frontiers | Research Topics

Health (in)equity - examinations of the role of culture and trust

Edited by
Miodraga Stefanovska-Petkovska and Violeta Alarcão

Published in
Frontiers in Public Health

FRONTIERS EBOOK COPYRIGHT STATEMENT

The copyright in the text of individual articles in this ebook is the property of their respective authors or their respective institutions or funders. The copyright in graphics and images within each article may be subject to copyright of other parties. In both cases this is subject to a license granted to Frontiers.

The compilation of articles constituting this ebook is the property of Frontiers.

Each article within this ebook, and the ebook itself, are published under the most recent version of the Creative Commons CC-BY licence. The version current at the date of publication of this ebook is CC-BY 4.0. If the CC-BY licence is updated, the licence granted by Frontiers is automatically updated to the new version.

When exercising any right under the CC-BY licence, Frontiers must be attributed as the original publisher of the article or ebook, as applicable.

Authors have the responsibility of ensuring that any graphics or other materials which are the property of others may be included in the CC-BY licence, but this should be checked before relying on the CC-BY licence to reproduce those materials. Any copyright notices relating to those materials must be complied with.

Copyright and source acknowledgement notices may not be removed and must be displayed in any copy, derivative work or partial copy which includes the elements in question.

All copyright, and all rights therein, are protected by national and international copyright laws. The above represents a summary only. For further information please read Frontiers' Conditions for Website Use and Copyright Statement, and the applicable CC-BY licence.

ISSN 1664-8714
ISBN 978-2-83251-899-1
DOI 10.3389/978-2-83251-899-1

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

Health (in)equity - examinations of the role of culture and trust

Topic editors
Miodraga Stefanovska-Petkovska — Universidade de Lisboa, Portugal
Violeta Alarcão — University Institute of Lisbon, Portugal

Citation
Stefanovska-Petkovska, M., Alarcão, V., eds. (2023). *Health (in)equity - examinations of the role of culture and trust*. Lausanne: Frontiers Media SA.
doi: 10.3389/978-2-83251-899-1

Table of contents

05 Editorial: Health (in)equity - examinations of the role of culture and trust
Miodraga Stefanovska-Petkovska and Violeta Alarcão

10 Reducing Sexual and Reproductive Health Inequities Between Natives and Migrants: A Delphi Consensus for Sustainable Cross-Cultural Healthcare Pathways
Pedro Candeias, Violeta Alarcão, Miodraga Stefanovska-Petkovska, Osvaldo Santos, Ana Virgolino, Sónia Pintassilgo, Patrícia M. Pascoal, Andreia Silva Costa and Fernando Luís Machado

31 The Barriers and Enablers of Primary Healthcare Service Transition From Government to Community Control in Yarrabah: A Grounded Theory Study
Janya McCalman, Crystal Sky Jongen, Sandy Campbell, Ruth Fagan, Kingsley Pearson and Suzanne Andrews

46 Ingroup Bias in Healthcare Contexts: Israeli-Jewish Perceptions of Arab and Jewish Doctors
Elliot Graham, Samer Halabi and Arie Nadler

56 Health Literacy and Migrant Communities in Primary Health Care
Patrícia Medina, Ana Catarina Maia and Andreia Costa

64 Connections and Biases in Health Equity and Culture Research: A Semantic Network Analysis
Mireya Martínez-García, José Manuel Villegas Camacho and Enrique Hernández-Lemus

85 Complicated Appendicitis Among Adults With and Without Disabilities: A Cross-Sectional Nationwide Study in South Korea
Kyoung Eun Yeob, So Young Kim, Jong Eun Park and Jong Hyock Park

95 Beliefs, Perceptions, and Behaviors Regarding Chronic Respiratory Diseases of Roma in Crete, Greece: A Qualitative FRESH AIR Study
Marilena Anastasaki, Egid M. van Bree, Evelyn A. Brakema, Ioanna Tsiligianni, Dimitra Sifaki-Pistolla, Vasiliki E. Chatzea, Matty C. Crone, Andreas Karelis, Rianne M. J. J. van der Kleij, Charlotte C. Poot, Ria Reis, Niels H. Chavannes and Christos Lionis on behalf of the FRESH AIR collaborators

107 Empirical Analysis of Health Assessment Objective and Subjective Methods on the Determinants of Health
Abdou Khadre Dit Jadir Fall, Florence Migot-Nabias and Najoua Zidi

116 Public Views About Involvement in Decision-Making on Health Data Sharing, Access, Use and Reuse: The Importance of Trust in Science and Other Institutions
Ngozi Nwebonyi, Susana Silva and Cláudia de Freitas

127 **Association Between Residence Status Regularization and Access to Healthcare for Undocumented Migrants in Switzerland: A Panel Study**
Julien Fakhoury, Claudine Burton-Jeangros, Liala Consoli, Aline Duvoisin and Yves Jackson

140 **A latent profile analysis of residents' knowledge, attitude, and practice toward common chronic diseases among ethnic minority area in China**
Huaqin Hu, Yihua Xu, Yingshan Shao, Yaxin Liang, Qionghua Wang, Shunmei Luo, Heyun Lu, Heng Meng and Chenxi Liu

150 **Development and validation of cultural competence assessment tool for healthcare professionals, India**
Parvathy Balachandran, Vineetha Karuveettil and Chandrashekar Janakiram

161 **The care types choice in filial culture: A cross-sectional study of disabled elderly in China**
Zheng Zang

frontiers | Frontiers in Public Health

TYPE Editorial
PUBLISHED 23 November 2022
DOI 10.3389/fpubh.2022.1078967

Editorial: Health (in)equity - examinations of the role of culture and trust

Miodraga Stefanovska-Petkovska[1]* and Violeta Alarcão[1,2]

[1]Instituto de Saúde Ambiental, Faculdade de Medicina, Universidade de Lisboa, Lisbon, Portugal, [2]Centro de Investigação e Estudos de Sociologia, Instituto Universitário de Lisboa (Iscte), Lisbon, Portugal

KEYWORDS

inequity, public health, culture, trust, health, health disparities

Editorial on the Research Topic
Health (in)equity - examinations of the role of culture and trust

Our health throughout the life course is a peculiar, individualized interaction of nature and nurture. Achievements in science have provided an improved understanding of the role of genetics and environment contribute to disease and disability, and biomedical interventions have often been able to provide the prospect of bringing a person back to full health or living with the disease with reduced discomfort. Nevertheless, how sociocultural behaviors and environmental factors (nurture) can trigger biological and genetic processes (nature), not just the other way around, has been largely neglected (1–4). New approaches to boundaries between internal and external environments, health and disease, and social and biological are needed to merge the gaps while contributing to the understanding of the influence of socioeconomic factors on health (1). Global migrations, changes in the demographic and cultural profile of countries, emerging disease vectors, and communicable and non-communicable illnesses are just some of the issues that in the last several decades have spurred the growth of multi-disciplinary attention on the importance of culture to health. Cultural and linguistic diversity, socioeconomic differences in healthcare utilization, the technologization of health, and the degree of empowerment of patients to make their own decision, all these issues raised awareness of how inseparable health is from culturally affected perceptions of wellbeing and integration, and how understanding culture is imperative to the advancement of health worldwide. For example, ethnic/racial minority communities experience worse health outcomes due to underutilization of healthcare services as a result of language barriers, differences in the cultural understanding of health, healthcare and health-seeking behavior, the inability of the healthcare system and workforce to identify and understand the specific needs and circumstances of the patient, among other factors (5, 6). According to MBRRACE-UK - Mothers and Babies: Reducing Risk through Audits and Confidential Enquiries across the UK - last report, Black and Asian Ethnic Women in the U.K. are 5 and 2 times more likely to die during pregnancy and after childbirth compared to White Women (7). However, behind

Check for updates

OPEN ACCESS

EDITED AND REVIEWED BY
Cyrille Delpierre,
INSERM U1027 Epidémiologie et analyses en santé publique: Risques, Maladies Chroniques et Handicap, France

*CORRESPONDENCE
Miodraga Stefanovska-Petkovska
mpetkovska@medicina.ulisboa.pt

SPECIALTY SECTION
This article was submitted to Life-Course Epidemiology and Social Inequalities in Health, a section of the journal Frontiers in Public Health

RECEIVED 24 October 2022
ACCEPTED 10 November 2022
PUBLISHED 23 November 2022

CITATION
Stefanovska-Petkovska M and Alarcão V (2022) Editorial: Health (in)equity - examinations of the role of culture and trust.
Front. Public Health 10:1078967.
doi: 10.3389/fpubh.2022.1078967

COPYRIGHT
© 2022 Stefanovska-Petkovska and Alarcão. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

every maternal death, there are whole groups of women suffering negative health outcomes. These unacceptable racial maternal health disparities are not limited to the UK and not limited solely to race, but other countries/regions/continents and cultural groups as well. Despite that, the word "culture" with its modern technical or anthropological meaning was established by Tylor as far back as 1871, and it took fifty years to penetrate British and American dictionaries (8). Likewise, although the awareness of the importance of the interaction culture-health is present for a long time, the awareness of the importance of a closer definition of culture as relating to health emerged within the last decade. A comprehensive definition was only recently provided by the Lancet Commission defining culture *"as the shared, overt and covert understandings that constitute conventions and practices, and the ideas, symbols, and concrete artifacts that sustain conventions and practices, and make them meaningful"* (9).

