analysis. *Int J Biol Sci.* (2018) 14:717–25. doi: 10.7150/ijbs.24063

7. Gbd 2019 Mental Disorders Collaborators. Global, regional, and national burden of 12 mental disorders in 204 countries and territories, 1990-2019: A systematic analysis for the global burden of disease study 2019. *Lancet Psychiatry.* (2022) 9:137–50. doi: 10.1016/S2215-0366(21)00395-3

8. Buescher AV, Cidav Z, Knapp M, Mandell DS. Costs of autism spectrum disorders in the United Kingdom and the United States. *JAMA Pediatr.* (2014) 168:721–8. doi: 10.1001/jamapediatrics.2014.210

9. Hong M, Lee SM, Park S, Yoon SJ, Kim YE, Oh IH. Prevalence and economic burden of autism spectrum disorder in South Korea using national health insurance data from 2008 to 2015. *J Autism Dev Disord.* (2020) 50:333–9. doi: 10.1007/s10803-019-04255-y

10. Yildiz M, Demir Y, Kircali A, Incedere A. Caregiver burden in schizophrenia and autism spectrum disorders: A comparative study. *Psychiatry Investig.* (2021) 18:1180–7. doi: 10.30773/pi.2021.0165

11. Xiong N, Yang L, Yu Y, Hou J, Li J, Li Y, et al. Investigation of raising burden of children with autism, physical disability and mental disability in China. *Res Dev Disabil.* (2011) 32:306–11. doi: 10.1016/j.ridd.2010.10.003

12. Ou JJ, Shi LJ, Xun GL, Chen C, Wu RR, Luo XR, et al. Employment and financial burden of families with preschool children diagnosed with autism spectrum disorders in urban China: Results from a descriptive study. *BMC Psychiatry.* (2015) 15:3. doi: 10.1186/s12888-015-0382-4

13. Zhang Y, Coyte PC. Inequality of opportunity in healthcare expenditures: Evidence from China. *BMC Health Serv Res.* (2020) 20:379. doi: 10.1186/s12913-020-05252-z

14. Ma Y, Zhou Y, Liu Y, Ping Y, Wang Y, Hu X, et al. Urgency in improving child health care workers' awareness and knowledge of ASD: Findings from a cross-sectional study in Southwest China. *Front Psychiatry.* (2021) 12:703609. doi: 10.3389/fpsyt.2021.703609

15. Elder JH, Kreider CM, Brasher SN, Ansell M. Clinical impact of early diagnosis of autism on the prognosis and parent-child relationships. *Psychol Res Behav Manag.* (2017) 10:283–92. doi: 10.2147/PRBM.S117499

16. Wang K, Qi Q, Luo Z, Zhou J, Chen S, Wang L. Autism spectrum disorder: Status of primary care in China. *Biosci Trends.* (2022) 16:172–5. doi: 10.5582/bst.2022.01093

17. Li C, Zhu G, Feng J, Xu Q, Zhou Z, Zhou B, et al. Improving the early screening procedure for autism spectrum disorder in young children: Experience from a community-based model in shanghai. *Autism Res.* (2018) 11:1206–1217. doi: 10.1002/aur.1984

18. Huang JP, Cui SS, Han Y, Irva HP, Qi LH, Zhang X. Prevalence and early signs of autism spectrum disorder (ASD) among 18-36 month-old children of Tianjin in China. *Biomed Environ Sci.* (2014) 27:453–61. doi: 10.3967/bes2014.008

19. Zhou WZ, Ye AY, Sun ZK, Tian HH, Pu TZ, Wu YY, et al. Statistical analysis of twenty years (1993 to 2012) of data from mainland China's first intervention center for children with autism spectrum disorder. *Mol Autism.* (2014) 5:52. doi: 10.1186/2040-2392-5-52

20. Althoff CE, Dammann CP, Hope SJ, Ausderau KK. Parent-mediated interventions for children with autism spectrum disorder: A systematic review. *Am J Occup Ther.* (2019) 73:7303205010p1–7303205010p13. doi: 10.5014/ajot.2019.030015

21. Green J, Charman T, McConachie H, Aldred C, Slonims V, Howlin P, et al. Parent-mediated communication-focused treatment in children with autism (PACT): A randomised controlled trial. *Lancet.* (2010) 375:2152–60. doi: 10.1016/S0140-6736(10)60587-9

22. Akhani A, Dehghani M, Gharraee B, Hakim SM. Parent training intervention for autism symptoms, functional emotional development, and parental stress in children with autism disorder: A randomized clinical trial. *Asian J Psychiatr.* (2021) 62:102735. doi: 10.1016/j.ajp.2021.102735

23. Zhang Y, Huang L, Zhou X, Zhang X, Ke Z, Wang Z, et al. Characteristics and workload of pediatricians in China. *Pediatrics.* (2019) 144:e20183532. doi: 10.1542/peds.2018-3532

24. Liu Y, Yang LL, Xu SY, Zhao ZY. Pediatrics in China: Challenges and prospects. *World J Pediatr.* (2018) 14:1–3. doi: 10.1007/s12519-017-0117-3

frontiers | Frontiers in Psychiatry

TYPE Mini Review
PUBLISHED 21 December 2022
DOI 10.3389/fpsyt.2022.916532

OPEN ACCESS

EDITED BY
Diana Schendel,
Drexel University, United States

REVIEWED BY
Jessica Brian,
Holland Bloorview Kids Rehabilitation Hospital, Canada
Matthew O'Brien,
University of Iowa Stead Family Children's Hospital, United States

*CORRESPONDENCE
Laura Pacione
laura.pacione@utoronto.ca

SPECIALTY SECTION
This article was submitted to Autism,
a section of the journal
Frontiers in Psychiatry

RECEIVED 09 April 2022
ACCEPTED 23 November 2022
PUBLISHED 21 December 2022

CITATION
Pacione L (2022) Telehealth-delivered caregiver training for autism: Recent innovations.
Front. Psychiatry 13:916532.
doi: 10.3389/fpsyt.2022.916532

Telehealth-delivered caregiver training for autism: Recent innovations

Laura Pacione[1,2]*

[1]Division of Child and Youth Mental Health, Department of Psychiatry, University of Toronto, Toronto, ON, Canada, [2]Department of Mental Health and Substance Use, World Health Organization, Geneva, Switzerland

Providing treatment to children with autism is a global health priority, and research demonstrates that caregivers can be trained in techniques to promote their child's social interaction, communication, play, positive behavior and skills. These caregiver-mediated interventions have been shown to promote a number of positive outcomes in children with autism, as well as their caregivers. When provided by telehealth, data indicate that caregiver training is acceptable and feasible, and associated with similar positive outcomes as live face-to-face training. Telehealth innovations, which have accelerated during the COVID-19 era, have demonstrated advantages over in-person delivery of services in terms of cost effectiveness and increased accessibility, however, more research is needed on feasibility, acceptability and effectiveness for different populations in different contexts. This brief review will highlight recent caregiver skills training interventions for autism that have been successfully adapted or designed for telehealth delivery. Telehealth interventions that are scalable, adaptable, caregiver-mediated, open-access, and delivered as part of a stepped care model, have the potential to address the global treatment gap for families of children with autism and other neurodevelopmental disabilities. Considerations relevant to the global scale-up of caregiver-mediated interventions will also be discussed.

KEYWORDS

autism (ASD), telehealth, caregiver-mediated intervention, parent training, global health, neurodevelopmental disabilities, eLearning, online training

Introduction

Autism is a common neurodevelopmental condition affecting an estimated 1% of the population globally (1). It is characterized by difficulties in communication, social interaction and flexibility, differences in sensory processing, patterns of intense interests and repetitive behaviors (2, 3). While clinical information stresses deficits, certain strengths appear to be common in autistic individuals, including memory, attention to detail, skills related to strong special interests, and positive personality traits, including fairness and authenticity (4–7). Furthermore there is increasing recognition that valuing autism and neurodiversity benefits society as a whole (8).

Significant evidence shows that caregivers of children with autism can learn skills to promote their children's development and wellbeing (9, 10). Domains addressed by caregiver skills training include caregiver-child interaction, child joint attention, social interaction, communication, positive behavior, play, adaptive functioning, and other concerns, such as repetitive behaviors, self-regulation and sensory processing (11). Caregiver stress and lower perceived competence are common issues for caregivers of children with autism that can be improved through caregiver skills training (12–14).

Caregiver skills training is an attractive option for reducing the treatment gap globally because the majority of children with developmental disabilities, including autism, live in low- and middle -income countries where the availability of interventions is extremely limited (15). Even in high resource settings, wait times for services are lengthy, and the availability of trained personnel is a barrier (16).

The COVID-19 pandemic and the measures to contain it—including social isolation, school and workplace closures and interruptions in health and social services—have negatively affected children with autism and their families (17–19). Despite this, it is important to remember that children and families are often resilient in the face of adversity (20, 21). While causing undeniable hardship, the pandemic and the measures to contain it have also inspired innovation and propelled the use of digital technologies forward, including for training caregivers (22, 23).

This review focuses on interventions designed to train caregivers of young children with autism to support their child's development that are supported by randomized controlled trial (RCT) evidence of gains in child and/or caregiver outcomes and were adapted for telehealth delivery and published between April 2021 and Nov 2022. Selected examples of other telehealth-delivered caregiver training in Naturalistic Developmental Behavioral Interventions (NDBIs) are included to illustrate a diversity of approaches, especially those that show potential for broader scale-up.

Caregiver-mediated interventions

Caregiver-mediated interventions are adaptations of professionally delivered interventions or interventions developed specifically for implementation by caregivers. They give children the opportunity to benefit from daily exposure to treatment techniques during regular interactions and activities with family members (24). Many caregiver-mediated interventions belong to the category of NDBIs, which are implemented through play and daily activities, informed by developmental and behavioral science, and focused on improving social communication and development (24). As a group, NDBIs share certain features, such as utilizing typical daily interactions, following the child's interest, turn taking in shared activities, imitation of the child, adult modeling of language and other skills, and natural reinforcers of the child's communication attempts and skills (24). Training caregivers to deliver NDBIs for children with autism has been proposed as a cost-effective method for reducing the treatment gap for children with developmental disabilities because it enables children to receive interventions that are otherwise unavailable due to a lack of trained professionals (24).

Programs that train caregivers to deliver NDBIs have typically been offered in person, in group or individual format and typically involve teaching skills and strategies using instruction, modeling, rehearsal, and feedback (25, 26). The use of telehealth (the provision of health services, information and education over the Internet and related technologies) offers an attractive option for scaling up care globally and has been proposed to address barriers to traditional caregiver training, including cost, lack of time, geographic isolation, lack of transportation and lack of childcare (27, 28). According to WHO, digital health technologies can improve accessibility, quality and affordability of health services, and should "complement and enhance existing health service delivery models, strengthen integrated, people-centered health services and contribute to improved population health, and health equity…" (29).

Telehealth adaptations of caregiver training programs have mainly used video conferencing software to replicate in-person delivery (30). On the other hand, some interventions, have been adapted solely for asynchronous self-directed internet-based instruction (eLearning), while others use a hybrid telehealth approach, combining telehealth-delivered caregiver coaching with self-directed eLearning and/or in-person elements. eLearning approaches that include an element of real-time remote contact with trainers has been advocated for, given that opportunities for coaching, feedback and support are associated with better outcomes (31, 32). Asynchronous telehealth interventions may include adjunctive peer-to-peer support via virtual support groups, chatrooms, discussion groups, bulletin boards or forums (33, 34) and these options should also be investigated in the context of telehealth research.

Telehealth adaptations of evidence-based caregiver-mediated interventions

In a recent systematic review, Ellison et al. analyzed diverse telehealth interventions provided to children and adolescents with autism and their families and findings suggested that telehealth services were equivalent or better than face-to-face services (30). The review sought to extend the findings of reviews by Sutherland et al. and Boisvert et al., which reported high caregiver satisfaction with telehealth interventions (35, 36). Across studies identified by Ellison et al., the most common telehealth modality was synchronous (real-time intervention) using video conferencing software. For caregiver coaching specifically, telehealth has advantages over in-person delivery of services, including cost effectiveness and increased ability to access underserved populations (31). Telehealth-provided interventions for children with autism were also systematically reviewed by de Nocker and Toolan, revealing improvements in child level outcomes that were similar between telehealth and in-person delivery (37). Consistently, a systematic review by Pacia et al. found that caregiver-mediated interventions designed to improve social communication for children with autism that used videoconferencing and other eLearning strategies demonstrated treatment effects that were similar to in-person interventions (38). However, a recent meta-analysis of 8 RCTs of technology-assisted parent-mediated interventions for children with autism that utilized apps, websites, DVDs and computer-based interventions showed no significant differences in social communication, social functioning or language outcomes, although the majority of the interventions used technology only and did not include caregiver coaching (39).

Studies of telehealth-adapted caregiver training in evidence-based NDBIs published between April 2021 and Nov 2022 that included a caregiver coaching component and evaluated child and/or caregiver outcomes are described below. The search strategy is described elsewhere (30).

Shire et al. (40) adapted JASPER (Joint Attention, Symbolic Play, Engagement, and Regulation) for delivery using video conferencing plus 3 home visits to coach caregivers (40). The proof-of-concept study, which was conducted with caregivers of 6 children aged 2-9 years with autism who were living in rural and remote Canadian communities, found gains in children's joint engagement and caregivers' use of JASPER strategies that were similar to those seen in the face-to-face intervention, however, conclusions about effectiveness could not be made given the scope of this study. Importantly, participating families found the telehealth-delivered intervention to be acceptable and effective, and descibed the ability to connect from home and flexibility as advantages, with new technology, time, and managing other children in the home as disadvantages (40).

The Social ABCs program was adapted for telehealth delivery and tested with urban Canadian caregivers from diverse linguistic, ethnic, and educational backgrounds (41). In-person ($n = 45$) was compared to telehealth delivery ($n = 37$) for six group learning sessions and nine 60-min individual coaching sessions. Results showed good caregiver fidelity of intervention and toddler gains in social-communication skills that were similar for in-person and telehealth delivery. Importantly, the telehealth-delivered program, which included individual coaching via videoconferencing, was acceptable and feasible for a diverse group of caregivers.

Internet-based Parent-implemented Communication Strategies-Storybook (i-PiCSS) trains and coaches caregivers to use naturalistic communication teaching strategies while reading storybooks with young children with developmental disabilities (42). A single case multiple-baseline study with 3 parents used self-directed eLearning modules and coaching via videoconferencing, and demonstrated fidelity of implementation and improvements in child communication.

Preschool Autism Communication Trial (PACT) is an evidence-based communication-focused treatment for children with autism that has been adapted for use in low resource settings, and was adapted to PACT-Generalized (PACT-G), which includes caregiver- and educator-mediated components (43, 44). PACT-G was provided in a hybrid model, with both in-person and telehealth-delivered sessions. In a parallel, single-blind, an RCT, 555 children aged 2–7 years were randomized to either PACT-G ($n = 122$) or treatment as usual ($n = 127$) (45). PACT-G demonstrated improved social communication and positive effects on parental wellbeing and child disruptive behavior, although autism symptom outcomes were not different from treatment as usual and intervention dosage and poorer remote session quality were named as potential contributors to these findings (45).