The COVID-19 experiences have provided another reason to raise attention to the relationship between cultural origins and health. The events in 2020 and the beginning of 2021, taught us more about the urgency of addressing the cultural origins of health inequities, compared to the previous decades altogether. They demonstrated how interconnected the world and society are; more important than the path toward herd immunity, primarily starting with the awareness that the herd is not a homogenous group. A recently published article highlighted the effects of the COVID-19 pandemic on the existing deep-rooted and enduring health and wider inequalities (10). For example, the disproportionally higher mortality rate in the poorest communities in the European Union, such as Seine-St-Denis, one of the poorest areas in France, attracted the attention of the world. Poorer communities were particularly affected during the lockdown due to their social conditions (no separate room to isolate ill persons) combined with the inability to understand medical prescriptions and instructions (11). Interestingly, the same pandemic pronounced the issues of trust and its relations with cultural systems of value relating to health and illness (12–14). In fact, it is thought that the "epidemic of mistrust" had indeed become a global crisis threatening to characterize public perceptions of healthcare (15, 16) along with the recontextualization of medical knowledge by competing agencies agents and human and non-human actors in the era of "post-truth" (16). The issue of trust and health was especially highlighted amongst those living in the most deprived regions, communities, or neighborhoods, as well as people from minority racial and ethnic communities (17, 18).

Unfortunately, despite the rising awareness of the public health significance of health equity, there is a conspicuous lack of focus on the intersection of cultural diversity, trust, and health. Hence, the existing knowledge gap may make building a trusting and positive relationship with ethnic and racial minority patients particularly challenging. Under this background, the development of an increased understanding of the role of culture and trust in achieving health equity is a top priority to ensure the success of public health interventions. In this Research Topic, a total of 13 excellent articles presenting five different perspectives on health equity are included that contribute to the field by including texts that analyze how "*health inequity*" may become "*health in equity*".

Diversity and health equity

In today's globalized world we are continuously exposed to the richness of diverse social groups - ethnicities, religions, cultures, etc. Although intergroup communication between people belonging to diverse groups has been extensively researched and discussed, it has unfortunately remained understudied in the specific context of healthcare. Yet, it remains crucial for the existence of health equity. According to the WHO, one of the major burdens to global health are Chronic respiratory diseases (CRDs) (19), and a particularly vulnerable group to CRDs are the Roma (20), Europe's largest ethnic minority. Despite national and European efforts to improve access to care for Roma, health improvements remain limited. The study done by Anastasaki et al. studied CRD-related beliefs, perceptions, and behaviors among a Greek Roma population, focusing on asthma and COPD. They concluded that to tackle CRD within the Roma community, a multilevel approach should be adopted: bridging awareness gaps at the population level, providing resources to enhance the adoption of healthy behaviors, and fighting discrimination at the societal level, whilst establishing trusted relationships at the local level. The authors recommend that similar locally-tailored methodologies may strengthen the implementation of effective interventions for similarly vulnerable and/or low-resource populations. Another perspective relevant to equal public health access is given by McCalman et al. in which they sought to identify the barriers and enablers to transitioning the delivery of primary healthcare services from Queensland Health to Gurriny Yealamucka community-controlled health service in Yarrabah. Their evaluation of Yarrabah's transition process suggests that future such transitions will require planning and commitment to a long-term, multi-faceted and complex process, encompassing the required level of authorization and resourcing. Furthermore, it is well established that timely and appropriate healthcare plays a key role in wellness, illness prevention, and optimal recovery when illness occurs. However, healthcare disparities exist between people with and without disabilities, with the former group being more likely to experience a delay in healthcare that could contribute to differences in outcomes, such as mortality. The study done by Yeob et al. sought to compare 10-year trends of complicated appendicitis between South Koreans with and without a disability. They found that the incidence of complicated appendicitis was higher in people with disabilities, especially those with severe disabilities. Therefore, based on the findings it is recommended that public health policies should focus on people with disabilities to reduce disparities in health outcomes. Additionally, healthcare professionals should be educated toward improving equal access to diagnosis and treatment of people with disabilities. The role of healthcare professionals, and especially the level of their cultural competence, remains a cornerstone in health equity. India is one such example since existing assessment scales have limited application in the country due to the nation's rich cultural diversity and heterogeneous healthcare streams. Despite the tremendous improvement in the healthcare system owing to advancements in technology and research, the disease burden in the country remains unchanged, particularly among the underprivileged and underrepresented communities. The study by Balachandran et al. was undertaken to develop and validate a cultural competence assessment tool for healthcare professionals in India. The resulting tool can be used to assess the cultural competence level of healthcare professionals as the first step toward designing cultural competence training for healthcare manpower and the establishment of culturally sensitive healthcare organizations in India.

Further within this context, a group of special, but sometimes understudied, interest is older people with disabilities. The study by Zang examined the influence of the factors in the cultural context of filial piety on the choice of care types for older people with disability in China. According to the characteristics of filial culture, the factors influencing the choice of care type for older people in China are summarized as family endowment and support. The study concludes that gender, residence, living alone or not, family income, real estate, pension, and community service have momentous effects on the choice of care type of older people with disability; informal care has a substitutive effect on formal care. Hence, the government should consider informal care official support such as cash and services, to change the attribute of the private domain of filial culture and enhance the quality of long-term care.

International migrants and healthcare utilization

The increasing number of international migrants (ranging from 153 million in 1990 to ∼272 million in 2019) brought to attention the wide variation of national contexts concerning the policy measures to protect migrants' rights and ensure their equal access to basic and essential services, namely in health. In this context, one of the most frequently discussed issues in the area of health inequities concerns the health of migrants and its determinants. Even in a universal healthcare system, such as the one in Switzerland, undocumented migrants face barriers at different levels that hinder their access to healthcare services. Therefore, Fakhoury et al. aimed to assess whether undocumented migrants' healthcare utilization improves with residence status regularization. The study results confirmed that residence status regularization is associated with improved healthcare utilization among undocumented migrants. Therefore, future research is needed to understand the mechanisms through which regularization improves undocumented migrants' use of healthcare services. Another key component to the overall health and quality of life of migrants is sexual and reproductive health. The study done by Candeias et al. through the use of the Delphi panel technique, identified good practices in the SRH field, with a particular focus, whenever possible, on migrant populations, and to identify relevant and inclusive indicators to monitor SRH in Portugal. Their findings provide extended opportunities for the healthcare system to engage in better-informed decisions and more inclusive and integrative strategies regarding SRH, contributing to building political measures toward sexual and reproductive justice.

Health literacy

By acknowledging the importance of health literacy as a fundamental strategy for empowering migrants and promoting equity in their access to health care, Medina et al. investigated the level of health literacy of the migrant population attending a primary health care unit in the Lisbon region. They found that problematic and inadequate levels of health literacy were significantly frequent among the migrant population. Therefore, the authors suggested that the enhancement of health literacy among migrants is essential to the reduction of health inequalities, achieving better health outcomes, and contributing to the defense of the human rights of this vulnerable population. Furthermore, health literacy plays an important role in preventing and managing chronic diseases, while low levels of health literacy among ethnic minorities are a major manifestation of health inequities. The study by Hu et al. updated insights on health literacy among ethnic minorities by investigating the knowledge, attitude, and practice (KAP) profile of common chronic diseases in ethnic minority areas, and discussed the KAP profiles in detail to inspire future health education interventions. The authors noted that a more specific and nuanced understanding of ethnic minority health literacy could allow providers to conduct more effective health education with their recipients.

Improving health research methodology

A crucial step toward an integrated understanding of social determinants and cultural issues contributing to determining the health inequity status and related issues, consists, not only in enlisting them but also in sketching the interplay that these features may have among themselves to give rise to the observed impact of social constraints upon population-level health conditions (21). Nevertheless, the future of health equity assessment also depends on our continued innovation in developing methods to monitor them and intervene from an integral, inclusive perspective. The paper published by Martínez-García et al. presented the state of affairs regarding the scholarly discussion on these quite relevant subjects, to serve as a starting point for deeper analyses. Through the use of data analytics, the authors highlighted potential pathways for future research by identifying certain biases and under-representation of several relevant concepts, likely influenced by the fact that the academic literature is both relatively scarce and produced in a few countries, most of which are developed or emerging economies characterized by firmly established trends in their health systems. The final study in this group, by Fall et al., investigated the empirical differences between health assessment objective and subjective methods, to identify a possible long-term relationship between methods and health determinants and the influence of these methods on the perceived level of risk according to health determinants. Using data from 1970 to 2018 in the United States, they found that health assessment methods influence the determinants of health and the perceived risk of health determinants changes according to the method used. Therefore, the impact of health assessment methods must be considered to adequately prioritize the determinants of health.

Healthcare quality improvement

Although healthcare quality improvement can be bolstered by data-intensive and needs-driven research, mounting reports of data breaches and mismanagement have generated concern for privacy loss, undisclosed surveillance, and discrimination thus undermining public trust in data processing organizations. The final study, conducted by Nwebonyi et al., assessed the data sharing, access, use, and reuse views of rare disease patients and their informal carers, and found that most participants perceived involvement in decision-making about data sharing, access, use and reuse to be important or very important. This high value attributed by participants to involvement in individual-level data governance stresses the need to rethink opportunities for public participation in health data decision-making.

The variety of topics submitted to this Research Topic demonstrates that this field of knowledge is growing progressively, incorporating new areas into the concept of health (in) equity - linking disabilities, old age, family caregiving culture, health literacy among ethnic and migrant groups, chronic disease and access to healthcare, trust in individual-level data governance as well as the potential impact of health assessment methods on the prioritization of the determinants of health. Altogether, they acknowledge that failure to adequately and timely address health inequity may worsen the outcomes for vulnerable groups, even more, and take its toll during another future pandemic. If timely addressed, the development of health systems and approaches that are sensitive to cultural characteristics would result in building a feeling of trust and inclusion with multiple positive consequences for the health of the patients, their families, and the communities in which they live. We hope that this Research Topic can contribute to increasing our understanding to link culture, trust, and health, by identifying and promoting sustainable health-in-equity practices.

Author contributions

MS-P wrote the first draft of the editorial and VA provided comments and recommended amendments. Both authors contributed to the article and approved the submitted version.

Acknowledgments

We deeply thank all the authors and reviewers who have participated in this Research Topic.