WHO's Caregiver Skills Training for Families of Children with Developmental Delays or Disabilities (CST) has been tested with diverse caregivers and was developed as an open-access program for families of children aged 2–9 years to improve access to evidence-based care (46). The program, which was designed to be culturally and contextually adapted, consists of 9 group sessions and 3 home visits delivered by trained and supervised non-specialist facilitators. It has demonstrated acceptability and feasibility when delivered in-person in Ethiopia, India, Italy and Hong Kong, and virtually in Hong Kong and rural Missouri (47–51). A pilot RCT of CST in Italy ($n = 86$) demonstrated a large and significant effect on parent skills supporting joint engagement and benefits in terms of parental stress, self-efficacy, and child gestures, although changes in autism symptom severity and joint engagement were not statistically significant (52). CST was evaluated in Hong Kong and different methods of delivery were compared to waitlist control: asynchronous eLearning (digital versions of participant guides and pre-recorded videos demonstrating CST skills and strategies) ($n = 9$), videoconferencing ($n = 7$), and a hybrid model with group sessions delivered by videoconferencing and in-person home visits with coaching ($n = 9$) (49). In this study, all modes of delivery showed high acceptability and feasibility, and in-person and video conferencing were associated with greater improvements in caregivers' wellbeing and child's communication and behaviors compared to self-directed eLearning and control. An eLearning version of CST (eCST) was recently released in English on WHO's free learning platform, OpenWHO[1], and is currently in field testing.

Caregiver training to improve child behavior has also been effectively delivered via telehealth. Collaborative Model for

---

[1] https://openwho.org/courses/caregiver-skills-training

Promoting Competence and Success (COMPASS for Hope) delivered in 4 group and 4 individual sessions was compared for videoconference-delivery ($n = 10$), in-person delivery ($n = 13$) and waitlist control ($n = 10$), and showed similar benefits for telehealth and in-person delivery compared to control (53). Additionally, telehealth-provided Research Unit for Behavior Intervention (RUBI), a parent-mediated intervention protocol for children with autism was compared in an RCT to treatment as usual ($n = 38$, ages 21–84 months) and showed good efficacy in reducing disruptive behavior and high acceptability when provided in 12 weeks of real-time remote training and coaching (54). RUBI was also compared for face-to-face ($n = 24$) and virtual delivery ($n = 31$) in Israel with a sociodemographically diverse population and a reduction of disruptive behaviors was shown for both groups (55). Evidence also indicates that caregiver training to improve disruptive behavior is effective and acceptable when offered by telehealth in multiple countries and in low resource settings (56).

An applied behavior analysis (ABA)-based naturalistic communication intervention that was delivered by didactic training and live feedback sessions by teleconference was conducted with trainers and 5 families of children aged 2–7 years located in Ireland and Scotland. It was associated with variable gains in child communication and improved positive affect (57).

Potential for digital innovation in the delivery of caregiver training

Recently, Gentile et al. evaluated the ATHENA telehealth program, which was delivered via an app and tablet, and included both synchronous and asynchronous sessions, modeling and parent coaching (58). The intervention, which tested caregiver outcomes only ($n = 27$) using a pre-post design, was associated with improvements in caregivers' empowerment, stress level, and behaviors to promote child learning, however, increased caregiver age was found to be a moderating factor that was associated with decreased empowerment and increased stress over the course of the intervention. The authors hypothesized that this may be due low acceptability and familiarity with technology among older caregivers, which has been proposed by others (59). This speaks to the need for a diversity of intervention approaches to meet the diverse needs of caregivers.

Wainer and Ingersoll evaluated a caregiver-mediated intervention for young children with autism, Improving Parents As Communication Teachers (ImPACT) Online, that combined self-directed internet-based instruction with therapist coaching (60). ImpACT Online consists of 12 eLearning lessons with explanatory video clips, a manual, self-assessments, interactive exercises and homework assignments. The study showed that caregivers were able to learn and use program skills and their children's spontaneous imitation skills improved. Furthermore, the hybrid model was acceptable and feasible, raising the potential that such hybrid telehealth programs that utilize self-directed and facilitator-provided elements can be used to increase access to evidence-based autism services. The same online intervention was assessed in a pilot RCT which compared self-directed eLearning vs. therapist-assisted eLearning for caregivers of children aged 2 to 6 years ($n = 27$). Both interventions showed high caregiver engagement and satisfaction but therapist assistance increased engagement and course completion (61). Child outcomes were not measured.

Ingersoll et al. randomized caregivers of children aged 1.5–6 years to receive self-directed ($n = 13$) or therapist-assisted ($n = 14$) telehealth delivery models of ImPACT Online (62). Both groups were given access to the training website and support from a technology navigator, with the therapist-assisted group receiving two 30-min telehealth sessions per week for 12 weeks. Both groups showed improved child language and caregiver self-efficacy, stress, and positive parental perceptions of their child, however, the therapist-assisted group showed improved child social skills, and greater gains in parental intervention fidelity and positive perceptions of the child (62).

Hao et al. used a quasi-experimental design to evaluate SKILLS (Skills and Knowledge of Intervention for Language Learning Success) an adaptation of Project ImPACT, in which caregivers of children aged 1–10 years chose between in-person ($n = 15$) or virtual training provided by videoconference ($n = 15$) (63). Groups were matched based on the child's age and gender, and maternal education. The intervention, which consisted of eight 1-h sessions, showed equivalent and comparable outcomes between groups, specifically, parents demonstrated fidelity of implementation and children showed gains in vocabulary and language complexity (63).

A stepped care approach to the use of telehealth interventions is increasingly being advocated for, whereby families are first offered a lower intensity intervention, and then a more intensive intervention is offered to those children and families with higher needs, certain characteristics or poorer treatment response (8). Wainer et al. conducted a small proof of concept trial in which families of children with autism ages 1.5 to 5 years ($n = 20$) were offered a self-directed eLearning program to enhance social imitation (64). Subsequently, families that demonstrated lower fidelity and no improvement in caregiver self-efficacy were directed into 5 sessions of coaching by video conference. The stepped care model showed high acceptability and feasibility with improvements in fidelity, parental self-efficacy and child social communication, although differences in child imitation ability and family quality of life were not significant.

Considerations for global scale-up

Caregiver training that can be delivered by telehealth at scale has the potential to address global health inequities. However, the "digital divide" is used to describe the disparity between people in high-income contexts, and low- and middle-income contexts who may have limited access to internet-enabled devices, electricity, high speed internet and sufficient data to access digital interventions (65). The use of A.I.-based caregiver coaching is on the horizon and could help facilitate scale-up of caregiver-mediated interventions by mitigating the lack of skilled personnel, however, use of such advanced technologies will likely exacerbate the digital divide. There are some strategies for reducing barriers, such as making flexible digital interventions freely available through smart phone apps (65), but it will not be possible to address the treatment gap with a one-size-fits all model, and continued support of low-tech delivery of interventions, especially in certain contexts, can prevent widening of the digital divide (66). Multiple approaches will be needed to adapt existing evidence-based interventions and develop novel interventions that meet the needs of caregivers who are diverse in terms of language, culture, geography and context, including access to internet-enabled technologies (8, 62, 63).

Certain characteristics increase the scalability of evidence-based interventions and should be considered during the process of intervention design, including being manualized, deliverable by trained non-specialists, culturally and contextually adaptable, brief, affordable, group-based, and utilizing self-directed eLearning (67). Interventions need to be adaptable to the social, linguistic, economic, cultural and geographic context in which they are delivered (68–70). Some adapted evidence-based interventions may show decreased effectiveness compared to the original intervention, however, if modest, this may be acceptable given the trade-off with increased coverage and accessibility (67). At the same time, adapted evidence-based interventions should demonstrate acceptability, feasibility, relevance and cultural and contextual appropriateness across diverse populations, while also demonstrating effectiveness. The heterogeneous needs of children with autism and their families highlight the need for a stepped care approach and personalized models of intervention that consider the preferences, needs, and costs to individuals and families (8). Also, whenever possible, inclusion of children with other neurodevelopmental conditions should be prioritized in autism intervention research to potentially benefit a wider range of children and families (8).

Discussion

Given the established body of knowledge that earlier identification and intervention leads to better outcomes (71), and the ability to detect autism in younger populations (72), the ability to scale up caregiver training and reach caregivers of newly identified children early, are critical to improving the trajectories for these children. Telehealth interventions that are scalable, open-access, caregiver-mediated and delivered as part of a stepped care approach have the potential to address the global treatment gap for families of children with autism and other neurodevelopmental disabilities. However, to address rather than exacerbate health inequities, interventions should be provided as part of universal health coverage (73). Future research should continue to assess acceptability and feasibility of telehealth interventions while acknowledging the digital divide these interventions can create (30).

Lack of data on the effectiveness of telehealth interventions remains a major barrier to implementation and scale up, therefore, larger randomized controlled studies that collect data on acceptability, feasibility and relevance, and utilize standard outcome measures when possible, are needed (30, 31, 74). Shreiberman and colleagues advocate for six research considerations to advance the development of NDBIs, including larger scale RCTs to address moderators, mediators and efficiency of treatments; measurement of functional changes in child behavior; analysis of the active ingredients within multicomponent interventions; measurements of treatment fidelity; testing treatment strategies for children who are poor responders; and use of implementation science to facilitate broad implementation (24). Furthermore, there is a need to test interventions across a range of different populations and contexts (27).

Increased investment and sustained funding for telehealth interventions is critical if these research priorities are to be realized, especially those that improve health equity, and allow interventions to reach rural, marginalized and disadvantaged families globally. Implementation of universal health coverage also requires adequate funding, political will and a commitment to the idea that all people deserve access to health (73, 75). It is also aided by the understanding that health equity benefits whole societies, not just the most marginalized (76).

Finally, it is critical to involve autistic adolescents and adults in the development and adaptation of interventions for children with autism and their families. There are now more self-identified autistic researchers and scholars in the field, although greater progress could be made in the adoption of participatory and emancipatory research methods (77). Resources for and by people with autism [see, for example, (78–80)] should be studied to determine potential benefit when included in telehealth interventions for families of children with autism. Meaningful involvement of people with autism in shaping research agendas to meet local needs has the potential to improve the relevance and impact of research and reduce health disparities for children and adults with autism and other neurodevelopmental disabilities (81–83).

Author contributions

The author confirms being the sole contributor of this work and has approved it for publication.

Acknowledgments

The author is grateful to Erica Salomone and Chiara Servili for their guidance on an early version of this manuscript.

Conflict of interest

LP serves as a paid consultant for WHO. Typically this is not a conflict of interest but could be considered one in the context of this article.

Publisher's note

References

1. Zeidan J, Fombonne E, Scorah J, Ibrahim A, Durkin MS, Saxena S, et al. Global prevalence of autism: a systematic review update. *Autism Res.* (2022) 15:778–90. doi: 10.1002/aur.2696

2. American Psychiatric Association. *Diagnostic and Statistical Manual of Mental Disorders.* Virgina: American Psychiatric Association (2013). doi: 10.1176/appi.books.9780890425596

3. World Health Organization. *International Classification of Diseases, 11th Revision (ICD-11).* Geneva: World Health Organization. (2018). Available online at: https://icd.who.int/en (accessed April 8, 2022).

4. Kirchner J, Ruch W, Dziobek I. Brief report: character strengths in adults with autism spectrum disorder without intellectual impairment. *J Autism Dev Disord.* (2016) 46:3330–7. doi: 10.1007/s10803-016-2865-7

5. Dawson M, Soulières I, Gernsbacher MA, Mottron L. The level and nature of autistic intelligence. *Psychol Sci.* (2007) 18:657–62. doi: 10.1111/j.1467-9280.2007.01954.x

6. Russell G, Kapp SK, Elliott D, Elphick C, Gwernan-Jones R, Owens C. Mapping the autistic advantage from the accounts of adults diagnosed with autism: a qualitative study. *Autism Adulthood.* (2019) 1:124–33. doi: 10.1089/aut.2018.0035

7. Kirchner JC, Dziobek I. Toward the successful employment of adults with autism: a first analysis of special interests and factors deemed important for vocational performance. *Scand J Child Adolesc Psychiatr Psychol.* (2013) 2:77–85. doi: 10.21307/sjcapp-2014-011

8. Lord C, Charman T, Havdahl A, Carbone P, Anagnostou E, Boyd B, et al. The lancet commission on the future of care and clinical research in autism. *Lancet.* (2022) 399:271–334. doi: 10.1016/S0140-6736(21)01541-5

9. Oono IP, Honey EJ, Mcconachie H. *Parent-Mediated Early Intervention for Young Children with autism Spectrum Disorders (ASD). Vol. 2013, Cochrane Database of Systematic Reviews.* Hoboken: John Wiley and Sons Ltd. (2013). doi: 10.1002/14651858.CD009774.pub2

10. Naveed S, Waqas A, Amray AN, Memon RI, Javed N, Tahir MA, et al. Implementation and effectiveness of non-specialist mediated interventions for children with Autism Spectrum Disorder: a systematic review and meta-analysis. *PLoS ONE.* (2019) 14:e0224362. doi: 10.1371/journal.pone.0224362

11. Althoff CE, Dammann CP, Hope SJ, Ausderau KK. Parent-mediated interventions for children with autism spectrum disorder: a systematic review. *Am J Occup Therapy.* (2019) 73:1–3. doi: 10.5014/ajot.2019.030015

12. Valicenti-Mcdermott M, Lawson K, Hottinger K, Seijo R, Schechtman M, Shulman L, et al. Parental stress in families of children with autism and other developmental disabilities. *J Child Neurol.* (2015) 30:1728–35. doi: 10.1177/0883073815579705

13. Estes A, Yoder P, McEachin J, Hellemann G, Munson J, Greenson J, et al. The effect of early autism intervention on parental sense of efficacy in a randomized trial depends on the initial level of parent stress. *Autism.* (2021) 25:1924–34. doi: 10.1177/13623613211005613

14. Kurzrok J, McBride E, Grossman RB. Autism-specific parenting self-efficacy: An examination of the role of parent-reported intervention involvement, satisfaction with intervention-related training, and caregiver burden. *Autism.* (2021) 25:1395–408. doi: 10.1177/1362361321990931

15. Reichow B, Servili C, Yasamy MT, Barbui C, Saxena S. Non-specialist psychosocial interventions for children and adolescents with intellectual disability or lower-functioning autism spectrum disorders: a systematic review. *PLoS Med.* (2013) 10:e1001572. doi: 10.1371/journal.pmed.1001572

16. Yingling ME, Hock RM, Bell BA. Time-lag between diagnosis of autism spectrum disorder and onset of publicly-funded early intensive behavioral intervention: do race-ethnicity and neighborhood matter? *J Autism Dev Disord.* (2018) 48:561–71. doi: 10.1007/s10803-017-3354-3

17. Colizzi M, Sironi E, Antonini F, Ciceri ML, Bovo C, Zoccante L. Psychosocial and behavioral impact of COVID-19 in autism spectrum disorder: an online parent survey. *Brain Sci.* (2020) 10:341. doi: 10.3390/brainsci10060341

18. White SW, Stoppelbein L, Scott H, Spain D. It took a pandemic: Perspectives on impact, stress, and telehealth from caregivers of people with autism. *Res Dev Disabil.* (2021) 113:1–8. doi: 10.1016/j.ridd.2021.103938

19. Dekker L, Hooijman L, Louwerse A, Visser K, Bastiaansen D, ten Hoopen L, et al. Impact of the COVID-19 pandemic on children and adolescents with autism spectrum disorder and their families: a mixed-methods study protocol. *BMJ Open.* (2022) 12:e049336. doi: 10.1136/bmjopen-2021-049336

20. Iacob CI, Avram E, Cojocaru D, Podina IR. Resilience in familial caregivers of children with developmental disabilities: a meta-analysis. *J Autism Dev Disord.* (2020) 50:4053–68. doi: 10.1007/s10803-020-04473-9

21. Bekhet AK, Johnson NL, Zauszniewski JA. Resilience in family members of persons with autism spectrum disorder: a review of the literature. *Issues Ment Health Nurs.* (2012) 33:650–6. doi: 10.3109/01612840.2012.671441

22. Amaral DG, Anagnostou E, Bal VH, Barbaro J. phine, Barber AB, Canal-Bedia R, et al. COVID-19 and autism research: perspectives from around the globe. *Autism Res.* (2020) 13:844–69. doi: 10.1002/aur.2329

23. Vanegas SB, Dueñas AD, Kunze M, Xu Y. Adapting parent-focused interventions for diverse caregivers of children with intellectual and developmental disabilities: lessons learned during global crises. *J Policy Pract Intellect Disabil.* (2022). Available online at: https://onlinelibrary-wiley-com.myaccess.library.utoronto.ca/doi/full/10.1111/jppi.12422 (accessed Nov 20, 2022) doi: 10.1111/jppi.12422

24. Schreibman L, Dawson G, Stahmer AC, Landa R, Rogers SJ, McGee GG, et al. Naturalistic developmental behavioral interventions: empirically validated treatments for autism spectrum disorder. *J Autism Dev Disord.* (2015) 45:2411–28. doi: 10.1007/s10803-015-2407-8

25. Rahman A, Divan G, Hamdani SU, Vajaratkar V, Taylor C, Leadbitter K, et al. Effectiveness of the parent-mediated intervention for children with autism spectrum disorder in south Asia in India and Pakistan (PASS): a randomised controlled trial. *Lancet Psychiatry.* (2016) 3:128–36. doi: 10.1016/S2215-0366(15)00388-0

26. Sun X. Behavior skills training for family caregivers of people with intellectual or developmental disabilities: a systematic review of literature. *Int J Develop Disabil.* (2020) 68:247–73. doi: 10.1080/20473869.2020.1793650

27. Franz L, Howard J, Viljoen M, Sikich L, Chandrasekhar T, Kollins SH, et al. Pragmatic adaptations of telehealth-delivered caregiver coaching for children with autism in the context of COVID-19: perspectives from the United States

and South Africa. *Autism.* (2022) 26:270–5. doi: 10.1177/13623613211022585

28. Harris L, Gilmore D, Hanks C, Coury D, Moffatt-Bruce S, Garvin JH, et al. It was surprisingly equivalent to the appointment I had in person: advantages and disadvantages of synchronous telehealth for delivering primary care for autistic adults. *Autism.* (2022) 26:1573–80. doi: 10.1177/13623613211060589

29. WHA71.7 Digital health. Available online at: https://www.who.int/publications/i/item/10665-279505 (accessed Aug 2, 2022)

30. Ellison KS, Guidry J, Picou P, Adenuga P, Davis TE. Telehealth and autism prior to and in the age of COVID-19: a systematic and critical review of the last decade. *Clin Child Fam Psychol Rev.* (2021) 24:599–630. doi: 10.1007/s10567-021-00358-0

31. Knutsen J, Wolfe A, Burke BL, Hepburn S, Lindgren S, Coury D, et al. Systematic review of telemedicine in autism spectrum disorders. *Rev J Autism Dev Disord.* (2016) 3:330–44. doi: 10.1007/s40489-016-0086-9

32. Yosep I, Prayogo SA, Kohar K, Andrew H, Mardhiyah A, Amirah S, et al. Managing autism spectrum disorder in the face of pandemic using internet-based parent-mediated interventions: a systematic review of randomized controlled trials. *Children.* (2022) 9:1483. doi: 10.3390/children9101483

33. Ali K, Farrer L, Gulliver A, Griffiths KM. Online peer-to-peer support for young people with mental health problems: a systematic review. *JMIR Ment Health.* (2015) 2:e19. doi: 10.2196/mental.4418

34. Niela-Vilén H, Axelin A, Salanterä S, Melender HL. Internet-based peer support for parents: a systematic integrative review. *Int J Nurs Stud.* (2014) 51:1524–37. doi: 10.1016/j.ijnurstu.2014.06.009

35. Sutherland R, Trembath D, Hodge MA, Rose V, Roberts J. Telehealth and autism: Are telehealth language assessments reliable and feasible for children with autism? *Int J Lang Commun Disord.* (2019) 54:281–91. doi: 10.1111/1460-6984.12440

36. Boisvert M, Lang R, Andrianopoulos M, Boscardin ML. Telepractice in the assessment and treatment of individuals with autism spectrum disorders: a systematic review. *Dev Neurorehabil.* (2010) 13:423–32. doi: 10.3109/17518423.2010.499889

37. de Nocker YL, Toolan CK. Using telehealth to provide interventions for children with ASD: a systematic review. *Rev J Autism Develop Disorders Springer.* (2021) 1–31. doi: 10.1007/s40489-021-00278-3

38. Pacia C, Holloway J, Gunning C, Lee H. A systematic review of family-mediated social communication interventions for young children with autism. *Rev J Autism Develop Disorders.* (2021) 9:208–34. doi: 10.1007/s40489-021-00249-8

39. Pi HJ, Kallapiran K, Munivenkatappa S, Kandasamy P, Kirubakaran R, Russell P, et al. Meta-analysis of RCTs of technology-assisted parent-mediated interventions for children with ASD. *J Autism Dev Disord.* (2022) 52:3325–43. doi: 10.1007/s10803-021-05206-2

40. Shire SY, Worthman LB, Arbuckle S. A technology-enabled adaptation of face-to-face caregiver-mediated JASPER intervention: preliminary examination of video conferenced caregiver coaching. *Am J Intellect Dev Disabil.* (2021) 126:421–34. doi: 10.1352/1944-7558-126.5.421

41. Brian J, Solish A, Dowds E, Roth I, Bernardi K, Perry K, et al. "Going Mobile"-increasing the reach of parent-mediated intervention for toddlers with ASD *via* group-based and virtual delivery. *J Autism Dev Disord.* (2022) 52:5207–20. doi: 10.1007/s10803-022-05554-7

42. Akemoglu Y, Laroue D, Kudesey C, Stahlman M. A module-based telepractice intervention for parents of children with developmental disabilities. *J Autism Dev Disord.* (2022) 52:5177–90. doi: 10.1007/s10803-022-05549-4

43. Green J, Charman T, McConachie H, Aldred C, Slonims V, Howlin P, et al. Parent-mediated communication-focused treatment in children with autism (PACT): a randomised controlled trial. *Lancet.* (2010) 375:2152–60. doi: 10.1016/S0140-6736(10)60587-9

44. Divan G, Hamdani SU, Vajartkar V, Minhas A, Taylor C, Aldred C, et al. Adapting an evidence-based intervention for autism spectrum disorder for scaling up in resource-constrained settings: the development of the PASS intervention in South Asia. *Glob Health Action.* (2015) 8:27278. doi: 10.3402/gha.v8.27278

45. Green J, Leadbitter K, Ellis C, Taylor L, Moore HL, Carruthers S, et al. Combined social communication therapy at home and in education for young autistic children in England (PACT-G): a parallel, single-blind, randomized controlled trial. *Lancet Psychiatry.* (2022) 9:307–20. doi: 10.1016/S2215-0366(22)00029-3

46. Salomone E, Pacione L, Shire S, Brown FL, Reichow B, Servili C. Development of the WHO caregiver skills training program for developmental disorders or delays. *Front Psychiatry.* (2019) 10:769. doi: 10.3389/fpsyt.2019.00769

47. Tekola B, Girma F, Kinfe M, Abdurahman R, Tesfaye M, Yenus Z, et al. Adapting and pre-testing the World Health Organization's caregiver skills training programme for autism and other developmental disorders in a very low-resource setting: findings from Ethiopia. *Autism.* (2020) 24:51–63. doi: 10.1177/1362361319848532

48. Salomone E, Ferrante C, Salandin A, Ferrara F, Torchio E, Foletti G, et al. Acceptability and feasibility of the World Health Organization's Caregiver Skills Training implemented in the Italian National Health System. *Autism.* (2021) 26:859–74. doi: 10.1177/13623613211035228

49. Lau J, Lai SMK, Ip FTS, Wong PWC, Team WC, Shire S, et al. Acceptability and feasibility of the world health organization's caregiver skills training programme delivered *via* e-learning, video-conferencing, and in-person hybrid modalities in Hong Kong. *Front Psychiatry.* (2022) 1855. doi: 10.3389/fpsyt.2022.915263

50. Wong PWC, Lam YY, Lau JSP, Fok HK, Servili C, Salomone E, et al. Adapting and pretesting the World Health Organization's Caregiver Skills Training Program for children with autism and developmental disorders or delays in Hong Kong. *Sci Rep.* (2022) 12:1. doi: 10.1038/s41598-022-21343-9

51. Montiel-Nava C, Tregnago M, Marshall J, Sohl K, Curran AB, Mahurin M, et al. Implementing the WHO caregivers skills training program with caregivers of autistic children *via* telehealth in rural communities. *Front Psychiatry.* (2022) 909947. doi: 10.3389/fpsyt.2022.909947

52. Salomone E, Settanni M, McConachie H, Suma K, Ferrara F, Foletti G, et al. Pilot randomized controlled trial of the WHO caregiver skills training in public health services in Italy. *J Autism Dev Disord.* (2021) 52: 4286–300. doi: 10.1007/s10803-021-05297-x

53. Kuravackel GM, Ruble LA, Reese RJ, Ables AP, Rodgers AD, Toland MD, et al. for Hope: evaluating the effectiveness of a parent training and support program for children with ASD. *J Autism Dev Disord.* (2018) 48:404–16. doi: 10.1007/s10803-017-3333-8

54. Lindgren S, Wacker D, Schieltz K, Suess A, Pelzel K, Kopelman T, et al. A randomized controlled trial of functional communication training *via* telehealth for young children with autism spectrum disorder. *J Autism Dev Disord.* (2020) 50:4449–62. doi: 10.1007/s10803-020-04451-1

55. Graucher T, Sinai-Gavrilov Y, Mor Y, Netzer[1] S, Cohen EY, Levi L, et al. From clinic room to zoom: delivery of an evidence-based, parent-mediated intervention in the community before and during the pandemic. *J Autism Dev Disord.* (2022) 52:5222–31. doi: 10.1007/s10803-022-05592-1

56. Tsami L, Lerman D, Toper-Korkmaz O. Effectiveness and acceptability of parent training *via* telehealth among families around the world. (2019). Available online at: https://onlinelibrary.wiley.com/doi/10.1002/jaba.645 (accessed Oct 18, 2022). doi: 10.1002/jaba.645

57. Ferguson J, Dounavi K, Craig EA. The impact of a telehealth platform on ABA-based parent training targeting social communication in children with autism spectrum disorder. *J Dev Phys Disabil.* (2022) 34:1089–120. doi: 10.1007/s10882-022-09839-8

58. Gentile M, Messineo L, la Guardia D, Arrigo M, Città G, Ayala A, et al. A parent-mediated telehealth program for children with autism spectrum disorder: promoting parents' ability to stimulate the children's learning, reduce parenting stress, and boost their sense of parenting empowerment. *J Autism Dev Disord.* (2022) 1–16. doi: 10.1007/s10803-022-05482-6

59. Venkatesh V, Morris MG, Davis GB, Davis FD. User acceptance of information technology: toward a unified view. *MIS Quarterly.* (2003) 425–78. doi: 10.2307/30036540

60. Wainer AL, Ingersoll BR. Increasing access to an ASD imitation intervention *via* a telehealth parent training program. *J Autism Dev Disord.* (2015) 45:3877–90. doi: 10.1007/s10803-014-2186-7

61. Ingersoll B, Berger NI. Parent engagement with a telehealth-based parent-mediated intervention program for children with autism spectrum disorders: predictors of program use and parent outcomes. *J Med Internet Res.* (2015) 17:e4913. doi: 10.2196/jmir.5329

62. Ingersoll B, Wainer AL, Berger NI, Pickard KE, Bonter N. Comparison of a self-directed and therapist-assisted telehealth parent-mediated intervention for children with ASD: a pilot RCT. *J Autism Dev Disord.* (2016) 46:2275–84. doi: 10.1007/s10803-016-2755-z

63. Hao Y, Franco JH, Sundarrajan M, Chen Y. A pilot study comparing tele-therapy and in-person therapy: perspectives from parent-mediated intervention for children with autism spectrum disorders. *J Autism Dev Disord.* (2021) 51:129–43. doi: 10.1007/s10803-020-04439-x

64. Wainer AL, Arnold ZE, Leonczyk C, Valluripalli Soorya L. Examining a stepped-care telehealth program for parents of young children with autism: a proof-of-concept trial. *Mol Autism.* (2021) 12:1–14. doi: 10.1186/s13229-021-00443-9

65. Kumm AJ, Viljoen M, de Vries PJ. The digital divide in technologies for autism: feasibility considerations for low- and middle-income countries. *J Autism Dev Disord*. (2021) 52:2300–13. doi: 10.1007/s10803-021-05084-8

66. Alkureishi MA, Choo ZY, Rahman A, Ho K, Benning-Shorb J, Lenti G, et al. Digitally disconnected: qualitative study of patient perspectives on the digital divide and potential solutions. *JMIR Hum Factors*. (2021) 8:e33364. doi: 10.2196/33364

67. Department of Mental Health and Substance Abuse. *Scalable Psychological Interventions for People in Communities Affected by Adversity. A New Area of Mental Health and Psychosocial Work at WHO*. Geneva: WHO (2017). p. 8.

68. Escoffery C, Mullen PD, Lebow-Skelley E, Haardoerfer R, Böing E, Udelson H, et al. A systematic review of adaptations of evidence-based public health interventions globally. *Implementation Sci*. (2018) 13:125 doi: 10.1186/s13012-018-0815-9

69. Rathod S, Gega L, Degnan A, Pikard J, Khan T, Husain N, et al. The current status of culturally adapted mental health interventions: a practice-focused review of meta-analyses. *Neuropsychiatr Dis Treat*. (2018) 14:165–78. doi: 10.2147/NDT.S138430

70. Castro FG, Barrera M, Steiker LKH. Issues and challenges in the design of culturally adapted evidence-based interventions. *Annu Rev Clin Psychol*. (2010) 6:213–39. doi: 10.1146/annurev-clinpsy-033109-132032

71. Hyman SL, Levy SE, Myers SM, Kuo DZ, Apkon CS, Davidson LF, et al. Identification, evaluation, and management of children with autism spectrum disorder. *Pediatrics*. (2020) 145:1. doi: 10.1542/9781610024716-part01-ch002

72. Shaw KA, McArthur D, Hughes MM, Bakian A v., Lee LC, Pettygrove S, et al. Progress and disparities in early identification of autism spectrum disorder: autism and developmental disabilities monitoring network, 2002–2016. *J Am Acad Child Adolesc Psychiatry*. (2021) 61:905–14 doi: 10.1016/j.jaac.2021.11.019

73. Verrecchia R, Thompson R, Yates R. Universal health coverage and public health: a truly sustainable approach. *Lancet Public Health*. (2019) 4:e10–1. doi: 10.1016/S2468-2667(18)30264-0

74. Parsons D, Cordier R, Vaz S, Lee HC. Parent-mediated intervention training delivered remotely for children with autism spectrum disorder living outside of urban areas: systematic review. *J Med Internet Res*. (2017) 19:e6651. doi: 10.2196/jmir.6651

75. World Health Organization. *Primary Health Care on the Road to Universal Health Coverage: 2019 Monitoring Report*. Available online at: https://www.who.int/publications/i/item/9789240029040 (accessed April 8, 2022).