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

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

References

1. Canali S, Leonelli S. Reframing the environment in data-intensive health sciences. *Stud Hist Philos Sci*. (2022) 93:203–14. doi: 10.1016/j.shpsa.2022.04.006

2. Landecker H, Panofsky A. From social structure to gene regulation, and back: a critical introduction to environmental epigenetics for sociology. *Annu Rev Sociol.* (2013) 39:333–57. doi: 10.1146/annurev-soc-071312-145707

3. Lock M. The epigenome and nature/nurture reunification: A challenge for anthropology. *Med Anthropol.* (2013) 32:291–308. doi: 10.1080/01459740.2012.746973

4. Singh I. Thinking on the Edge Human development, nature and nurture: Working beyond the divide. *BioSocieties.* (2012) 7:308–21. doi: 10.1057/biosoc.2012.20

5. Dune T, Perz J, Mengesha Z, Ayika D. Culture Clash? Investigating constructions of sexual and reproductive health from the perspective of 1.5 generation migrants in Australia using Q methodology. *Reprod Health*. (2017) 14:50. doi: 10.1186/s12978-017-0310-9

6. Sheikh-Mohammed M, MacIntyre CR, Wood NJ, Leask J, Isaacs D. Barriers to access to health care for newly resettled sub-Saharan refugees in Australia. *Med J Aust*. (2006) 185:594–597. doi: 10.5694/j.1326-5377.2006.tb00721.x

7. Draper E, Gallimore I, Smith L, Fenton A, Kurinczuk J, Smith P, et al. *MBRRACE-UK perinatal mortality surveillance report, UK perinatal deaths for births from January to December 2019.* The Infant Mortality and Morbidity Studies, Department of Health Sciences, University of Leicester. (2021).

8. Stocking GW. Matthew Arnold, EB Tylor, and the Uses of Invention. *Am Anthropol.* (1963) 65:783–99. doi: 10.1525/aa.1963.65.4.02a00010

9. Napier AD, Ancarno C, Butler B, Calabrese J, Chater A, Chatterjee H, et al. Culture and health. *Lancet*. (2014) 384:1607–39. doi: 10.1016/S0140-6736(14)61603-2

10. Lal A, Erondu NA, Heymann DL, Gitahi G, Yates R. Fragmented health systems in COVID-19: rectifying the misalignment between global health security and universal health coverage. *Lancet*. (2021) 397:61–7. doi: 10.1016/S0140-6736(20)32228-5

11. Marine M, Joannes C, Neufcourt L, Delpierre C, Kelly-Irving M. *Living Conditions and Understanding Medical Guidelines During Covid-19 | Covid-19 et conséquences du confinement de la population*. EPIDEMIC Project First Results (2020). Available online at: https://epidemic.hypotheses.org/153#more-153

12. Marmot M, Allen J, Goldblatt P, Herd E, Morrison J. Build Back Fairer: The COVID-19 Marmot Review. The Pandemic, Socioeconomic and Health Inequalities in England. Institute of Health Equity. (2020). Available online at: http://www.instituteofhealthequity.org/resources-reports/build-back-fairer-the-covid-19-marmot-review/build-back-fairer-the-covid-19-marmot-review-executive-summary.pdf (accessed December 28, 2020).

13. Montel L, Kapilashrami A, Coleman MP, Allemani C. The right to health in times of pandemic: what can we learn from the UK's response to the COVID-19 outbreak? *Health Hum Rights.* (2020) 22: 227–41.

14. OECD. COVID-19 crisis puts migration and progress on integration at risk - OECD. (2020). Available online at: https://www.oecd.org/migration/covid-19-crisis-puts-migration-and-progress-on-integration-at-risk.htm (accessed December 13, 2020).

15. Cheung ATM, Parent B. Mistrust and inconsistency during COVID-19: Considerations for resource allocation guidelines that prioritise healthcare workers. *J Med Ethics.* (2020) 47:73–7. doi: 10.1136/medethics-2020-106801

16. Kwok H, Singh P, Heimans S. The regime of 'post-truth': COVID-19 and the politics of knowledge. In: *Discourse: Studies in the Cultural Politics of Education.* (2021) p. 1–15. doi: 10.1080/01596306.2021.1965544

17. Armstrong K, Ravenell KL, McMurphy S, Putt M. Racial/ethnic differences in physician distrust in the United States. *Am J Public Health.* (2007) 97:1283–9. doi: 10.2105/AJPH.2005.080762

18. Paul E, Fancourt D, Razai M. Racial discrimination, low trust in the health system and COVID-19 vaccine uptake: a longitudinal observational study of 633 UK adults from ethnic minority groups. *J R Soc Med.* (2022). doi: 10.1177/01410768221095241. [Epub ahead of print].

19. WHO. Chronic obstructive pulmonary disease (COPD). (2017). Available online at: https://www.who.int/news-room/fact-sheets/detail/chronic-obstructive-pulmonary-disease-(copd) (accessed May 15, 2022).

20. Marmot M, Allen J, Bell R, Bloomer E, Goldblatt P. WHO European review of social determinants of health and the health divide. *Lancet.* (2012) 380:1011–29. doi: 10.1016/S0140-6736(12)61228-8

21. Lathrop B. Moving toward health equity by addressing social determinants of health. *Nurs Women's Health.* (2020) 24:36–44. doi: 10.1016/j.nwh.2019.11.003

frontiers in Public Health

ORIGINAL RESEARCH
published: 13 May 2021
doi: 10.3389/fpubh.2021.656454

Reducing Sexual and Reproductive Health Inequities Between Natives and Migrants: A Delphi Consensus for Sustainable Cross-Cultural Healthcare Pathways

Pedro Candeias[1†], *Violeta Alarcão*[1,2*†], *Miodraga Stefanovska-Petkovska*[1], *Osvaldo Santos*[1,3], *Ana Virgolino*[1], *Sónia Pintassilgo*[2], *Patrícia M. Pascoal*[4,5], *Andreia Silva Costa*[1,6] and *Fernando Luís Machado*[2]

[1] Instituto de Saúde Ambiental, Faculdade de Medicina, Universidade de Lisboa, Lisboa, Portugal, [2] Centro de Investigação e Estudos de Sociologia, ISCTE - Instituto Universitário de Lisboa (ISCTE-IUL), Lisboa, Portugal, [3] Unbreakable Idea Research, Painho, Portugal, [4] Centro de Investigação em Ciência Psicológica, Faculdade de Psicologia, Universidade de Lisboa, Lisboa, Portugal, [5] Digital Human-Environment Interaction Lab, Universidade Lusófona, Lisboa, Portugal, [6] Centro de Investigação, Inovação e Desenvolvimento em Enfermagem de Lisboa, Escola Superior de Enfermagem de Lisboa, Lisboa, Portugal

OPEN ACCESS

Edited by:
Aldo Rosano,
National Institute for the Analysis of Public Policy, Italy

Reviewed by:
Angela Freitas,
University of Coimbra, Portugal
Elena Riza,
National and Kapodistrian University of Athens, Greece

***Correspondence:**
Violeta Alarcão
violeta_sabina_alarcao@iscte-iul.pt

† *These authors share first authorship*

Specialty section:
This article was submitted to Life-Course Epidemiology and Social Inequalities in Health, a section of the journal Frontiers in Public Health

Received: *20 January 2021*
Accepted: *14 April 2021*
Published: *13 May 2021*

Citation:
Candeias P, Alarcão V, Stefanovska-Petkovska M, Santos O, Virgolino A, Pintassilgo S, Pascoal PM, Costa AS and Machado FL (2021) Reducing Sexual and Reproductive Health Inequities Between Natives and Migrants: A Delphi Consensus for Sustainable Cross-Cultural Healthcare Pathways. Front. Public Health 9:656454. doi: 10.3389/fpubh.2021.656454

The increasing number of international migrants (ranging from 153 million in 1990 to ∼272 million in 2019) brought to attention the wide variation of national contexts concerning the policy measures to protect migrants' rights and ensuring their equal access to basic and essential services, namely in health. Sexual and Reproductive Health (SRH) is a key component to the overall health and quality of life and is impacted by power inequities inherent to society's institutions, environment, economics, and culture. In Portugal, guidelines for intervention in SRH are insufficient, a gap that is more pronounced with migrant populations due to the absence of culturally sensitive indicators to assess and monitor SRH. The aim of this work was 2-fold: to identify good practices in the SRH field, with a particular focus, whenever possible, on migrant populations, and to identify relevant and inclusive indicators to monitor SRH in Portugal. A Delphi panel (via online survey) with 66 experts (researchers, teachers, and health professionals) and 16 stakeholders (non-governmental organizations, civil society, and governmental organizations) was implemented in two rounds. Panelists were asked to state their level of agreement (5-point Likert-type scale) regarding four different SRH areas: Sexual Health, Reproductive Health, Social-Structural Factors, and Good Practices. Items were based on literature review and a World Café with 15 experts and stakeholders. Participation rate was 68% and response rate was 97% on the first round. From the initial list of 142 items, a total of 118 (83%) were approved by consensus. Findings may provide extended opportunities for the healthcare system to engage in better informed decisions and more inclusive and integrative strategies regarding SRH, contributing to build political measures toward sexual and reproductive justice.

Keywords: sexual and reproductive health, health equity, migrants, Delphi panels, inequities and inequalities in health

INTRODUCTION

According to the World Migration Report 2020, the total number of international migrants is estimated to be almost 272 million, with nearly two-thirds being labor migrants and nearly half being female (1). In 2020, female migrants accounted for 47.6% of all migrants in high-income countries, 48.2% in middle-income countries, and 50.9% in low-income countries. The share of female migrants was highest in North America (51.8%) and Europe (51.4%). In addition, the current estimated number and proportion of international migrants already surpasses the projections made for the year 2050 (1).

The relationship between migration and health is well-established in the literature. In general, the existing research studies investigate and suggest interventions in four key aspects of the relationship between migration and health: (1) health of migrants; (2) the impact of migration on public health; (3) the response of the healthcare system; and (4) the global governance of migration and health (1, 2). Each of these aspects is discussed in detail bellow and illustrated in **Figure 1**.