76. Weinstein JN, Geller A, Negussie Y, Baciu A. *Communities in Action: Pathways to Health Equity*. Washington, DC: National Academies Press (2017). p. 1–558. doi: 10.17226/24624

77. Chown N, Robinson J, Beardon L, Downing J, Hughes L, Leatherland J, et al. Improving research about us, with us: a draft framework for inclusive autism research. *Disabil Soc*. (2017) 32:720–34. doi: 10.1080/09687599.2017.1320273

78. Autistic Self Advocacy Network. *Welcome to the Autistic Community*. Washington, DC: The Autistic Press (2020).

79. Ambitious about Autism. *Young People Explain Autism*. (2022). Available online at: https://www.ambitiousaboutautism.org.uk/information-about-autism/understanding-autism/what-is-autism (accessed April 8, 2022).

80. Castellon S. *The Spectrum Girl's Survival Guide : How to Grow up Awesome and Autistic*. London: Jessica Kingsley Publishers (2020).

81. de Vries PJ. Thinking globally to meet local needs: Autism spectrum disorders in Africa and other low-resource environments. *Curr Opin Neurol*. (2016) 29:130–6. doi: 10.1097/WCO.0000000000000297

82. Abma TA, Broerse JEW. Patient participation as dialogue: setting research agendas. *Health Expect*. (2010) 13:160. doi: 10.1111/j.1369-7625.2009.00549.x

83. Fletcher-Watson S, Adams J, Brook K, Charman T, Crane L, Cusack J, et al. Making the future together: shaping autism research through meaningful participation. *Autism*. (2019) 23:943–53. doi: 10.1177/1362361318786721

frontiers | Frontiers in Psychiatry

TYPE Original Research
PUBLISHED 22 December 2022
DOI 10.3389/fpsyt.2022.937516

OPEN ACCESS

EDITED BY
Maria Luisa Scattoni,
National Institute of Health (ISS), Italy

REVIEWED BY
Liza Hopkins,
The Alfred Hospital, Australia
Megan Thomas,
Dalhousie University, Canada
Matthew Kivel,
Oregon Health and Science University,
United States

*CORRESPONDENCE
Mihaela D. Barokova
✉ mihaela.barokova@gmail.com

SPECIALTY SECTION
This article was submitted to
Autism,
a section of the journal
Frontiers in Psychiatry

RECEIVED 06 May 2022
ACCEPTED 05 December 2022
PUBLISHED 22 December 2022

CITATION
Barokova MD,
Andreeva-Sapundzhieva A,
Andonova E, Markova-Derelieva G
and Karpur A (2022) Diagnostic paths
and service needs of children with
autism spectrum disorder and with
other neurodevelopmental disorders
in Bulgaria.
Front. Psychiatry 13:937516.
doi: 10.3389/fpsyt.2022.937516

Diagnostic paths and service needs of children with autism spectrum disorder and with other neurodevelopmental disorders in Bulgaria

Mihaela D. Barokova[1]*, Ana Andreeva-Sapundzhieva[2], Elena Andonova[1], Galina Markova-Derelieva[3] and Arun Karpur[4]

[1]Department of Cognitive Science and Psychology, New Bulgarian University, Sofia, Bulgaria, [2]Center for Social Rehabilitation and Integration, Sofia, Bulgaria, [3]Department of Health Care and Social Work, New Bulgarian University, Sofia, Bulgaria, [4]Autism Speaks, Princeton, NJ, United States

Background: Currently, there are no official statistics about the number of children with developmental disorders in Bulgaria. This is the first systematic investigation of the needs, access to services, and priorities of families of children with developmental disorders in the country.

Aims: The study aims to: (1) characterize the needs of children with developmental disorders in Bulgaria; (2) to compare the needs and access to services of children with Autism Spectrum Disorders (ASD) and other neurodevelopmental disorders (oNDD); (3) and to examine the daily burden of their caregivers and how it varies based on their demographic characteristics, such as income and education.

Methods: We used an online family needs assessment survey to collect data from caregivers of children with developmental disorders in Bulgaria between April and July 2020. 195 parents of children with ASD and 73 parents of children with oNDD completed the questionnaire.

Results: Children with ASD waited longer than children with oNDD to receive a diagnosis. Caregivers in the ASD group also expressed first concerns about their child's development when their children were older and for different reasons than caregivers in the oNDD group. There were no significant differences between groups in service encounters, including access to and delay of medical, counseling, and educational services, with approximately 50% of all caregivers experiencing some delay and/or difficulties in access to services. There were no associations between access to services and caregiver education and family income, with the exception of higher education being linked to receiving a diagnosis earlier for the oNDD group.

Discussion: This study has three main findings: (1) children with ASD and children with oNDD in Bulgaria have different needs and paths to diagnosis; (2) nevertheless, children in both groups experience similar challenges in accessing medical, counseling, and educational services, regardless of their demographic characteristics; and (3) parents' priorities focus on education, counseling, and medical support, protecting children's basic rights, and raising awareness. A comparison of our findings to past research in the region shows a relative improvement in diagnostic services with families not having to travel outside their city to receive a diagnosis. Based on our findings, we provide specific recommendations for changes in services and policy.

KEYWORDS

diagnosis, service needs, Bulgaria, ASD, neurodevelopmental disorders

1 Introduction

Over the last decade, a concerted effort has been made to shed light on the needs of families of children with developmental delays, disorders, and disabilities in low- and middle-income countries in Europe. Specifically, the establishment of the Southeast European Autism Network (SEAN) in 2010 as part of the Autism Speaks Global Autism Public Health Initiative helped raise awareness about autism spectrum disorders (ASD) and other neurodevelopmental disorders (oNDD) (1). The increasing number of scientific meetings, conferences, clinical and parent workshops combined with the rapidly increasing number of parent-led organizations and non-governmental organizations suggests that there is a growing awareness and demand for services for children with disabilities in the region. Nevertheless, still very little is known about the specific needs and challenges families of children with developmental disorders[1] experience. To address this gap the present study reports primary data on the needs and daily burdens of families of children with developmental delays, disorders, and disabilities in Bulgaria collected online between April and July 2020.

1.1 Health care in Bulgaria

In order to situate our study in the context of services for children with developmental disorders provided in Bulgaria, we provide a brief overview of the country's health care system. In Bulgaria, there is a National Health Insurance Fund, whose main goal is "to ensure and guarantee free and equal access to health care for insured persons – through a set of types, scope and volume healthcare activities, as well as free choice of an executor who has a contract with a regional health insurance fund" (2). The fund is financed through mandatory taxes, specifically for health insurance. People can also enroll in private health insurance plans for additional benefits. In the context of seeking a diagnosis for a neurodevelopmental disorder, with referrals from a pediatrician or a general practitioner, families can receive consultation from a child psychiatrist and get medical tests done for free or for a subsidized fee. Once the child receives a formal diagnosis and a referral, they can access state-funded service providers for treatment and intervention. Families can also seek additional services through paid providers with varying fees. The wait times for accessing these services vary, and so do the kinds of services that are available at a specific resource center (counseling, speech therapy, occupational therapy, physical therapy, etc.).

1.2 The needs and experiences of children with developmental disorders in Bulgaria

In Bulgaria, there are no publicly available data on the total number of children with disabilities and their breakdown by diagnosis and specific medical, counseling, and educational needs. In the fall of 2019, the Bulgarian UNICEF office published statistics – provided by the Ministry of Education and Science – showing that during the 2019—2020 school year, the number of children with disabilities and with specific educational needs was over 25,000 (2.5% of all school-age children in Bulgaria in 2019), of whom 15,000 presented with difficulties in communication (3). At that time, additional 10,000 children with disabilities (1% of all school-age children in Bulgaria in 2019) were reported to not be enrolled in an educational institution. Based on these statistics, there are close to 35,000 school-aged children with disabilities in Bulgaria, which makes up 3.5% of all school-age children in the country ($N = 994\ 667$) based on data from the National Statistical Institute. This percentage would potentially be higher when infants and toddlers with developmental disorders are included. Nevertheless, there is no

---

[1] In the paper, the label "developmental disorder" is used to represent all formal diagnoses of a developmental delay, disorder, and/or disability as defined by the International Classification of Diseases.

published systematic investigation of the needs and priorities of these children and their families.

Of the few published, peer-reviewed papers and books on the topic, to our knowledge, only one used primary data from families (4), while the rest provide useful, albeit broad overview of the available services (5), relevant policy and governmental structures/mechanisms and the history and development of intervention, therapeutic, and research practices in the country (e.g., 6, 7). Daniels et al. (4) reported results from a parent questionnaire administered to caregivers of children with ASD ($N = 147$) in Bulgaria between 2013 and 2015. Based on their responses, parents expressed a first concern about their children when they were on average 24.7 months old, and the average age of diagnosis was 46.6 months. In addition, half of the parents reported traveling over 100 km to obtain their child's diagnosis. Parents reported on utilizing services, with 94% receiving speech and language therapy in the past and 83% receiving it at the time of assessment. In terms of receiving other supports for their children, 70% reported receiving government assistance, and 30% reported relying on advocacy groups and 42% of parents reported receiving training or assistance in general. Many parents (43%) endorsed feelings of helplessness in providing care for their children (4).

In terms of paths to a formal diagnosis, families' experiences vary. Based on anecdotal reports from parents, some seek out a diagnosis themselves by signing their children up for a diagnostic assessment. Other parents report that it is their pediatrician that encouraged them and provided them with a referral for a child psychiatrist. In Bulgaria, only a child psychiatrist can provide a formal diagnosis of ASD or other neurodevelopmental disorders. Typically, the diagnostic evaluation includes an assessment by a child psychiatrist, a neurologist, and a clinical psychologist among other professionals. An autism diagnosis is typically not given before the age of 3 years. Yet, other parents prefer to undergo an informal diagnostic evaluation that does not lead to the issuance of an official document. Overall, diagnostic experiences vary, but to date, there is no published formal evaluation of age of diagnosis, diagnostic procedures and protocols across clinics, and duration of the diagnostic process.

When it comes to access to services, there is also a lot of variation across families and providers. Access is greatly influenced by how their children's disorders are conceptualized. In the past, the medical model of disability identification was central (8). As a result, children were classified based on their disability and they were more likely to be placed in specialized institutions. In recent years, there has been a transition toward a more social model of conceptualizing these children's conditions and consequently educational experiences by focusing on their potential for learning and change.

In addition to the way developmental disorders are conceptualized, community awareness and attitudes could also influence the experiences of children with developmental disorders. An independent report on parental attitudes published by the Center for Inclusive Education demonstrated that parents' subjective level of acceptance of children with developmental disorders in the classroom varied by their diagnosis (9). Children with motor difficulties and speech-language disorders were accepted by 66% and 63% of respondents, respectively. Parents were much less likely to accept the integration of children with ASD in the classroom (33%) and even less likely to accept children with intellectual disabilities and multiple comorbid conditions (13–16%). The authors attributed these differences to lack of awareness about these children's conditions, their educational needs, and how they can be addressed in the classroom.

Although informative, these studies were all conducted over 5 years ago. Since then, many new government initiatives have taken place to address the needs of these children with developmental disorders and their families.

1.3 Major initiatives and policy changes in the last 5 years

As of the beginning of 2019, the Persons with Disabilities Act guarantees institutional/governmental support for persons with disabilities through means of medical, professional, social, occupational, and psychological rehabilitation, education and professional training, and access to information among others (10). Furthermore, this act regulates a monthly financial assistance for individuals with disabilities and imposes new obligations on employers to promote their employment.

Specifically targeting children's welfare and rights, the National Strategy for Children (2019–2030) was drafted, albeit delayed in its formal approval and implementation (11). This national strategy follows the aims and priorities set forth by the UN Convention on Children's Rights focusing on ensuring the children's rights to life and development. In the meantime, the Social Services Act that went into effect on 1 July 2020 guarantees individuals' right to social services, including therapy and rehabilitation, assistant services, and day and residential care (12). It also emphasizes the need for providing services not only for the child but for its family as a unit as well, which had not been emphasized in the past.

Perhaps, most progress has been made in the educational sector, specifically as it pertains to inclusive education. As of 2017, the ordinance on inclusive education regulates mandatory screening of all children between 3 and 3.5 years of age upon enrollment in kindergarten (13). Based on the screening and evaluation, an individual educational plan is to be designed. This new regulation complements earlier reforms of the Public Education Act (Article 21) now part of the Pre-School and School Education Act, which mandated the inclusion of children with specific educational needs in the schools, and allowed for the development of individual educational plans, while the child still attended school with typically developing peers (14).

Despite reforms in laws, regulations, and national strategies, whether and how they are implemented in practice and what their effects are on the daily lives of families with children with disabilities has not been examined systematically. The only document providing an evaluation of how the country ensures children's welfare is the yearly publication of the National Network for Children's Report Card (15). The Network is an alliance of organizations united by the common goal to promote and protect the rights of children in Bulgaria. In their yearly report card, they summarize and present the evaluations of various organizations, clinicians, teachers, and even children on whether and how the governmental structures have implemented laws and policies. The report focuses on key aspects of children's welfare including early child development, health, education, and protection of children's rights among others. Their evaluation claims that little progress has been made in early child development with key issues being the lack of enough trained professionals and lack of coordination between providers and governmental agencies. The report suggests that most progress has been made in the educational sector due to strong political commitment. Despite the well-developed plans and strategies for inclusive education, there is still a dearth of resource teachers (trained educators at school that work one-on-one with children inside and outside of the classroom) to address the needs of children with special educational needs in the schools (15).

1.4 Early intervention in Bulgaria

Although the policy has changed over the past 5 years, special note should be taken on the state of early intervention in the country. Based on data collected between 2018 and 2019, UNICEF published a report on the topic (16). Data were collected through focus groups and online questionnaires with parents, teachers, psychologists, speech-language pathologists (SLPs), kindergarten staff, and other professionals involved in early intervention. Based on the Ordinance on Prophylactic Examinations and Dispanserization Services from November 2016 (17), every general practitioner and pediatrician in Bulgaria is required to monitor the development of children visiting their practice. Based on the UNICEF report, indeed over 70% of parents report that their pediatrician monitored the physical development of their child (in terms of weight and height), but only 12% reported that their child's learning and behavior had been evaluated (16). When medical professionals themselves were asked about barriers to developmental screening, 51.4% of them indicated that they do not know what instruments to use and that instruments were lacking, and 62.9% reported lack of trained personnel to do the screening. In addition, when early intervention service providers were asked about screening their clients, only 28% reported conducting screening at all, and only 15% listed a specific instrument that they used. Among the used listed instruments were the Ages and Stages Questionnaire (ASQ; 18), Denver Developmental Screening Test-II (19), Modified Checklist for Autism in Toddlers, Revised (M-CHAT-R; 20), and Vasilka Manova-Tomova (21). In addition, according to the report, 38% of professionals have no experience working with children under the age of 3 years (16). In addition, 44% of surveyed kindergarten and nursery staff reported having no experience working with children with developmental disorders or disabilities. Overall, early intervention is extremely limited in the country and many screening and assessment instruments have yet to be normed and validated.

1.5 Current study

Regardless of these reports and evaluations, there is still a need for a systematic investigation of the needs and priorities of families of children with developmental disorders, delays, and disabilities, and how the system of services interacts with their demographic characteristics and diagnosis. The present study addressed this gap in past research by collecting an online caregiver needs survey from parents of children with developmental disorders in Bulgaria. The study had three aims: (1) to characterize the needs of children with developmental disorders in Bulgaria; (2) to compare the needs and access to services of children with ASD and other neurodevelopmental disorders; (3) to examine the daily burden of their caregivers and how it varies based on their demographic characteristics, such as income and education.

2 Materials and methods

2.1 Survey

The survey used in the present study was adapted from the Caregiver Needs Survey developed by Autism Speaks (22). The aim of the original survey was to assess the needs of families, who have a child with a confirmed ASD diagnosis. For the present purposes, the survey was adapted for use with families of children with a confirmed diagnosis of developmental delays, disorders, and/or disabilities more broadly. We acknowledge that by including only children who have a confirmed diagnosis we are not able to assess the needs of families who are unable or unwilling to seek diagnostic services. Assessing the needs of such families would require a very different methodological and recruitment approach. Nevertheless, this study is a first step toward understanding the family needs of children with neurodevelopmental disorders in Bulgaria. The survey consisted of three parts.

In ***Part 1,*** we collected demographic information about the respondent, including age, sex, relationship to the child, education, employment status, marital status, and household income. Similar information was also collected about the respondent's partner, if applicable.

In ***Part 2,*** we collected information about the child's characteristics and the caregiver's first concerns. Questions covered the reasons why the caregiver sought a diagnosis, the age of diagnosis, and the current diagnosis of the child. We also asked caregivers about what they thought caused their child's condition (e.g., genetic predisposition, vaccines, act of God, traumatic experiences in early childhood, unknown, etc.).

In ***Part 3,*** we collected information about service encounters and the caregiver's needs and perceptions. Questions covered whether in the past year the child received medical and counseling services and if not, why. We collected information about whether the child attended kindergarten or school, what type, and whether the school knew about their diagnosis. Caregivers were asked about the availability of parent trainings and workshops in their region and about whether they experienced difficulty in finding information about their child's condition. We also asked caregivers to pick the 3 main challenges associated with their child's condition, the three main challenges associated with their access to care, and the three main priorities when receiving support.

At the end of the survey, we added a comment box, not included in the original Caregiver Needs Survey, in order to give respondents the opportunity to share their experiences that might not have been reflected in the survey questions. The prompt for it was as follows: "Comments, recommendations, questions (e.g., What kind of professionals do you need access to? What should the main priorities be when changing current policy related to ensuring services for children with developmental disorders and their families?)."

2.2 Preparation of the Bulgarian version

The survey was first translated into Bulgarian by a licensed translator. The translation was then reviewed and adapted to the Bulgarian context by two clinicians, who work with children with developmental disorders and their families, and two researchers with experience in developing instruments. This adapted version was then presented to two separate focus groups. The focus groups were facilitated by a clinician and a researcher and consisted of five primary caregivers of children with developmental disorders (primarily ASD) of varying age. The focus groups were presented with the survey questions verbally and were asked to provide feedback. Based on the received feedback, the wording of some questions and their associated responses was changed to better reflect the experiences of caregivers. In particular, more options were included in the questions about caregiver's employment status to reflect individual experiences of being the primary caregiver, as well as working full-time. Yearly income was changed to monthly income. For the caregivers' perceptions of the causes of their child's diagnosis, we added a medical/doctor's error/malpractice option. With regard to who encouraged the caregiver to seek a diagnosis, we added the option of "encouragement/recommendation from pre-K and kindergarten personnel" again based on the focus group feedback.

2.3 Procedure

In Bulgaria, there is no population-based sampling of children with developmental disorders, so a sample of convenience was used. Caregivers of children with developmental delays, disorders, and disabilities were recruited through listservs of service providers in the big cities around the country, through posts on NGO websites and parent groups on social media platforms, and through posts on news and media outlets (radio, newspapers, etc.). The survey was sent out in the form of a link.

Respondents had to read and fill out an online consent form prior to starting their participation. The survey took between 30 and 40 mins to complete. Data collection took place between April and July 2020.

The Research Ethics Committee of the Cognitive Science and Psychology Department at the New Bulgarian University approved this project prior to respondent participation.

2.4 Participants

While the survey was active online, 422 respondents opened it, 7 did not give consent to participate, and 98 gave consent but did not fill out any questions, which left us with a sample of 317 respondents. Out of them, 7 did not indicate their relationship to the child and 5 indicated that they were a clinician, resource teacher, and/or a medical professional working with children with developmental disorders. These 12 respondents were excluded, which left us with a final sample of 305 respondents.

2.5 Data analysis plan

The data from the survey was collected using SurveyMonkey (SurveyMonkey Inc., San Mateo, CA, United States). Data were then imported and analyzed using the Statistical Package for Social Sciences Version 26.0 (SPSS 26.0). Data analysis included descriptive statistics such as frequency distributions, and inferential statistics comparing responses across child diagnostic status and respondent's income and education as proxy for SES.

3 Results

3.1 Child diagnosis and groups

To categorize our sample by diagnosis, we asked respondents to report their child's most recent diagnosis, with the option of including more than one. Table 1 includes the distribution of diagnoses across children. Note that clinical diagnoses in Bulgaria are based on the International Statistical Classification of Diseases and Related Health Problems (ICD-10; 23). The most common diagnosis in our sample was autism (27.9%), followed by PDD (24.9%) and ASD (16.7%) as defined by the ICD-10. Under other/written in diagnoses, responses included hydrocephaly ($N = 4$), Asperger's syndrome ($N = 1$), childhood autism ($N = 1$), Coffin-Siris Syndrome ($N = 1$), Prader-Willi Syndrome ($N = 1$), Down Syndrome ($N = 1$), Rett Syndrome ($N = 1$), and dyslexia ($N = 1$) among others. We also examined the overlap across diagnostic categories. 63.9% of children in our sample had an autism diagnosis, including autism, autism spectrum disorder, pervasive developmental disorder, pervasive developmental disorder – not otherwise specified, Asperger's Syndrome, and/or childhood autism. The second most common category of diagnoses was a mix with 13.4% of children receiving multiple diagnoses across autism, ADHD, intellectual disability, cerebral palsy, and epilepsy. In terms of the distribution of total number of diagnoses in our sample, the majority of children (61%; $N = 186$) had a single diagnosis, 18.7% ($N = 57$) had two, and 8.1% ($N = 25$) had 3 or more.

For the purposes of the following analyses, we divided participant responses into two groups based on child diagnosis. The first group, hereby called ASD Group, comprises respondents ($N = 182$), whose child had an autism and/or related diagnosis including childhood autism, autism spectrum disorder, pervasive developmental disorder, pervasive developmental disorder – not otherwise specified, and/or Asperger's Syndrome. The second group, hereby called oNDD Group, comprises of respondents ($N = 74$), whose child had any of the other neurodevelopmental disorders including ADHD, intellectual disability, cerebral palsy, epilepsy, different syndromes, or multiple diagnoses spanning across these categories. Children who received both an ASD diagnosis and a diagnosis that belonged to the oNDD category were classified as oNDD. This was done because we hypothesized that a child who had an ASD and an additional NDD diagnosis, for example, epilepsy or cerebral palsy, would have different needs and would try to access different services as compared to a child who only had an ASD diagnosis.

TABLE 1 Distribution of diagnoses and total number of diagnoses in the full participant sample ($N = 305$).

Characteristic	*N*	% Out of total
Diagnosis (could choose more than one)		
Autism	85	27.9
Pervasive Developmental Disorder (PDD)	76	24.9
Autism Spectrum Disorder (ASD/PAC)	51	16.7
Pervasive Developmental Disorder – Not Otherwise Specified (PDD-NOS)	35	11.5
Developmental delay	29	9.5
Intellectual disability	28	9.2
ADHD	23	7.5
Written in diagnosis	23	7.5
Cerebral palsy	22	7.2
Epilepsy	15	4.9
No diagnosis	11	3.6
Did not respond	27	8.9
Distribution of diagnoses		
Autism, ASD, PDD, PDD-NOS, DD, Asperger, Childhood Autism	195	63.9
Developmental delay	5	1.6
ADHD	8	2.6
Intellectual disability	2	0.7
Cerebral palsy	11	3.6
Epilepsy	1	0.3
Across categories – combined	41	13.4
Other	10	3.3
No diagnosis	10	3.3
Did not respond	27	8.9
Number of diagnoses		
0	37	12.1
1	186	61.0
2	57	18.7
3	15	4.9
4	8	2.6
5	1	0.3
6	1	0.3

3.2 Needs of the child

First, we focused on characteristics that could directly affect the child's needs and access to services including family demographic information and child characteristics.

3.2.1 ASD group

3.2.1.1 Family characteristics

The majority of respondents were female (94%) between the ages of 36 and 45 (56%) and identified themselves as the biological mother (89%) and primary caregiver (89.6%) of the child for whom they were filling out the survey (see Table 2). In terms of race and ethnicity, which was operationalized as native language, the vast majority of respondents (98.9%) identified as Bulgarian.

When characterizing the families' socio-economic status (SES), we looked at the education of the respondent and the family monthly income (in leva). The majority of respondents had an undergraduate degree or higher (79.1%). Even though the caregiver education was skewed toward higher attained degrees, the household monthly income resembled a normal distribution with 39% of respondent earning 1.220 lv, which is at or below country average (1.148 lv; 24), 26.4% earning between 1.220 and 1830 lv per month, and 28.6% earning over 1.830 lv per month. In terms of employment, 39.6% reported working full-time, and 17% reported working part-time.

3.2.1.2 Child characteristics

We did not collect age and gender information on the children. However, we have information about the children's kindergarten or school enrollment. Based on it, 30.8% ($N = 56$) children were under the age of 6 and 58.2% ($N = 106$) of the children were school-age or older than 6 years (see Table 3). Next, we examined caregivers' reasons for first concern regarding their child's development. 75.3% of caregivers chose communication difficulties, 66.5% chose social challenges, and 58.2% chose restricted and repetitive behaviors (see Table 3). Respondents were the ones to first notice and express concern about their child's development in 67% of cases, followed by other family members (12.6%), and spouses (4.4%). The majority of children (58.2%) were between 12 and 24 months at the time when the first concern about their development was noticed/expressed. 13.2% of children were under the age of one and 28.6 were between 2 and 6 years of age when someone expressed a concern about them.

With regard to children's diagnosis, 58.2% received a diagnosis by the age of 3 years, and additional 37.9% received a diagnosis by the age of 8 years, with less than 5% receiving a diagnosis past that age. The most common factors that lead to the caregiver seeking a diagnosis for their child were that the child's symptoms worsened (41.2%) and that family members or friends encouraged it (30.8%), followed by encouragement from a kindergarten teacher (14.3%). Interestingly, 18.1% of parents selected "other reasons" for pursuing a diagnosis for their child, and the majority of them wrote in their response explaining that it was their desire to help their child develop and achieve their full potential that encouraged them.

There were no significant associations between caregiver education or income with age of first concern or with age of diagnosis.

TABLE 2 Caregiver demographic characteristics across the participant groups.

	ASD group $N = 182$		oNDD group $N = 74$	
Characteristics	N	Valid%	N	Valid%
Gender				
Female	171	94.0	74	100
Male	11	6.0	–	–
Age (in years)				
18–25	3	1.6	–	–
26–35	33	18.1	17	23.0
36–45	102	56.0	38	51.4
46–55	37	20.3	18	24.3
56–65	1	0.5	–	–
>65	6	3.3	1	1.4
Relationship to child				
Biological mother	162	89.0	68	91.9
Biological father	9	4.9	–	–
Adoptive mother	3	1.6	3	4.1
Foster parent	0	0	2	2.7
Grandparent	5	2.7	1	1.4
Other family member	3	1.6	–	–
Primary caregiver of the child				
Yes	163	89.6	69	93.2
No	19	10.4	5	6.8
Native language				
Bulgarian	180	98.9	73	98.6
Other	2	1.1	1	1.4
Family status				
Married or living together	152	83.5	65	87.8
Widowed, divorced or separated	17	9.3	3	4.1
Single	13	7.1	5	6.8
Did not respond	–	–	1	1.4
Education *Simplified*				
School education	38	20.9	9	12.2
Undergraduate (classes and/or degree)	57	31.3	33	44.6
Graduate degree	87	47.8	32	43.2
Income *Simplified*				
<1220l v	71	39.0	27	36.5
1220–1830 lv	48	26.4	20	27.0
>1830 lv	52	28.6	23	31.1
Did not respond	11	6.0	4	5.4
Employment *Simplified*				
Full-time	72	39.6	27	36.5
Part-time	31	17.0	13	17.6
Other	77	42.3	34	45.9
Did not respond	2	1.1	–	–

TABLE 3 Caregiver-reported child characteristics, reasons for first concern, and diagnosis information.

Question	ASD group (*N* = 182)		oNDD group (*N* = 73)	
	N	%	*N*	%
Type of School/Kindergarten				
Kindergarten	56	30.8	18	24.3
State School	56	30.8	22	29.7
Private School	4	2.2	5	6.8
Specialized School	22	12.1	13	17.6
Not enrolled	11	6.0	8	10.8
Other	9	4.9	3	4.1
Did not respond	24	13.2	5	6.8
Proxy for age				
Under 6	56	30.8	18	24.3
Over 6	106	58.2	45	60.8
Unknown	20	11.0	11	14.9
Reasons for first concern				
Communication difficulties	137	75.3	39	52.7
Social challenges	121	66.5	18	24.3
Repetitive behaviors	106	58.2	13	17.6
Motor difficulties	35	19.2	42	56.8
Medical difficulties	36	19.8	23	31.1
Behavioral difficulties	51	28.0	24	32.4
First to notice the concern				
I was	122	67.0	43	58.1
My spouse/domestic partner	8	4.4	3	4.1
Other family member	23	12.6	6	8.1
Health care provider	11	6.0	16	21.6
Teacher	9	4.9	4	5.4
Other	9	4.9	2	2.7
Child's age when first concern was noticed				
0–3 months	4	2.2	22	29.7
3–6 months	4	2.2	9	12.2
6–12 months	16	8.8	13	17.6
12–18 months	63	34.6	11	14.9
18–24 months	43	23.6	6	8.1
24 months–3 years	40	22.0	9	12.2
3–6 years	12	6.6	3	4.1
7–12 years	–	–	1	1.4
Child's age at diagnosis				
<1 year old	–	–	34	45.9
1–3 years	106	58.2	25	33.8
4–8 years	69	37.9	14	18.9
9–12 years	4	2.2	1	1.4
13–17 years	–	–	–	–
>18 years	1	0.5	–	–
No diagnosis	2	1.1	–	–
Primary factor to pursue diagnosis				
Symptoms worsened	75	41.2	33	44.6
Encouragement from family members and friends	56	30.8	15	20.3
Encouragement from kindergarten teacher	26	14.3	10	13.5
Encouragement from community leaders to seek medical evaluation	6	3.3	3	4.1
Public service announcement that encouraged pursuit of medical advice	2	1.1	–	–
Previously unavailable healthcare services became available	4	2.2	2	2.7
Other (written in)	33	18.1	21	28.4
Number of diagnoses				
1	150	82.4	32	43.2
2	30	16.5	23	31.1
3	2	1.1	10	13.5
4	–	–	7	9.5
5	–	–	1	1.4
6	–	–	1	1.4

3.2.2 oNDD group

3.2.2.1 Family characteristics

All family demographic characteristics for the oNDD group can be found in Table 2.