1) Health of migrants—The area that concerns the health of migrants focuses on the differences in the health status between the migrants and their counterparts in the origin and destination country. The determinants of changes in the health status of the migrants are dependent on the exposure to risk factors at departure, during transit and at arrival (3–5). In example, some migrants are faced with increased risk for sexual violence and exploitation during the migration journey (5).
2) Impact of migration on public health—The second key element is public health and the global target of universal health coverage (UHC) (6). Access to affordable quality and culturally competent healthcare is an important concern for all vulnerable groups, especially migrant workers, and poses a neglected challenge to progress toward universal health coverage. Therefore, national systems should identify migrant population in order to understand the scale of migration, develop evidence-based policies, and know the extent to which refugees and labor migrants are able to access health and other social services. As 64% of all migration is related to work, it would therefore benefits the host country to invest in their health (7).
3) The healthcare systems response is one of the essential elements of the intersection between migrants and health. Developing systems that are sensitive to migrants cultural and health characteristics would result with multiple positive consequences for the health of the migrants, their families and the communities in which they live. In example, services for sexual and reproductive health are typically under-utilized by migrant and refugee communities and certain studies indicate a lower utilization rate of health services of migrant, compared to native women (8) due to lack of knowledge about available services and how to access them, language barriers, differences in the cultural understanding of health, healthcare and health-seeking behavior, inability of the healthcare system and workforce to identify and understand the specific needs and circumstances of the migrant population, as well as unresolved administrative status of the person (9).
4) Finally, the global governance of migration and health encompasses the integration of equity, accountability, impartiality, fairness, justice and probity into the global governance processes (10).

During 2019, the stock of foreigners in Portugal accounted for 590,348 people (5.7% of the total population) (11). In terms of age, 57.5% of foreigners were aged between 15 and 44 years with a 50/50 ratio between men and women (12). The Portuguese Observatory for Migration publishes an annual statistical report of the migrant integration indicators that allows access to organized indicators on social, economic, educational and civic indicators, based on nationality. It also provides an understanding of the challenges that persist in monitoring the integration of migrants in Portugal, namely in the health dimension, with indicators related to access to and use of health services, and the needs of resident populations and health systems (13). As in other countries, in Portugal there are differences in health indicator outcomes between migrants and the autochthonous population placing migrants in an unfavorable position in terms of their access and utilization of healthcare services, specifically concerning certain health risk factors such as inadequate diet, tobacco, and alcohol consumption (5). The systematic health status differences between natives and migrants may reflect inequities in the accessibility of health services, as well as diverse health inequalities and health protection needs due to the socio-economic characteristics of the population (13–16). By definition inequity refers to unfair, avoidable differences arising from poor governance, corruption or cultural exclusion while inequality simply refers to the uneven distribution of health or health resources as a result of genetic or other factors or the lack of resources (17). Inequality was listed as a global risks in 2012, while in 2017 it was considered that in the following decade the rising income and wealth disparity will be one of most powerful determinants of global development (18). An example to the significance of addressing this issue can be found within the issue of maternal deaths in low- and middle- income countries. Although the majority of maternal deaths are avoidable through quality obstetric care, such as cesarean section, evidence suggests inequality and inequities among women in low-and middle-income countries concerning obstetric services. Findings from a 19 year study in Tanzania indicated that women who were uneducated, poorest/poor, living in rural settings and from certain regions demonstrated lower utilization of obstetric services (19). In regards to Portugal, the country has already implemented the concrete measures in the past 5 years to increase women's access to comprehensive sexual and reproductive health services, regardless of marital status and age as well as support for family planning and specific programs to ensure the access of adolescents and youth to sexual and reproductive health information (20). However,

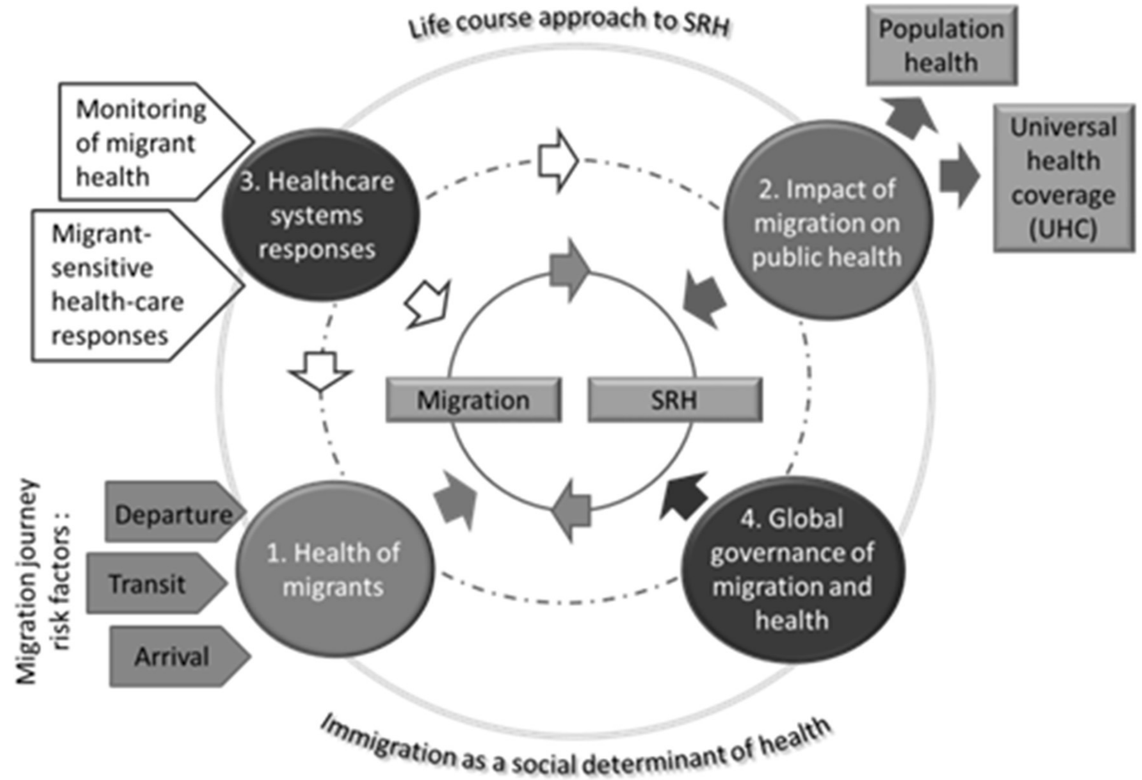


FIGURE 1 | Key aspects and determinants of migration and health. Source: Authors own compilation based on (1, 2).

intergroup differences are observed between migrant and domestic population. A study found that the families of newborn children in Amadora and Sintra Council districts (districts with the highest proportion of migrants) face increased socio-material deprivation compared to the general population of the Greater Metropolitan Area of Lisbon. Their health vulnerability is reflected in the greater fetal and post-natal mortalities and more deaths during pregnancy, mainly due to infectious diseases (21). Another study that used data on births registered between 1995 and 2002 and classified by reported nationality of mothers, found that among African births there was an increase in births to teenaged mothers and a decline to mothers from advantaged socioeconomic backgrounds. Additionally, in the investigated period there was a decline on mean birth weight among African babies that was found to be associated with socioeconomic advantage (22). The impact of structural inequities and socioeconomic health determinants in ethnic and migrant health inequities has increased during the COVID-19 pandemic (23). In this context, it is essential to address the wide variation of national experiences in what concerns policy measures to protect migrants' rights and well-being and ensuring equal access to essential services, with special emphasis on healthcare.

Addressing Sexual and Reproductive Health Related Inequities

Sexual and reproductive health (SRH) is shortly defined by the World Health Organization (WHO) (24) as a state of physical, emotional, mental and social well-being related to sexuality. The health issues covered by SRH include, but are not limited to improving maternal and newborn care, providing high quality services for family planning, eliminating unsafe abortion, combatting sexually transmitted infections, and promoting sexual health, which includes protecting sexual rights, improve sexual function and promote sexual pleasure free of coercion (25). Although the foundation of SRH health outcomes lies in individual behavior, there is an array of forces and systems shaping the conditions of migrants' daily lives that cannot be ignored (26). Therefore, effective SRH can only be achieved when considering the full range of factors that make a critical difference to health outcomes. This is especially important since services for SRH are typically under-utilized by migrant and refugee communities, when compared to the native population (8). Reasons include lack of knowledge about available services and how to access them, language barriers, differences in the cultural understanding of health, healthcare and health-seeking behavior, inability of the healthcare system and workforce to identify and understand the specific needs and circumstances of the migrant population, as well as unresolved administrative status of the person (9).

One of the novel approaches in sexual health monitoring and evaluation with specific focus on migrant population highlights the importance of envisaging the diversity of individual needs at various points across life course and in various settings or circumstances (27).

The importance of this interaction is highlighted in the fifth key principle from the WHO operation framework—*Diversity of needs across life course and populations* (28, 29). This principle highlights three forces that shape SRH—individual, environment and time. More specifically, it views sexual health as a complex interaction between individual characteristics, the role of the cultural, socioeconomic, geopolitical and legal environment in SRH outcomes, but also the changes incurred over time and across the lifespan. In addition this goal is complementary with the UN's Sustainable Development Goal 3 (SDG 3) for 2030 which aims to "ensure healthy lives and promote well-being for all at all ages." This goal crosscuts with the other SDGs (30, 31). Hence, in regards to SRH the intersectional approach envisages the importance of 'the different stages in one's life cycle' and of being aware of where people are in the life cycle as their capacities and needs change over time. It has also been incorporated into the United Nations Refugee Agency's (UNHCR) Age, Gender, and Diversity (AGD) framework (32) (**Figure 2**). This framework sets out a definition of diversity for 'one community, many people', and draws attention to the roles and needs of women and girls, men and boys, children (including adolescents), people who are lesbian, gay, bisexual, transsexual, or intersex (LGBTI), older men and women, disabled people, and those belonging to national or ethnic, religious and linguistic minorities or indigenous groups (33). In accordance with the AGD framework, the approach should be used to plan, program, implement, monitor, and evaluate the relevant indicators. The adoption of the life-course approach promotes functional ability of the individual, as the sum of the individual and environmental attributes that enable a person to be or do what they have reason to value, that in turn enables well-being and is interdependent with the realization of rights (31, 34). Estimates suggest that long-term investment in the life-course approach can results in with benefits that are not limited to health, but extend to social and economic development as well (31). In example, the reduction of preventable diseases in low- and middle-income countries has resulted in their increased economic growth (35). An example of a more locally oriented action is the Madsen's Institute for Tribal and Rural Advancement program that utilized the life-course approach in their cross-sectorial programs to transform the health of people in 48 villages in Orissa in India by targeting primarily malaria control and afterwards including other interdependent health, educational, environmental and poverty-reduction goals. The result was a halved infant mortality rate over a 15 year period, and a range of advances in the areas of health, social and developmental areas, that in contrast remained very low in villages not covered by the program (36).

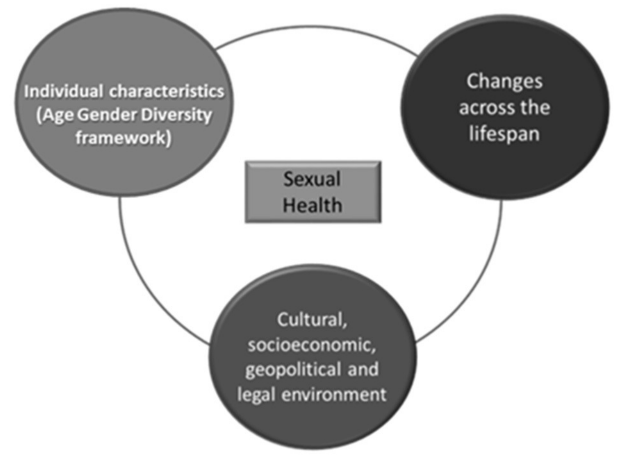


FIGURE 2 | Determinants of sexual health. Source: Authors own compilation based on WHO operation framework of SRH interventions (10) and the United Nations Refugee Agency's (UNHCR) Age, Gender, and Diversity (AGD) framework (11).

This fifth key principle is complemented by two main approaches found in the literature: *the life course approach to SRH* (37) and *the migration as a social determinant of health* (38). The first approach argues that events at different stages of life must be understood as fundamentally connected (39). According to this approach individual life courses are composed of multiple, simultaneously occurring trajectories through various dimensions of life (e.g., family, work, sexuality). Each trajectory extends from birth until death and can be divided into a sequence of transitions (i.e., retirement or virginity loss). This framework posits that sexual beliefs and behaviors result from individuals' lifelong accumulations of advantageous and disadvantageous experiences—social, psychological, and physiological—and their adoption or rejection of sexual scripts within specific socio-historical contexts.

In regards to migration, in their lives migrants undergo experiences that ultimately affect their health in a setting characterized with legal, cultural, social, economic, and behavioral barriers. Migration itself can be a strong determinant of physical and mental health. Therefore, it should be viewed as a social determinants of health which emphasizes the *racialized-gendered social determinant of health*—the dominance of race and gender identities, along with other identities such as social class, sexual orientation, age, ethnicity and nativity, and legal status, that form the basis for education and health frameworks (40). It is considered that the ability to treat migration as a social determinants of health has the potential to result in a comprehensive and targeted response to the health of the populations affected by the global phenomenon of migration (38).

Taking into consideration all the relevant aspects, approaches and arguments that surround SRH and migration, the Delphi method was chosen to identify guidelines for intervention with migrant populations in Portugal that are currently insufficient due to the absence of culturally sensitive indicators to assess and monitor SRH. The Delphi method has been commonly applied in the selection processes of health indicators where group opinion is needed from an audience with varied views, such as in the health field (41). This method has been used in studies to select indicators on healthcare services (42, 43), perinatal health in Europe (44, 45), health inequalities and inequities (46, 47) and population health (41). Therefore, this Delphi study was implemented to generate consensus on:

a) what constitutes good practices in the SRH field, with emphasis on SRH equity across migrant populations;
b) relevant and inclusive indicators to monitor SRH, namely among migrants, in Portugal.

MATERIALS AND METHODS

The study was approved by the Ethics Committee of the Centro Académico de Medicina de Lisboa (CAML). A Delphi panel approach was used to achieve agreement on the best indicators to monitor SRH in Portugal, establishing good practices in the SRH field to both the host and the migrant populations. In general terms, the Delphi method assumes that the opinion of experts can have a scientific application (48). It consists of a participatory methodology that aims to generate consensus, where several experts participate, building consensus between their ideas on the subject in question, but without direct confrontation of opinions (49, 50). To this end, it implies a series of anonymous questionnaires with the particularity of the respondents having access to the group's statistics (48). This method has been used both in the field of social policies and public health (51). It has the potential to obtain viable data that allow informing policy makers (48). The obtained results are based, to a large extent, on personal perspectives, drawing on the experiences and knowledge of the group of qualified specialists carefully selected, with a multidisciplinary vision that allows the establishment of objectives and interventions (52).

Preliminary List of Indicators

The development of the Delphi form is illustrated in **Figure 3**. The set of items/indicators included in the Delphi panel were based on two distinct but still complementary approaches: a literature review and an initial input using expert opinion, collected through the World Café method. Firstly, a review of the existing literature that covered several sources that have been reflecting on issues related to sexual and reproductive health and rights and migration (Medline, Scopus, Web of Science, and the Cochrane Database of Systematic Reviews), including reports published by internationally recognized sources (WHO, UNFPA, and/or UNAIDS), was implemented to better understand the scenario of the current needs and gaps in existing data, and the nature of indicators that the project should entail (29, 41, 53–64). This collection of items resulted in a first list of 536 entries, which were divided into monitoring indicators (447 items) and good practices (88 items). Monitoring indicators are understood as standardized measures, which allow measuring processes that change over time and are considered essential for the creation of health policies (65). Good practices can be defined as an action, which can be compared with an alternative action and where can be established a link between this action and some desirable outcome (66). In other words, the good practices concern measures that must be taken, indicators concern ways of quantifying the impact of the measures and/or helping to define better or more appropriate measures.

For the presentation on the questionnaire forms, and in the subsequent analyses, the identified indicators were classified into three dimensions in accordance to the WHO operational definition of SRH (10): (1) Sexual Health, (2) Reproductive Health and (3) Social-Structural Factors. The indicators were finally subjected to segmentation between: (a) indicators already available in national statistics; (b) indicators that are not available and that should be monitored through regular population-based surveys; and (c) indicators that are (or can be made) available through administrative records or surveys conducted by healthcare units.

Secondly, complementary to this literature-based collection of items, a World Café (WC) (67) session was undertaken to enable obtaining new indicators, potentially different from those obtained through the literature review and more adapted to the national context. The WC method can be defined as a structured conversational process, that facilitates a group discussion, focused on a specific theme and that allows the construction of "collective wisdom" (68). The WC encourages people to speak in a relaxed environmental context. It is based on the assumption that *cafes* provide a creative atmosphere (69). Another assumption is that small group organization facilitates learning, in addition to being less intimidating, and allows everyone the opportunity to express themselves and comment others' ideas (70). For this purpose, a group of 15 experts and stakeholders were drawn from different relevant fields (Academia, Non-governmental Organizations [NGOs], Policymaking, from Healthcare Practice and Civil Society Organizations; multiple affiliation was possible). These specialists were selected due to their experience and expertise in the field and were asked to think about SRH indicators, in general, and specific SRH indicators suitable for migrant populations while considering the three dimensions of WHO (Sexual Health, Reproductive Health, and Social-Structural factors). Items listed by the specialists, together with the ones from the initial set were included in the final list of items launched for discussion within the Delphi panel.

The full list of items was then reviewed by four members of the research team who assessed the items' relevance and suitability. The following five criteria were used in the review of the items: (1) *Repetition or equivalence*. In situations of equal or quite similar indicators, the one that was formulated more clearly was chosen. (2) *Adequate clarity/depth*. Items that were not too abstract / vague / general were privileged. On the other hand, items that were too specific were avoided, as they could be outside the domain of some of the experts. As example "Time frame and coverage of national policy on abortion and fetal sex determination." This item mixes time frame with coverage, therefore being unclear. (3) *Link to the theme*. Items more directly close to the SRH intervention areas were

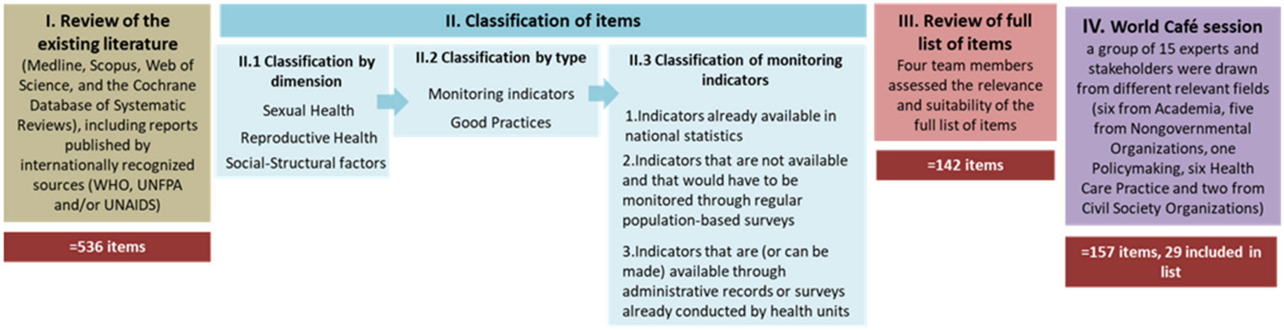


FIGURE 3 | Development of the Delphi form.

privileged. As example: "Percentage of people protected against catastrophic/impoverishing out of pocket health expenditure." It falls out of the SRH scope. (4) *Feasibility*. Since the outcome indicators are indicators derived from statistics or administrative records, it was accounted whether the indicator could be measured. As example "Percentage of facilities that report not experiencing a stock-out of a modern form of contraception in the past 6 months." This item was not included as it would imply monitoring the inventories of all facilities at the national level. Note that no distinction was made between existing indicators and indicators that would need to be created. (5) *Unidirectionality*. Since the objective of the outcome indicators is to be collected over time, in order to understand the effectiveness of the implemented measures, only unidirectional indicators were chosen. As example "Percentage of people who have had more than one sexual partner in the past 12 months." In a public health frame, a greater number of sexual partners can imply a greater risk of chronic diseases (71). However, the freedom of choice in matters relating to own sexual life is a sexual right.