3.2.2.2 Child characteristics

All child characteristics can be found in Table 3.

When examining how family income and caregiver education were related to the diagnostic experiences, no significant associations between caregiver income with age of first concern or age of diagnosis were found. There was a statistically significant association between caregiver education and child age of diagnosis, however. Parents with a higher attained degree were more likely to have a child who was diagnosed earlier [$\chi^2(6, N = 74) = 19.39, p = 0.004$].

3.2.3 ASD vs. oNDD comparison

When comparing the ASD and oNDD Groups, there were no differences between groups in terms of demographic characteristics such as parent education, monthly income, or employment status. There were some group differences in child characteristics, however. Caregivers in the ASD Group were more likely to choose communication difficulties [$\chi^2(1, N = 256) = 12.48, p < 0.001$], social challenges [$\chi^2(1, N = 256) = 37.68, p < 0.001$], and repetitive behaviors [$\chi^2(1, N = 256) = 34.99, p < 0.001$] as reasons for first concern, while caregivers in the oNDD group were more likely to pick motor difficulties [$\chi^2(1, N = 256) = 35.23, p < 0.001$]. Furthermore, there was a marginally significant difference in who expressed first concern about the child across the diagnostic group with caregivers in the oNDD Group being more likely to select a medical professional than parents in the ASD group [$\chi^2(5, N = 256) = 14.36, p = 0.013$].

There was a statistically significant association between diagnostic group and age of first concern as well [$\chi^2(7, N = 256) = 72.53, p < 0.001$]. Caregivers in the oNDD Group were more likely to notice a first concern about their child's development when the child was younger compared to caregivers in the ASD group. A similar pattern was observed with regard to child's age at diagnosis [$\chi^2(5, N = 256) = 97.04, p < 0.001$]. There were no group differences in reasons to pursue a diagnosis.

3.3 Daily burden of caregiver

Next, we focused on factors that contribute to the daily burden of caregivers, including caregiver knowledge and accessibility to treatments and services.

3.3.1 ASD group

When asked about their beliefs about the causes of their child's condition, 36.8% of caregivers reported that there were no known causes, 33.5% selected vaccinations as the cause, and 9.9% selected genetics or hereditary causes, while the remaining responses spanned across act of God, traumatic experiences early in life, bad luck, and medical error (see **Table 4**). There was a statistically significant association between caregivers' beliefs about the causes of their child's condition and their education [$\chi^2(16, N = 182) = 31.18, p = 0.013$]. Based on visual inspection of the distribution of responses across the three education categories, it appears that the caregivers, who had attained higher education degrees were more likely to select "unknown causes" and "genetic causes," while the distribution across the "vaccines" response was similar across education sub-groups. There was no association between beliefs and family income.

In terms of caregiver burden related to diagnosis, the majority of children (66.5%) received a diagnosis within their own town/village and additional 31.3% within the country. In terms of how long they had to wait from the initial pursuit of diagnosis to its ultimate confirmation, there was an even distribution across choices: 22% reported they waited less than a month, 29.1% reported between 1 and 3 months, 24.7% reported between 3 and 6 months, 9.3% between 6 months and a year, and 13.7% – over a year. The majority of caregivers (89.6%) had only one child with a developmental disorder in their household.

When reporting on their service experiences, 46.7% caregivers indicated that their family did not experience any difficulties or delays accessing medical services. However, 21.4% reported delays due to long wait times, 17% - due to lack of information about services, 10.4% - due to unreasonable costs, 17% – no services available in general or in their area, and 7.1% because of ineligibility. A similar pattern of responses was observed with regard to encounters with counseling services and educational services (see **Table 4**). There were no significant associations between caregivers' education and their experienced delays and/or difficulties accessing medical, counseling, or educational services. There was a significant association between family income and access to educational services, however [$\chi^2(2, N = 182) = 7.66, p = 0.022$]. Caregivers with lower income were more likely to experience delays in educational resources.

When asked about the availability of local centers in their region providing trainings for parents, only 31.3% of respondents indicated that there were such. In addition, the majority of caregivers (76.4%) indicated that they had experienced difficulty in finding accurate and helpful information about addressing their child's needs with 39.6% choosing that it was somewhat difficult, 19.2% that it was very difficult, and 17% that it was extremely difficult. There was no significant association between parent education and caregivers' difficulty finding information.

We asked parents about their top three greatest challenges to caring for their child (see **Table 4**). The most common challenge picked by 56.6% of caregivers was social interaction difficulties, including difficulty making friends, and not being able to read social cues. This was followed by communication difficulties (48.9%), problems with daily living skills (39%), troubling behaviors (36.3%), and repetitive behaviors/limited interests (35.7%). Close to one-fifth of caregivers reported challenges with safety concerns (24.2%), their child's sensory issues (22.5%), and sleep problems (19.2%).

When asked about caregivers' greatest challenges in getting support for their child, the majority of respondents (63.2%) picked adequate education, followed by adequate counseling help (47.3%), and making sure that the child's basic rights are protected (40.1%). Among the other popular choices were receiving financial support (35.7%) and adequate support from the community (29.7%). There were no significant associations between caregivers' top three problems and top three challenges with their education or income.

TABLE 4 Caregiver-reported beliefs, paths to diagnosis, and access to services.

	ASD group (*N* = 182)		oNDD group (*N* = 74)	
Question	*N*	%	*N*	%
Beliefs about the causes of their child's condition				
Act of God or supreme being	1	0.5	2	2.7
Traumatic experiences early in life or in womb	16	8.8	15	20.3
Genetics or hereditary in some families	18	9.9	13	17.6
Cold, rejecting parents	–	–	3	4.1
Vaccinations	61	33.5	10	13.5
Bad luck or a curse from one's past life	3	1.6	1	1.4
Medical error/malpractice	7	3.8	8	10.8
No known causes	67	36.8	14	18.9
Other	8	4.4	7	9.5
Did not respond	1	0.5	1	1.4
Number of children with dev. disorder				
None	5	2.7	2	2.7
One	163	89.6	69	93.2
Two	11	6.0	3	4.1
Three or more	1	0.5	–	–
Did not respond	2	1.1	–	–
Number of adults with disorder				
Yes	19	10.4	9	12.2
No	163	89.6	65	87.8
How far did you have to travel to obtain a diagnosis?				
Within my town/village	121	66.5	48	64.9
A few towns/villages away	26	14.3	11	14.9
Had to travel into another province	31	17.0	10	13.5
I traveled outside the country	3	1.6	5	6.8
Did not respond	1	0.5	–	–
How long did you have to wait between your initial pursuit of a diagnosis and the ultimate confirmation of a clinical diagnosis for your child?				
Less than a month	40	22.0	30	40.5
1–3 months	53	29.1	28	37.8
3–6 months	45	24.7	5	6.8
6–12 months	17	9.3	1	1.4
Over a year	25	13.7	7	9.5
We still have not received a diagnosis	2	1.1	3	4.1
During the past year, did your family have any difficulties or delays in getting medical services for your child for any of the following reasons?*				
There were no difficulties or delays	85	46.7	37	50.0
Delayed because of ineligibility	13	7.1	7	9.6
Delayed because of lack of information about services	31	17.0	10	13.5
Delayed because of lack of services in our area	15	8.2	8	10.8
Delayed because of long wait times	39	21.4	13	17.6

(Continued)

TABLE 4 (Continued)

	ASD group ($N = 182$)		oNDD group ($N = 74$)	
Question	N	%	N	%
Delayed because of unreasonable costs	19	10.4	7	9.5
No services available	16	8.8	9	12.2
Other	9	0.5	5	6.7
Has your child ever received counseling services to meet his/her behavioral needs?				
Yes	153	84.1	65	87.8
No	4	2.2	2	2.7
I do not know	3	1.6	1	1.4
Did not respond	22	12.1	6	8.1
During the past year, did your family have any difficulties or delays in getting counseling services for your child for any of the following reasons?*				
There were no difficulties or delays	103	56.6	39	52.7
Delayed because of ineligibility	7	3.8	6	8.1
Delayed because of lack of information about services	12	6.6	7	9.5
Delayed because of lack of services in our area	10	5.5	7	9.5
Delayed because of long wait times	16	8.8	3	4.1
Delayed because of unreasonable costs	18	9.9	9	12.2
No services available	9	4.9	7	9.5
Other	9	4.9	5	6.8
Does your child's school offer any additional academic support for children with developmental disabilities (such as tutors or resource teachers)?				
Yes	120	65.9	47	63.5
No	22	12.1	14	18.9
I do not know	6	3.3	7	9.5
Did not respond	34	18.7	6	8.1
During the past year, did your family have any difficulties or delays in getting educational services for your child for any of the following reasons?*				
There were no difficulties or delays	98	53.8	45	60.8
Delayed because of ineligibility	8	4.4	4	5.4
Delayed because of lack of information about services	7	3.8	5	6.8
Delayed because of lack of services in our area	5	2.7	3	4.1
Delayed because of long wait times	13	7.1	1	1.4
Delayed because of unreasonable costs	5	2.7	1	1.4
Delayed because the kindergarten/school did not want to enroll my child	22	12.1	5	6.8
Other	14	7.6	9	12.2
Are there any local service centers that specialize in teaching parents the best ways to manage and support the needs of their children?				
Yes	57	31.3	19	25.7
No	59	32.4	32	43.2
I do not know	44	24.2	17	23.0
Did not respond	22	12.1	6	8.1

(Continued)

TABLE 4 (Continued)

	ASD group ($N = 182$)		oNDD group ($N = 74$)	
Question	*N*	%	*N*	%
If so, do you currently use any of these services to learn more about managing and addressing the needs of your child?				
Yes	43	23.6	14	18.9
No	47	25.8	22	29.7
I do not know about such services	44	24.2	19	25.7
Did not respond	48	26.4	19	25.7
In the time since learning about your child's disability, how difficult have you found it to obtain accurate and helpful information on the best ways to address your child's needs?				
Not difficult at all	21	11.5	4	5.4
Somewhat difficult	72	39.6	30	40.5
Very difficult	35	19.2	21	28.4
Extremely difficult	31	17.0	13	17.6
I have not tried to obtain information	1	0.5	–	–
Did not respond	22	12.1	6	8.1
What do you consider to be the greatest challenges to caring for a child with developmental difficulties? *Please select the top three challenges from the list below.*				
Troubling behaviors (e.g., self-injury, aggression, tantrums)	66	36.3	29	39.2
Daily living skills (e.g., using the bathroom, dressing themselves, feeding themselves)	71	39.0	40	54.1
Health problems (e.g., mental/physical health illnesses occurring alongside your child's disability)	14	7.7	21	28.4
Sleep problems (e.g., trouble falling asleep, trouble staying asleep)	35	19.2	17	23.0
Diet/eating difficulties	42	23.1	5	6.8
Social interaction difficulties (e.g., has difficulty making friends, can't read social cues)	103	56.6	32	43.2
Repetitive behaviors/limited interests/insistence on sameness	65	35.7	15	20.3
Communication difficulties (e.g., cannot explain their needs, cannot express emotions)	89	48.9	23	31.1
Safety concerns (e.g., getting in trouble with police, neighbors, strangers)	44	24.2	11	14.9
Sensory issues (sensitivity to certain sounds or lights)	41	22.5	16	21.6
Other (Please specify: ______)	6	3.3	9	12.2
What are the greatest challenges you face in getting support for your child? *Please select the top three challenges from the list below*				
Making sure my child receives adequate medical help	50	27.5	30	40.5
Making sure my child receives adequate education	115	63.2	45	60.8
Making sure my child receives adequate counseling help	86	47.3	25	33.8
Making sure my child's basic rights are protected	73	40.1	25	33.8
Making sure my family and I receive adequate financial support	65	35.7	25	33.8
Making sure my family and I receive adequate support from the community	54	29.7	25	33.8
Other (Please specify: ________________)	7	3.8	4	5.4

(*Continued*)

TABLE 4 (Continued)

	ASD group ($N = 182$)		oNDD group ($N = 74$)	
Question	N	%	N	%
What do you consider to be the greatest priorities for families affected by developmental disabilities in your country? *Please select the top three priorities* from the list below.				
Improved medical services	53	29.1	31	41.9
Improved education services	118	64.8	45	60.8
Improved counseling services	85	46.7	31	41.9
Greater rights for individuals with disabilities	63	34.6	27	36.5
Greater protection of existing rights for individuals with disabilities	56	30.8	16	21.6
More information about autism/developmental delay	75	41.2	16	21.6
Greater in-home support	24	13.2	12	16.2
Greater community awareness	81	44.5	33	44.6
Greater financial support for the family	77	42.3	23	31.1
Other (Please specify: ______)	–	–	3	4.1

The symbol * means that participants could choose more than one answer.

In terms of greatest priorities for families affected by developmental disorders in Bulgaria, the majority of respondents in the ASD group (64.8%) similarly picked improved education services, followed by improved counseling services (46.7%), and greater community awareness (44.5%). These top three choices were followed by greater financial support for the family (42.3%), more information about the child's condition (41.2%), and greater rights for individuals with disabilities (30.8%). There was no significant association between caregivers' education and family income with the likelihood of choosing any of the priorities.

3.3.2 oNDD group

See Table 4 for caregivers' beliefs about the causes of their child's condition, distanced traveled and wait time to receive an official diagnosis, and delays in getting medical, counseling, and educational services. There were no associations between caregivers' education and their beliefs. There were no associations between caregiver education and family income and access to services or information.

See Table 4 for the top three challenges to caring for a child with a developmental disorder, the top three challenges to getting support for the child, and the top three greatest priorities for families. There were no associations between the top three challenges to care and the top three priorities with caregiver education and family income.

3.3.3 ASD vs. oNDD comparison

Caregivers' beliefs about the causes of their child's condition differed across groups, [$\chi^2(9, N = 256) = 37.780, p < 0.001$]. Specifically, caregivers in the ASD group were more likely to select "vaccines" or "unknown causes" than caregivers in the oNDD group.

There were no statistically significant differences between diagnostic groups and distance traveled to receive a diagnosis. However, there was an association between diagnostic group and the wait time between initial pursuit of diagnosis and the confirmation of the diagnosis [$\chi^2(5, N = 256) = 24.488, p < 0.001$] with oNDD group receiving a diagnosis more quickly.