Each item was evaluated by two investigators who were unaware of their peers' endorsement (blind process). In case of doubt or disagreement, the item was discussed by the extended team of four members. A list of 142 items was reached to be discussed with the Delphi panel.

Delphi Panel Recruitment and Formation

In order to tap on an adequate range of perspectives regarding SRH and migrants, a comprehensive list of experts and stakeholders with knowledge and experience in SRH among the migrant population living in Portugal was created. No quota criteria were used in relation to gender or geographic area of intervention, although recruitment has been the most inclusive as possible. In order to obtain an exhaustive list of participants, a web search was carried out on institutional sites of NGOs, civil society organizations, scientific societies, research, and teaching institutions. Research team members attended scientific events (congresses, seminars and workshops) in order to be able to establish personal contact with potential participants that had not been previously identified. After initial contacts were made, additional participants were included through a snowball referral. Snowball sampling, also known as "chain-referral-sampling" is a convenience sampling method (therefore not probabilistic) in which some of the participants recruit new participants through their network of contacts (72, 73). Attempts were made to distribute these sectors as evenly as possible. The following rationale was used to select members of each sector:

1. *Academia*—only demography, birth and related specialties were directly considered. In the case of migrations scholars, they were only considered if they were linked to the previously indicated specialties, or to migrations and health, i.e., migrations and demography, migrations and birth, migrations and health, etc. The specialties of family, sexuality and gender identity, gender violence or gender equality were not considered relevant to the case, unless they had some relation to the themes of intersectionality or migration.
2. *Civil society*—namely experts from migrant associations. Only those that acted on sexual, reproductive, intersectional, gender, and sexuality were considered. Recreational, legal rights, and support for young people associations were not considered.
3. *Non-governmental organizations*—only those having a professional activity related to migrations and health and to SRH were considered. For example, for the promotion of sexual and reproductive health and rights, or for the human rights of women in childbirth. Gender equality actors were not considered.
4. *Healthcare services or organizations*—comprised professionals from three sub-areas: (a) Public health, if they were specialized in working with migrant populations; (b) Gynecologists, obstetricians, and urologists; and (c) Sexologists, except for specialists in childhood sexology.
5. *Governmental organization*—included members of the central and local administration, and members of public institutes that had some connection to the issues under analysis.

Out of the 137 potential participants that had been initially identified, it was not possible to obtain a response from 28, either because they did not answer to the formal invitation sent by email, or because the email has bounced back. Furthermore, sixteen people were excluded because they replied to not having enough knowledge about the topic (though filling in the inclusion criteria). Only the responses of those 82 participants who fully completed the form (from 93 participants who accepted to participate) were used for this analysis.

Development of Delphi Questionnaire

The Delphi was designed using Limesurvey® online survey system. An invitation to participate was sent with the survey link to the questionnaire with a personalized access code, thus ensuring data confidentiality between experts' answers. Along with the questionnaire forms, all participants received an online consent form informing them on the project aims and their rights. The form was made up of six sections:(1) Introduction to the study and informed consent, (2) Socio-demographic characteristics, (3) Monitoring indicators of Sexual Health in Portugal, (4) Monitoring indicators of Reproductive Health in Portugal, (5) Monitoring indicators of social-structural factors with an impact on Sexual and Reproductive Health, and (6) Evaluation of good practices in Sexual and Reproductive Health. With reference to the indicators, sections Results, Discussion, and Conclusion, included information on how the indicator would be collected (by surveys of the population, through administrative data or through official statistics). Initially, three rounds of Delphi were planned (74). However, in agreement with certain literature that argues that it is possible to finish the panel at the end of the second round in case a satisfactory consensus is reached (75), the high consensus observed at the end of the second round determined that an additional third round was not needed. For each round, the opinion of the panelists about the suitability and relevance of each item were collected using a five-point Likert scale. The formulation of the questions and the answer options were the same in both rounds. In the sections that concerned the indicators, the replies were collected through the following item: "In your opinion, what is the relevance of each of the following indicators for the evaluation / monitoring of Sexual Health in Portugal?" and were recorded on a five-point Likert-type scale (*1 = Totally irrelevant, 2 = Irrelevant, 3 = More or less relevant, 4 = Relevant, 5 = Totally relevant*). In the section concerning good practices, replies were collected through the following item: "In your opinion, to what extent do you agree with the fact that each of the following items is good practice in the field of Sexual and Reproductive Health in Portugal?" and were recorded on a five-point Likert-type scale (: 1 = *"I strongly disagree"*, 2 = *"I disagree"* 3 = *"I neither agree nor disagree,"* 4 = *"I agree,"* 5 = *"I strongly agree"*) (41, 76). Additionally, both sections included a "*no opinion/don't know how to answer*" option. Furthermore, an open question was included at the end of each section of the Delphi form, asking participants to propose new indicators or to suggest potential changes to the already included indicators.

Round 1 took place between 18 February 2020 and 5 March. Round 2 took place between 12 March 2020 and 31 March 2020. At the end of each round, the participants were presented with the anonymous aggregation of the results regarding the items approved and rejected. In the second round, participants had access to aggregated responses in items where no consensus had been reached with the aim to question the relevance of the indicators and their agreement with good practices. To reduce the dropout rates and the effect of non-response bias, personal reminders without inclusion in BCC (Blind Carbon Copy), were sent to the participants who did not complete the survey within the specified time and deadlines were extended.

Data Analyses

Regarding group agreement rules, in Round 1, the same criterion was followed as that used in Freitas et al. (41) where the approval and rejection decision were based on the following criteria: 50% of "4" or "5" and at the same time no more than 1/3 (33.3%) of "1" or "2" would be accepted. Items with more than 50% of "1" or "2" would be rejected. In Round 2, the criterion was more demanding and based on literature that suggested as a criterion values between 60 and 90% according to what the researchers considered meaningful (77), in this way, only items that had more than 75% of "4" or "5" responses were approved. Items with lower approval percentages would be rejected.

In order to explore the obtained results, approval rates were used as a measure of consensus (78) and no opinion rates were calculated (79). The level of consensus among the panelists was assessed through the coefficient of variation (mean/standard-deviation) (80). The cut-off referred for a good degree of consensus was between 0 and 0.5 (81). This analysis was complemented with Kernel Density curves as a complementary method for analyzing panelists' consensus (41). Mean values were calculated by dimension and by round.

In order to analyze the changes of opinion by panelists between Round 1 and 2, the McNemar Test was used (82, 83). This test is similar to the chi-square test, but applicable to paired samples and dichotomous variables (2X2). It allows perceiving the change *vis-à-vis* stability of the panelists' position. The null hypothesis is that the respondents 'opinion does not change between R1 and R2 and the alternative hypothesis that the respondents' opinion changes between R1 and R2, either for greater acceptance or for greater rejection. A $p < 0.05$ was considered statistically significant.

Data analyses were done using Microsoft Excel 2011 and SPSS versions 23.

RESULTS

Panel Participation

Of the 137 initial contacts that were selected to participate in the Delphi process, 93 were considered eligible (68%). The remaining 16 were excluded for the following reasons: 5 have reported insufficient knowledge on the topic, 2 no longer held positions in the organizations they represented, 1 was on medical leave and 8 refused to participate. Of these 93 eligible contacts, 90 participants filled in the Delphi form at the first round, which represents 97% response rate. Eight questionnaires were incomplete and were not considered for the analyses, resulting in a total of 82 participants in Round 1. Flowchart for the selection of panelists is presented as **Annex 1**. In the second round, a total of 68 participants responded which corresponds to a dropout rate of 17%. For a clearer portrayal, please refer to **Table 1**.

The following sample characterization concerns the 82 participants who fully completed the first round. The majority

TABLE 1 | Delphi panelists' characteristics (%).

	n	%
Type of panelist		
Health professionals or researchers	66	80.5
Stakeholders	16	19.5
Gender		
Female	62	75.6
Male	20	24.4
Age		
26–45	42	51.2
46–70	40	48.2
Qualifications		
Secondary	1	1.2
Bachelor	17	20.7
Master	23	28.0
Doctorate	41	50.0
Professional activity		
Research/Teaching	36	43.9
Healthcare	23	28.0
NGOs/Civil society	12	14.6
Governmental Organizations	11	13.4
Years in the professional activity		
1–16	44	53.6
17–45	38	46.3
Municipality of activity		
Lisbon	63	78.8
Other municipalities	19	21.2
Field of Science		
Social sciences	50	61.0
Medical and health sciences	24	29.3
Natural sciences	3	3.7
Humanities	3	3.7
Engineering and technology	1	1.2
Field of clinical specialty		
Sexual Health	9	11.0
Reproductive Health	10	12.2
Minority health	5	6.1
Migrant health	4	4.9
Health equity	3	3.7
Social rights	1	1.2
Sexual violence	3	3.7
Field of Intervention		
Sexual Health	28	34.1
Reproductive Health	22	26.8
Minority health	23	28.0
Migrant health	16	19.5
Health equity	19	23.2
Social rights	28	34.1
Sexual violence	24	29.3
Field of Investigation		
Sexual Health	11	13.4
Reproductive Health	16	19.5
Minority health	10	12.2
Migrant health	17	20.7
Health equity	21	25.6
Social rights	21	25.6
Sexual violence	11	13.4

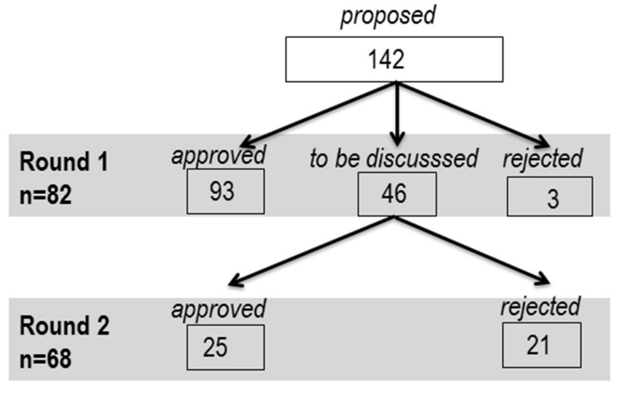


FIGURE 4 | Flowchart for items endorsement/rejection per round and dimension.

of the sample (80.5%) was consisted of experts. More than three quarters (76%) of the panelists were women. Average age was 46 years. Considering their main activity, panelists were classified as health professionals and academics, or stakeholders (associative leaders, members of governmental and non-governmental organizations and political positions). On average, panelists attended their professional positions for 17 years. Academic training was classified according to the Frascati manual (84) and, according to this classification, training in social sciences (61%) and health (29%) predominated as background areas of the participants. The graduates of health sciences were all from the group of researchers; in the graduates of human and social sciences group, a greater diversity was observed, although researchers and health professionals also prevailed. In the graduates of human and social sciences, some diversity was also observed regarding the contexts of activity, with research and teaching predominating. Graduates of health and medical sciences worked majorly in the health sector.

Indicators

From 142 items included in Round 1, 93 items were immediately endorsed to be integrated in the final list, 46 proceeded to Round 2 due to absence of consensus, and 3 were rejected. From the 46 items evaluated in Round 2, 25 were approved and 21 were rejected (**Figure 4**).

Item Analysis

From the initial list of 142 proposed items, a consensus was reached on 118 items (83%). The retained items are presented as **Annex 2**. The distribution of the consensual items by dimension and sub-dimension was the following: *Sexual Health* Indicators (31), *Reproductive Health Indicators* (32), *Social-Structural Factor Indicators* (15), and *Good Practices* (37). A detailed description three groups of indicators and one group of good practices can be found in **Table 2**.

In Round 1, 93 items reached consensus by the absolute majority ($5 > 50\%$ and $1 + 2 < 33.3\%$) and three items did not reach consensus ($1 + 2 = > 20\%$). In Round 2, the rule of

TABLE 2 | Number of indicators proposed, approved or rejected, by dimension and sub-dimension.

	Proposed (*n*)	Approved (*n*)	Rejected (*n*)
Sexual Health	*34*	*32*	*2*
Comprehensive education and information	10	10	0
Gender-based violence prevention, support, and care	8	8	0
Prevention and control of HIV and other sexually transmissible infections	11	11	0
Sexual function and psychosexual counseling	5	3	2
Reproductive Health	*42*	*33*	*9*
Contraception counseling and provision	8	5	3
Fertility care	9	8	1
Antenatal, intrapartum and postnatal care	21	16	5
Safe abortion care	4	4	0
Social-Structural Factors	*27*	*15*	*12*
Cultural and social norms around sexuality	5	3	2
Gender and socioeconomic inequalities	14	6	8
Human rights	5	4	1
Laws, policies, regulations, and strategies	3	2	1
Good practices	*39*	*38*	*1*
Total	142	118	24

TABLE 3 | Number of indicators, approved or rejected, by group decision rules and round.

	Round 1		Round 2	
	Absolute majority approval	Absolute majority rejection	Qualified majority approval	Qualified majority rejection
Sexual Health	20	0	12	2
Reproductive Health	28	1	5	8
Social-Structural Factors	12	1	3	11
Good practices	33	1	5	0

TABLE 4 | Mean values for response on Likert scale, Coefficient of variation, Percentage of "agreement" responses and Percentage of *"no opinion"* responses, by dimension.

	Mean	Coefficient of Variation	% of Agreement	% of No opinion
Sexual Health	4.31	0.21	84.22	0.86
Reproductive Health	4.23	0.22	80.23	2.86
Social-Structural Factors	3.98	0.25	71.66	1.93
Good Practices	4.47	0.21	88.43	1.38

Qualified Majority (5 +4> 75%) was applied, with 25 items being endorsed and 21 items being rejected (**Table 3**).

An analysis of the mean values calculated by dimension (**Table 4**) shows that the highest number of endorsed items was obtained in the *Good Practices* dimension and the lowest in the *Social-Structural Factors* dimension. The consensus was higher in the *Sexual Health* and *Good Practices* dimensions and lower in the *Social-Structural Factors*. The percentage of agreement (defined as the percentage of responses in the *"I agree"* or *"I strongly agree"* values of the scale) was higher in the *Good Practices* dimension and lower in *Social-Structural Factors*. Finally, the "no opinion/don't know how to answer" were more frequent in the *Reproductive Health* dimension.

In order to synthesize the collected information, the results for the agreement values at the end of Round 2 are projected in **Figure 5**. They represent the sum of the two rounds and can be interpreted as an endorsement rate.

Figure 5 shows that there is a high endorsement of items belonging to the *Good Practices* and *Sexual Health* dimensions. There is a high concentration of points near the outer vertex, with only one of the items having an approval rate below 75% (63% precisely). In comparison, the items from the *Reproductive Health* dimension have a lower acceptance rate while the items from the *Social-Structural Factors* demonstrate a greater concentration below 75% of acceptance, with the points scattered along the top vertical line.

TABLE 5 | Mean values for the coefficient of variation by round and dimension.

	Mean R1	Mean R2
Sexual Health	0.218	0.204
Reproductive Health	0.229	0.225
Social-Structural Factors	0.244	0.276
Good Practices	0.211	0.206

Coefficient of variation scale ranges from 0 to 1; higher values meaning higher variation.

In the *Reproductive Health dimension*, the highest endorsement rate was found for items referring to the safe abortion care, such as *"Number of terminations of pregnancy, total and at the option of the woman," "Number of health services that offer safe termination of pregnancy,"* and *"Number of hospitalizations due to unsafe abortion."* Regarding *Social-Structural Factors* and *Good Practices*, a regular pattern was not identified in the items with a higher endorsement rate.

"No Opinion" Rates

"No opinion" rates were used as indicators of unfamiliarity. Considering that there is a pattern of no opinions, they should be considered not missing at random. With these arguments in consideration, **Figure 6** represents the distribution of "*no opinion rates*" by dimension. The analysis indicates that "*no opinion*" rates are quite reduced within the *Sexual Health* dimension, while a wider distribution is observed among the *Reproductive Health* dimension and for *Socio-Structural Factors*. Within the *Reproductive Health* dimension, the greatest uncertainties are observed in items related to vaccination, such as *"Number of women of reproductive age who received tetanus vaccine"* and *"Coverage rate of tetanus vaccine by birth cohort."* Within the *Socio-Structural Factors*, the highest "*no opinion*" rate was observed for the item *"Average age at divorce."*

Consensus Analysis

High consensus was observed for all dimensions and in both rounds. The values of the coefficient of variation varied between 0.2 and 0.4 in Round 1, and 0.2 and 0.3 in the Round 2. Although the values are always of high consensus, it can be observed that in Round 1 there is a greater consensus on *Good Practices* and a fewer consensus on *Social-Structural Factors* (**Table 5**). In the passage to Round 2, the consensus becomes higher in 3 of the 4 dimensions, the exception being *Social-Structural Factors*.

The Kernel Density curves (**Figure 7**) demonstrate that, in the first round, the distributions are closer to the leptokurtic type (meaning flattened ends and more pronounced mean values), especially regarding the dimensions related to *Sexual Health* and *Good Practices*. The latter is where the greatest concentration on the right is observed, which means greater agreement with the items. In Round 2, the distributions are closer to the Platykurtic type (meaning lower agreement, since there is flatness along the line) and tend to concentrate more on the value 4 than on the 5.

Figure note: Each density curve represents the distribution of the mean of the panelists' responses on a 5-point Likert scale by dimension and round. The curves of the first round relate to the totality of the items that were discussed and include the responses of the 82 participants. The curves corresponding to the second round only refer to the items that were discussed in the second round because they did not have consensus in the first round.

Changes of Opinion Between Round 1 and Round 2

In order to identify the changes in opinion between the two rounds, a series of McNamar tests was used. After the items' dichotomization (1, 2, or 3 = 0 *"non-agree"*; 4 or 5 = 1 *"agree"*), statistical significance ($p < 0.05$) was observed in six out of the 46 items. Since for each item, the changes can be 2-folded (participants who disagree in R1, agree in R2 and participants who disagree in R1, agree in R2), the most relevant changes were indicated. Significant changes of opinion were observed in two items of the *Reproductive Health* dimension: The *"Number of people who have undergone sterilization,"* 50.0% of the participants who disagreed in R1 changed their position to agreement in R2. Also the *"Number of women who comply with gynecological surveillance recommendations"* in which 72.2% of the participants that disagreed with this item in R1 agreed in R2.The remaining changes of opinion occurred in items within the *Social-Structural Factors* dimension: *"Number of people with health insurance"*—42.5% of the participants who agreed with this item in R1, disagreed in R2; *"Economic well-being"*—35.1% of the participants who agreed with this item in R1, disagreed in R2; *"Gross Divorce Rate"*—42.9% of the participants who agreed with this item in R1, disagreed in R2; and finally the item *"Occupancy rate for Portuguese language courses for foreigners"*—48.8% of the participants who agreed on R1 disagreed on R2.

DISCUSSION

The aim of this study was to reach consensus on what constitute good practices in the SRH field, with emphasis on SRH equity across native and migrant populations, and to identify the most relevant and inclusive indicators in accordance with the Age Gender and Diversity framework to plan, program, implement, monitor and evaluate SRH in Portugal. Good practices and indicators were grouped into the WHO operational definition of SRH: Sexual Health; Reproductive Health; and Social-Structural Factors (29). The items that received the highest approval rate for each sub-dimension were selected for discussion.