There were no significant differences between groups in service encounters, including access to and delay of medical, counseling, and educational services, and access to parent training and information.

Some differences emerged, however, in top three challenges to caring for a child with developmental difficulties. Specifically, the oNDD group was more likely to select "health problems" [$\chi^2(1, N = 256) = 19.074, p < 0.001$]. Furthermore, the ASD group was more likely to select "diet" as a problem [$\chi^2(1, N = 256) = 9.349, p = 0.002$].

There were no group differences in top three challenges to care, and there was only one group difference in top three priorities. The ASD group was more likely to select "more information about autism/developmental delay" than the oNDD group [$\chi^2(1, N = 256) = 8.810, p = 0.003$].

3.4 Open responses

The very last question on the survey was a comment box asking respondents to provide any comments, suggestions, and/or recommendations about what services should become available, but also about what the main priorities should be when changing policy related to families affected by developmental disorders. Out of the 317 respondents, 118 (37.2%) filled out the comment box. Responses ranged from a list of specialists that the caregiver wanted available for their child to paragraph-long discussion of the situation in Bulgaria and how the respondent envisioned existing policy should change.

We went through the responses and based on their content, categorized them into 8 distinct categories: "specialized services and support," "access to services," "education," "financial support," "community awareness," "psychological and family support," "protection of the rights of individuals with disabilities," and "other." The order of these categories reflects the number of responses classified under them, with "specialized services and support" containing the highest number of responses (see **Figure 1**). Each category is described in **Table 5**.

4 Discussion

This is one of the first studies collecting primary data from families of children with developmental disorders in Bulgaria aimed at characterizing their needs and daily burdens. This study has three main findings: (1) children with ASD and children with oNDD in Bulgaria have different needs and paths to diagnosis; (2) nevertheless, children in both groups experience similar challenges to accessing medical, counseling, and educational services, regardless of their demographic characteristics; and (3) parents' priorities focus on education, counseling and medical support, protecting children's basic rights, and raising awareness. Based on our findings, we provide specific recommendations for changes in services and policy.

4.1 Comparison of ASD and oNDD children's needs and paths to diagnosis

Overall, the ASD group and the oNDD group did not differ in terms of demographic information. The majority of respondents were female between 36 and 45 years of age, primary caregivers and biological mothers of the child for whom they filled out the questionnaire. Our sample was highly educated with over 75% of respondents having received an undergraduate or a graduate degree compared to only 26.1% of Bulgarian population over the age of 25 years in 2020 that had attained at least a Bachelor's degree (25). We attribute this characteristic of our sample to our recruitment strategy, namely relying on parent groups online where parents proactively seek out advice and try to discuss treatment and intervention practices. The higher education level of our respondents might introduce some bias to our results considering that the sample is not representative of the general population when it to comes to educational attainment. Nevertheless, there was more variability in terms of family income, where over one-third of respondents reported income lower than the country average monthly wage (24). Perhaps, this can be linked to our respondents' employment status (46.6% were employed full-time or part-time), which was much lower than the 94% employment rate for the country in 2020 for people between the age of 20 and 64 years (24). The lower employment rate and lower income of our respondents but higher education than the national average could potentially be attributed to the fact that the majority of our respondents are the primary caregiver of their children with a developmental disorder, and the caregiving demands might be impacting their employment. Even though the two participant groups were comparable in demographic characteristics, they differed in terms of the characteristics of the children, their needs, and paths to diagnosis. Specifically, there were differences in caregivers' reasons for first concern. The oNDD parents noticed motor difficulties first, while ASD parents were more likely to notice communication difficulties, social challenges, and restrictive and repetitive behaviors first. In addition, oNDD parents were more likely to express concern about the development of their child earlier, with 58% of them expressing first concern in the first year of the child's life in comparison to only 16% of ASD parents. These differences in reasons for first concern and when it was expressed could be attributed to the different defining characteristics of the conditions across the two participant groups. Specifically, some oNDD and genetic syndromes have symptoms that might present immediately after birth. In contrast, differences in repetitive behaviors, cognition, and language between children with and without later ASD diagnosis have been found later by 14–16 months of age (26). Furthermore, physical/motor difficulties are perhaps easier to notice for parents without a training in early development than more subtle social and communication challenges that would start to affect the child's behavioral repertoire later (as their communication abilities develop). In past studies, parents from the United States and the United Kingdom expressed first concern for their child by 14—15 to 19 months of age (27–29). In our ASD sample, first concerns were expressed somewhat later with 82.2% of ASD parents expressing first concern by 24 months. However, our results are comparable to what was reported for Bulgarian parents in the past (4) with a mean age of first concern of 24.7 months and for Serbian parents with a mean age of 22.5 months (30). Efforts to improve awareness about child development and expected developmental milestones among parents could help lower the age of first concern.

The later age of first concern, in turn, is expected to have cascading effects on the age of diagnosis. Indeed, oNDD children were more likely to receive a diagnosis earlier with 45.2% receiving it within first year of life as compared to only 1% of ASD children. oNDD children not only were diagnosed earlier, but they received a diagnosis faster with 79.5% of them waiting

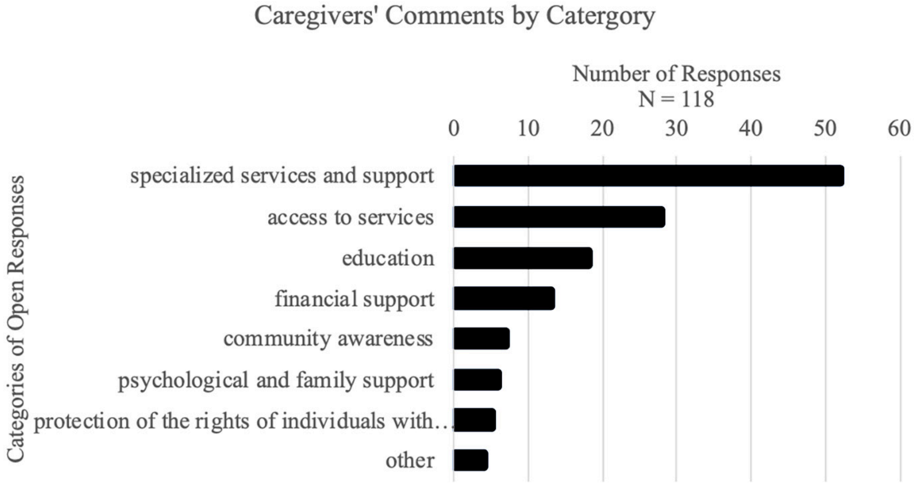


FIGURE 1
Distribution of caregivers' comments by category.

TABLE 5 Topics in open-ended comment box at the end of the survey.

Number of responses	Topic	Details
52	Specialized services and support	All responses that discussed the specific medical professionals and clinicians that the respondent considered lacking in the country. Common examples include SLPs, psychologists, occupational therapist, ABA therapist, etc. These responses often included exhaustive lists of medical professionals.
28	Access to services	All comments in which respondents wrote about how they envisioned they should receive services. Specifically, they described day centers that would provide all necessary services rather than having to take their child from professional to professional around the city. Another common theme was a consistent and individualized approach to each child across all service providers that they work with.
18	Education	All responses that talked about the need for better trained tutors and resource teachers, as well as the need to come up with a specialized curriculum for children with developmental disorders rather than following the curriculum of their typically developing peers.
13	Financial support	All responses that discussed the need for financial support for affected families, as well as the possibility of making services free or available at a reasonable cost. Some responses also mentioned the need to increase salaries of specialists providing services to children with developmental disorders.
7	Community awareness	All responses discussing the need for raising community awareness about developmental disorders. The responses described raising awareness both among peers and parents, but also among teachers and medical professionals.
6	Psychological and family support	All responses discussing the need for services and resources for parents/families, including support groups, resources for families right after they receive their child's diagnosis, and struggling with loneliness.
5	Protection of the rights of individuals with disabilities	All responses that talked about the need for better 'protection of the rights of individuals with disabilities.' Responses mentioned the need for the "archaic system" to adapt to the needs of the children rather than the other way around, and for the need of the "country" to protect these children as they transition to adulthood.
4	Other	All responses that did not fall into any of the other categories.

Note that a comment could be classified under more than one category. The total number of comments was 118.

less than 3 months from first attempt in comparison to 50.3% of ASD children who waited less than 3 months. What could account for the different wait times to diagnosis across the ASD and oNDD groups? On the one hand, it could be the length of the diagnostic procedures that is different with some oNDD conditions requiring genetic testing and EEG testing, while an ASD diagnosis requires a more extensive battery of behavioral assessments over an extended period of time.

Relatedly, the difference in wait times could be attributed to the number of specialists available and qualified to provide a diagnosis. In Bulgaria, only a child psychiatrist can provide a formal diagnosis of autism spectrum disorder based on the ICD-10. In contrast, conditions such as epilepsy, cerebral palsy, or genetic syndromes can be diagnosed by child neurologist and professionals specializing in genetic disorders. In comparison to past research, the average age of ASD diagnosis in North America is typically after 3 years of age and up to 5 and a half [(31) – 55.2 months; (32) – 38 months; (33) – 68 months; see (34) for review]. In Bulgaria over 5 years ago, the average age of ASD diagnosis was 46.6 months (4). These results are similar to the present study with 58.2% of ASD children receiving a diagnosis before 3 years and 37.9% between 4 and 8 years of age.

Another comparison to previously collected data in Bulgaria shows a relative improvement in diagnostic services. Based on data collected between 2013 and 2017, 50% of Bulgarian parents with ASD had to travel more than 100 km to receive a diagnosis and 17% had to travel between 25 and 100 km (4). In contrast, our results show that over 60% of parents across both the ASD and oNDD groups received a diagnosis within their own town or city. This improvement could be attributed to more diagnostic services becoming available or due to differences in the sampling of the two studies.

In summary, the diagnostic experiences of children with ASD and oNDD are different and so should be addressed accordingly. In particular, work needs to be done in raising awareness about developmental disorders, and in providing earlier and faster identification and diagnosis, especially for children with ASD.

4.2 Access to medical, counseling, and educational services

Regardless of the group differences in diagnostic experiences, both groups experienced similar delays in accessing services. Close to half of all children across both groups experienced delays in accessing medical, counseling, and educational services. These results support the evaluation of available services by the National Network for Children in their yearly report card (15). The reported reasons for the delays in access were somewhat evenly distributed across issues with eligibility, lack of information about such services, lack of existing services in the area, long wait times, and unreasonable costs. A notable exception was the long wait times associated with getting medical services, where 20.5% of children in the ASD group and 17.8% of children in the oNDD group experienced long wait times. Therefore, even though these children get diagnosed at different ages and for a different duration, when it comes to accessing services, they are similarly disadvantaged. The challenges associated with accessing services could be addressed by making more services available, thus reducing wait times, subsidizing costs, and generally promoting information about services and who is eligible for them. In addition, other efforts could include providing physicians with more information and training about developmental differences.

In addition to challenges associated with accessing services for their children, there were similar challenges across groups associated with access to parent trainings. In both the ASD and oNDD groups, less than one-third of parents (31.8% in the ASD group; 26% in the oNDD group) reported that there were any local centers teaching parents about the best ways to support the needs of their children. This issue of the lack of availability of parent trainings could have two related interpretations. On the one hand, the medical model of disability, which dominated the diagnostic process and educational services for children with developmental disorders and disabilities in the past (8), could account for the lack of services that are targeting the parents as the means for providing support for their children. This, in turn, could also lead to fewer parents even looking for such resources based on the expectation that it is only specialists who can support the development and functioning of their children. On the other hand, it could be the lack of parent training models available that are translated and adapted to the Bulgarian context that accounts for this finding. In fact, just recently the ImPACT training (35) was formally introduced in the country as part of the Stay-In Project co-funded by the Erasmus + Programme of the European Union (36). Data on parents' inclusion in already existing early intervention services showed that less than 10% of parents were present in the room with their child and actively trying out different treatment strategies (16), once again echoing the need for approaches engaging and training the parents. Engaging parents in applying treatment and intervention practices at home is perhaps the most scalable approach to reach the highest number of children, especially in contexts where there are not enough trained professionals and counseling services available.

Parents not only did not have access to parent training services but also reported difficulty finding helpful information on how best to address their children's needs. Specifically, 76.4% of ASD parents and 86.3% of oNDD parents found it somewhat difficult, very difficult, or extremely difficult to obtain useful information. Perhaps, this could partially be attributed to the fact that upon receiving a diagnosis, parents are not formally provided with guidance and materials about their child's condition and about what, where, and how to seek services. Parents, as the ones who spend most time with their children during childhood, have the potential to implement treatment and intervention strategies with their children on a daily basis. Supporting parents by providing them with freely available and accessible information about how to best promote their children's development is one very easy and cost-effective way to start addressing their needs and the general lack of services in low-resource contexts. In addition, raising awareness about developmental disorders and improving access to information could help improve caregivers' understanding of the etiology of developmental disorders, which should be a key goal considering that in our sample over 30% of parents selected vaccinations as the cause of their child's ASD diagnosis. When planning awareness campaigns, it is essential to not only present evidence-based advice but also to point out what the criteria are for a publication to be considered reliable and valid.

4.3 Parents' challenges and priorities

One way to move away from the strictly medical approach to developmental disorders and disabilities is to start addressing the social, communicative, and daily living needs of the children, as well as to treat them and their families as a unit. The third main finding of this study pertains specifically to the challenges, needs, and priorities of the families and could be used to build data-driven strategies to improve local services.

In our sample, parents were typically the first to notice and express concern about their child's development and it was their desire to help their child that motivated them to seek a formal diagnosis. Considering that parents are the ones who spend the most time with their children in early childhood, it would be beneficial to raise awareness among parents about typical developmental milestones in their children's functioning to lower the age at which parents express a first concern about their child's development. However, once the child has received a formal diagnosis, what are parents' greatest challenges to care? For the majority of parents in our sample (across both groups), the top three challenges to care were social interaction difficulties, communication difficulties, and daily living skills. Similar results were found from parents of children with ASD from Serbia (30) and from Low and Low Middle Income Countries (LMIC) in South America (37). Although social interaction and communication difficulties are often addressed in already available SLP and counseling services, parents in their open responses discussed the need for other services such as occupational therapy and applied behavioral analysis (ABA) therapy that could potentially improve the daily living skills of their children. Therefore, programs need to be developed that focus specifically on improving the daily functioning of children as it pertains to getting dressed, feeding, brushing their teeth, and doing chores. It should be pointed out that oNDD parents indicated that their children's health problems were a key challenge, while ASD parents selected their children's diet/eating difficulties. Furthermore, more oNDD parents experienced challenges in getting medical help for their children, which could be attributed to the nature of their diagnosis and associated medical conditions. In contrast, more ASD parents experienced challenges in receiving counseling help, again alluding to the different symptoms of the children, which translate into different needs. These differences in challenges should be accounted for in the way specific services and policies are made for children with ASD and children with oNDD.

In terms of parents' greatest priorities, over 60% of caregivers across both groups selected receiving adequate education. This key priority has been reported by parents in LMIC in Southeast Europe (4, 30) and in South America (37), as well as by parents in high-income countries (38). Education is one of the primary factors associated with positive long-term outcomes in children with ASD (39). Furthermore, education is perceived as children's path to socializing with their peers and to becoming independent. Parents valuing their children's education was further reflected in their open-ended responses, in which they discussed the need for better trained tutors and resource teachers, as well as for developing a specialized curriculum for children with developmental disorders. Therefore, even though progress has been made in education in Bulgaria over the last year (15), there is still more to be done to adequately support students with developmental disorders in the country.

In addition to education as a main challenge and a priority, caregivers also selected the protection of their children's basic rights and raising community awareness. Although community awareness has not been identified as a key priority for parents in high-income countries (39, 40), it has been identified as a key priority by over 40% of Bulgarian parents in parents our sample, as well as by parents from low- and middle-income countries in South America (37). Therefore, in addition to improving specific services for children with developmental disorders, there is a need to improve community awareness about developmental disorders. In their open-ended responses, caregivers discussed the need for raising awareness not only among other parents but also among teachers and medical professionals. A similar need for awareness has been found in relation to education suggesting that the majority of parents are not familiar with the needs of children with developmental disorders and thus are less likely to support their inclusion in the classroom (9). Raising community awareness will not only affect attitudes toward inclusive education but could also reduce stigma for families, which is frequently experienced by families of children with ASD (41, 42).

In addition to education, protection of children's basic rights, and raising community awareness, parents expressed other priorities. Specifically, caregivers envisioned that their children should receive services following an individualized and unified approach across professionals (SLP, counseling, OT, etc.). Furthermore, over one-third of all respondents identified the need for greater financial support, which is a reflection of the financial burden associated with raising a child with a developmental disorder. Therefore, future policy should also address the coordination between institutions and professionals working with the child, as well as the financial aspects associated with accessing services.

4.4 Limitations

Although very informative, the present study possesses a number of limitations. First, because there is no population-based sampling of children with developmental disorders in Bulgaria, we relied on a convenience sample. We recruited respondents on social media, through online parent forums and websites, and through listservs from centers providing services for children with developmental disorders. Furthermore, in the analyses, we only included families, whose child had a confirmed diagnosis of a developmental disorder. Therefore, our sample (1) might not be representative of the general population in the country, (2) could be subject to selection bias with more parents already seeking information and services for their children being more likely to participate, and (3) might not be representative of children who have not received a diagnosis yet or whose families are unwilling to seek one. Another limitation of our sample is that the caregivers of children with autism spectrum disorders were over-represented as they made up 73% of all respondents. Nevertheless, caregivers in the oNDD group had children with a wide range of neurodevelopmental disorders reflecting the heterogeneity of these conditions. Yet, another limitation is that the majority of respondents were the children's biological mothers and thus they might offer a different perspective on the needs of their children than other family members. Future studies should focus on sampling more caregivers of children with oNDD disorders to be able to investigate their specific needs based on diagnosis. Another limitation to our study is that children's diagnoses were not verified by a clinician, so we are relying on the accuracy of parent report. Furthermore, all responses were based on self-report and so could be subject to interpretation and be affected by the fact that parents were reflecting on past events.

Another limitation that needs to be addressed in the future is about children's characteristics, specifically age, sex, native language, etc. As of now, there are still no publicly available data on the number of children with developmental disorders in Bulgaria and their breakdown by diagnosis. Such information will help inform policy about the number of professionals and kinds of services that need to be made available to address the needs of these children and their families.

Data availability statement

The datasets presented in this article are not readily available because according to our Committee for Research Ethics application, we have not obtained permission to do that. Requests to access the datasets should be directed to MB, mihaela.barokova@gmail.com.

Ethics statement

The studies involving human participants were reviewed and approved by the Committee for Research Ethics of Department of Cognitive Science and Psychology at New Bulgarian University. The patients/participants provided their written informed consent to participate in this study.

Author contributions

MB and AA-S organized the data collection. MB and AK worked on data processing and analysis. MB drafted the first version of the manuscript. All authors participated in the conception and design of the study and contributed to the final written version of the manuscript.

Acknowledgments

We would like to thank all the families who took part in this study.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

References

1. Wallace S, Fein D, Rosanoff M, Dawson G, Hossain S, Bren Nan L, et al. A global public health strategy for autism spectrum disorders. *Autism Res.* (2012) 5:211–7. doi: 10.1002/aur.1236

2. National Health Insurance Fund. *Welcome to the Website of the National Health Insurance Fund!*. Sofia: National Health Insurance Fund (2022).

3. UNICEF. *UNICEF Launches a Campaign to Support Non-Verbal Children: The Data*. New York, NY: UNICEF (2020).

4. Daniels AM, Como A, Herguner S, Kostadinova K, Stosic J, Shih A. Autism in Southeast Europe: a survey of caregivers of children with autism spectrum disorders. *J Autism Dev Disord.* (2017) 47:2314. doi: 10.1007/s10803-017-3145-x

5. Levterova D. Social services for autistic children. *Trakia J Sci.* (2009) 8:406–10.

6. Koltcheva N. Research studies on child development and their relationship to social policy in the country [Научните изследвания на детското развитие и връзката им със социалната политика в държавата]. In: *Proceedings of the 3rd National Congress in Psychology Organized by the Bulgarian Psychological Association*. Sofia (2005).

7. Manolova H. Psychological Assessment, Therapy, and Training of Children With Autism Spectrum Disorder [Психологична оценка терапия и обучение на деца с разстройство от аутистичния спектър]. (2020).

8. Organisation for Economic Co-Operation and Development. *Education Policies for Students at Risk and Those With Disabilities in South Eastern Europe Bosnia-Herzegovina, Bulgaria, Croatia, Kosovo, FYR of Macedonia, Moldova, Montenegro, Romania and Serbia*. Paris: Organisation for Economic Co-Operation and Development (2006).

9. Center for Inclusive Education. *Attitudes Towards Inclusive Education* [Център за приобщаващо образование. Нагласи към приобщаващото образование]. Sofia: Center for Inclusive Education (2014). doi: 10.1108/S1479-363620140000003011

10. Ministry of Labour and Social Policy of Bulgaria. *Persons With Disabilities Act*. Sofia: Ministry of Labour and Social Policy of Bulgaria (2019).

11. Ministry of Labour and Social Policy of Bulgaria. *National Strategy for Children*. Sofia: Ministry of Labour and Social Policy of Bulgaria (2019).

12. Ministry of Labour and Social Policy of Bulgaria. *Social Services Act*. Sofia: Ministry of Labour and Social Policy of Bulgaria (2020).

13. Ministry of Education and Science. *Ordinance on Inclusive Education*. (2017). Available online at: http://www.sbubg.info/files/naredba_priobshavasho.pdf (accessed July 5, 2022).

14. Ministry of Education and Science. *Pre-School and School Education Act*. (2020). Available online at: https://www.mon.bg/bg/57 (accessed July 5, 2022).

15. National Network for Children. *Report Card 2020: What is the Average Government Score for Childcare?*. Sofia: National Network for Children (2020).

16. UNICEF. *Situational Analysis of Services for Young Children with Developmental Disorders and Disabilities in Bulgaria*. New York, NY: UNICEF (n.d.).

17. Ministry of Health. *Ordinance on Prophylactic Examinations and Dispanserization Services*. (2016). Available online at: https://www.nsoplb.com/uploads/docs/_8__03112016__________23112016_.pdf (accessed July 5, 2022).

18. Squires J, Bricker D. *Ages & Stages Questionnaires®, Third Edition (ASQ®-3): A Parent-Completed Child Monitoring System*. Baltimore, MD: Paul H. Brookes Publishing Co., Inc (2009). doi: 10.1037/t11523-000

19. Frankenburg WK, Dodds J, Archer P, Shapiro H, Bresnick B. The Denver II: a major revision and restandardization of the Denver Developmental Screening Test. *Pediatrics.* (1992) 89:91–7. doi: 10.1542/peds.89.1.91

20. Robins D, Fein D, Barton M. *Modified Checklist for Autism in Toddlers, Revised, With Follow-Up*. (2009). Available online at: https://mchatscreen.com/wp-content/uploads/2015/09/M-CHAT-R_F_Rev_Aug2018.pdf doi: 10.1037/t67574-000 (accessed July 5, 2022).

21. Manova-Tomova V. *Psychological Diagnosis in Early Childhood*. [Психологическа диагностика на ранното детство]. Belgrade: Prosveta (1974).

22. Autism Speaks. *Autism Speaks Global Public Health Initiative Website*. (2016). Available online at: https://www.autismspeaks.org/science/research-initiatives/global-autism-public-health (accessed Mar 27, 2021).

23. World Health Organization. *The ICD-10 Classification of Mental and Behavioural Disorders*. Geneva: World Health Organization (1993).

24. National Statistical Institute. *Statistical Data: Annual Data*. Sofia: National Statistical Institute (2020).

25. The World Bank. *Educational Attainment, at Least Bachelor's or Equivalent, Population 25+, Total (%) (Cumulative) – Bulgaria*. (2022). Available online at: https://data.worldbank.org/indicator/SE.TER.CUAT.BA.ZS?locations=BG (accessed July 4, 2022).

26. Landa R, Garrett-Mayer E. Development in infants with autism spectrum disorders: a prospective study. *J Child Psychol Psychiatry.* (2006) 46:629–38. doi: 10.1111/j.1469-7610.2006.01531.x

27. Chawarska K, Paul R, Klin A. Parental recognition of developmental problems in toddlers with autism spectrum disorders. *J Autism Dev Disord.* (2007) 37:62–72. doi: 10.1007/s10803-006-0330-8

28. De Giacomo A, Fombonne E. Parental recognition of developmental abnormalities in autism. *Eur J Child Adolesc Psychiatry.* (1998) 7:131–6. doi: 10.1007/s007870050058

29. Herlihy L, Knoch K, Vibert B, Fein D. Parents' first concerns about toddlers with autism spectrum disorder: effect of sibling status. *Autism.* (2015) 19:20–8. doi: 10.1177/1362361313509731

30. Pejovic-Milovancevic M, Stankovic M, Mitkovic-Voncina M, Rudic N, Grujicic R, Herrera AS, et al. Perceptions on support, challenges and needs among parents of children with autism: the Serbian experience. *Psychiatria Danubina.* (2018) 30(Suppl. 6):354–64.

31. Frenette P, Dodds L, Macpherson K. Factors affecting the age at diagnosis of autism spectrum disorders in Nova Scotia, Canada. *Autism.* (2013) 17:184–95. doi: 10.1177/1362361311413399

32. Valicenti-McDermott M, Hottinger K, Seijo R. Age at diagnosis of autism spectrum disorders. *J Pediatr.* (2012) 161:554–6. doi: 10.1016/j.jpeds.2012.05.012

33. Shattuck P, Durkin M, Maenner M. Timing of identification among children with an autism spectrum disorder: findings from a population-based surveillance study. *J Am Acad Child Adolesc Psychiatry.* (2009) 48:474–83. doi: 10.1097/CHI.0b013e31819b3848

34. Daniels AM, Mandell DS. Explaining differences in age at autism spectrum disorder diagnosis: a critical review. *Autism.* (2014) 18:583–97. doi: 10.1177/1362361313480277

35. Ingersoll B, Dvortcsak A. *Teaching social Communication to Children With Autism and Other Developmental Delays: The Project ImPACT Guide to Coaching Parents and the Project ImPACT Manual for Parents*. 2nd ed. New York, NY: Guilford Press (2019).

36. STAY-IN Project. *Early Start Intervention for Young Children With Autism Attending Educational Services in Low-Income Areas. STAY-IN Project Report*. (2019). Available online at: https://stay-in.adaptalab.org (accessed July 5, 2022).

37. Paula CS, Cukier S, Cunha GR, Irarrázaval M, Montiel-Nava C, Garcia R, et al. Challenges, priorities, barriers to care, and stigma in families of people with autism: similarities and differences among six Latin American countries. *Autism.* (2020) 24:2228–42. doi: 10.1177/1362361320940073

38. Bitterman A, Daley TC, Misra S, Carlson E, Markowitz J. A national sample of preschoolers with autism spectrum disorders: special education services and parent satisfaction. *J Autism Dev Disord.* (2008) 38:1509–17. doi: 10.1007/s10803-007-0531-9

39. McConachie H, Livingstone N, Morris C, Beresford B, Le Couteur A, Gringras P, et al. Parents suggest which indicators of progress and outcomes should be measured in young children with autism spectrum disorder. *J Autism Dev Disord.* (2018) 48:1041–51. doi: 10.1007/s10803-017-3282-2

40. Hoffman CD, Sweeney DP, Hodge D, Lopez-Wagner MC, Looney L. Parenting stress and closeness. *Focus Autism Dev Disabil.* (2009) 24:178–87. doi: 10.1177/1088357609338715

41. Farrugia D. Exploring stigma: medical knowledge and the stigmatisation of parents of children diagnosed with autism spectrum disorder. *Sociol Health Illness.* (2009) 31:1011–27. doi: 10.1111/j.1467-9566.2009.01174.x

42. Kinnear S, Link B, Ballan M, Fischbach R. Understanding the experience of stigma for parents of children with autism spectrum disorder and the role stigma plays in families' lives. *J Autism Dev Disord.* (2016) 46:942–53. doi: 10.1007/s10803-015-2637-9

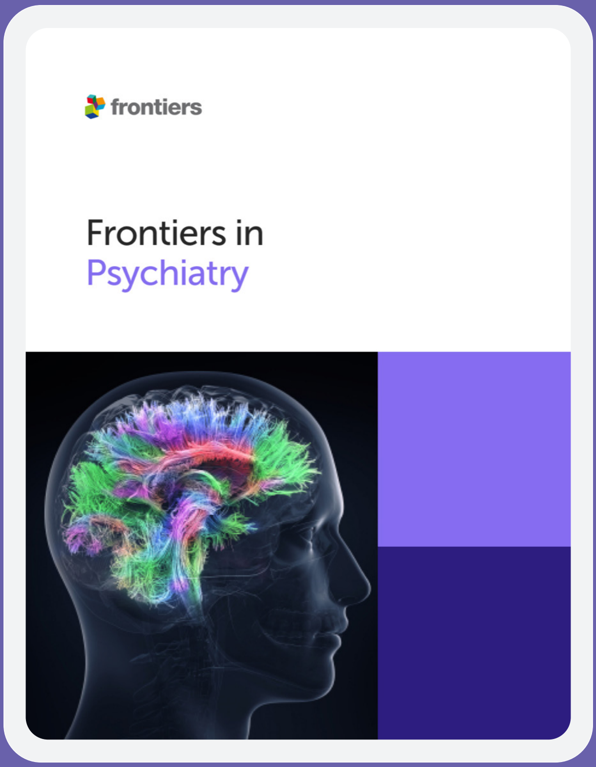
Frontiers in
Psychiatry

Explores and communicates innovation in the field of psychiatry to improve patient outcomes

The third most-cited journal in its field, using translational approaches to improve therapeutic options for mental illness, communicate progress to clinicians and researchers, and consequently to improve patient treatment outcomes.

Discover the latest Research Topics

See more →


frontiers

Frontiers in
Psychiatry

Frontiers

Avenue du Tribunal-Fédéral 34
1005 Lausanne, Switzerland
frontiersin.org

Contact us

+41 (0)21 510 17 00
frontiersin.org/about/contact