Sexual Health

Within the Sexual Health dimension which covers areas ranging from comprehensive education and information to sexual function and psychosexual counseling, the items with the highest endorsement rate concern the prevention and control of HIV and other sexually transmitted infections (STI), such as *"Number of new cases (incidence) of sexually transmitted infections," "Number of HIV/AIDS cases (prevalence),"* or *"Coverage of antiretroviral therapy."* This occurs even though there were panelists with experience and knowledge in all the other sub-dimensions of sexual health. This is in line with the investment in research intersecting sexual health and migration, where the thematic of the prevention and control of HIV/STI seems to have been

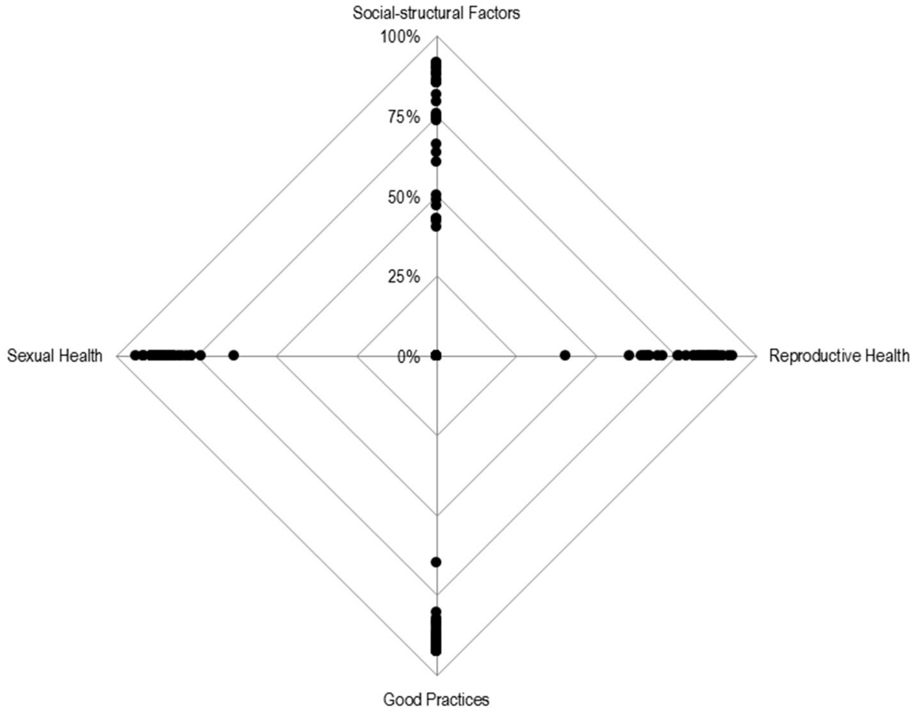


FIGURE 5 | Radar chart for the percentage of responses strongly agree + agree per dimension. Each point represents the percentage of agreement response ("I agree" and "I strongly agree") for each item, per dimension.

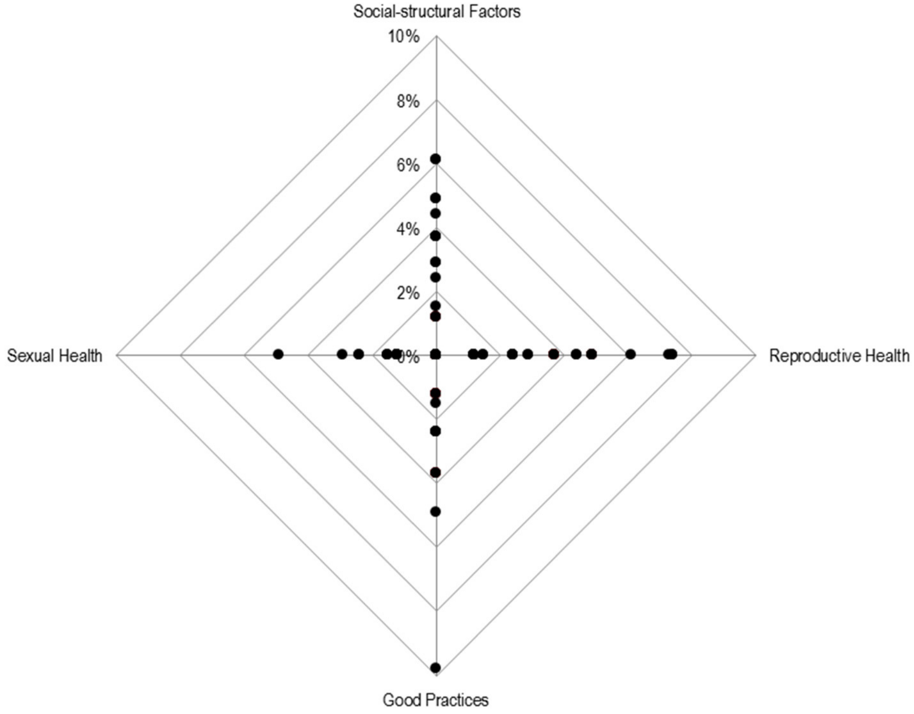


FIGURE 6 | Radar chart for the percentage of no opinions per dimension. Each point represents the percentage of no opinions for each item, per dimension.

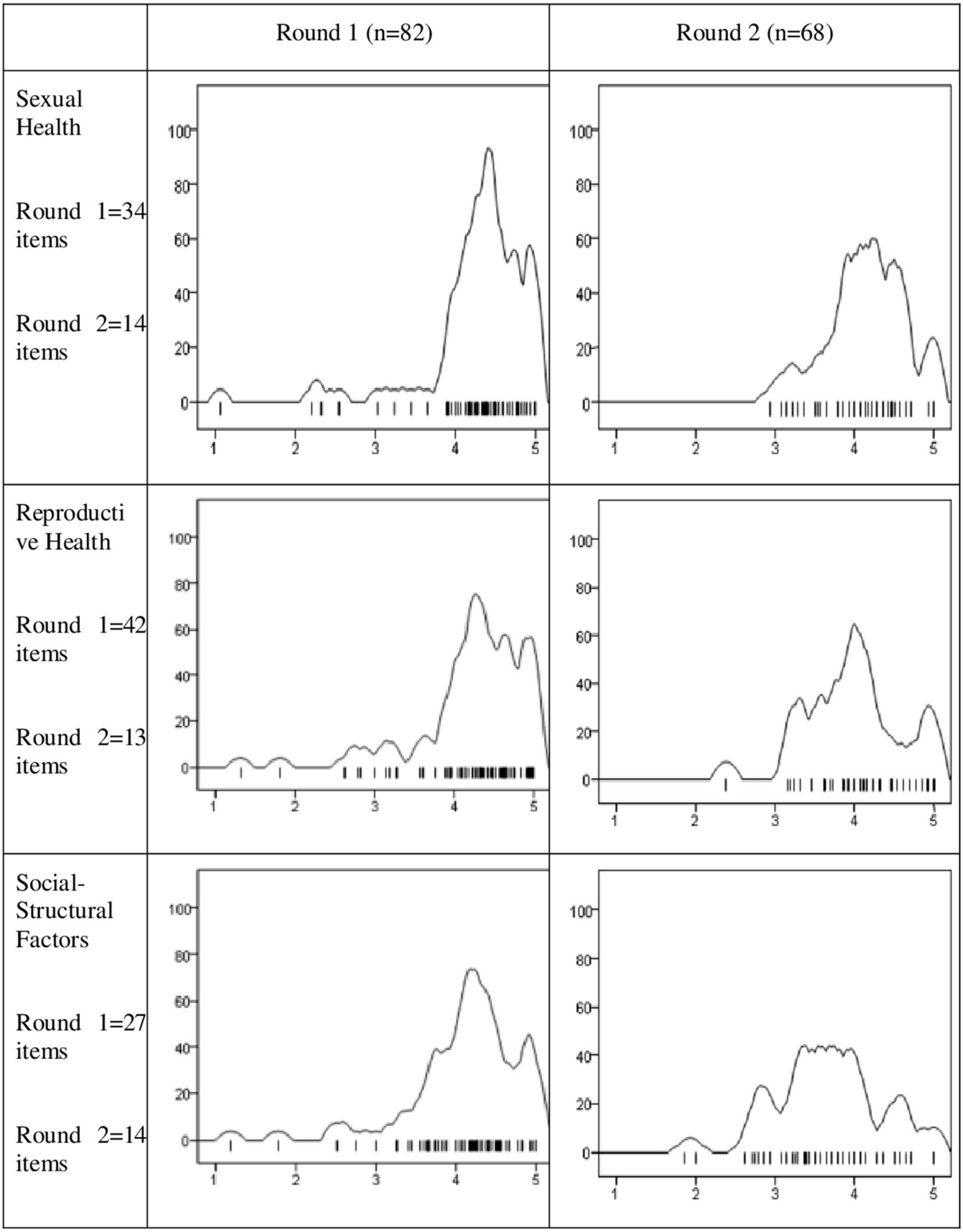


FIGURE 7 | Kernel Density Curves for the panelists mean responses for each round and dimension.

receiving more attention, while comprehensive education and information; gender-based violence prevention, support, and care; and sexual function and psychosexual counseling remain understudied areas (85).

Although exhibiting decreasing numbers, HIV/AIDS is a persisting global phenomenon. In Portugal, the cumulative number of people with HIV and AIDS is released annually by the Directorate-General for Health (86) and the PORDATA portal (the Database of Contemporary Portugal official statistics) only discloses data on HIV infection at the AIDS stage. This is one of the cases in which the data is segmented by national origin. In 2018, 61% of new infections were diagnosed in people born in Portugal, 19% in sub-Saharan Africa and 11% in Latin America (87). In addition, the WHO strategy on STIs in people of reproductive age proposes the improvement of the available data by paying special attention to the disaggregation by sex and age groups (88).

Previous studies have also highlighted the need for on-going monitoring of risk behaviors, STIs, and accessing services among migrant populations, as well as further research to help understand its intersecting inequities (89). It is important to identify key populations (including migrants) to be targeted with tailored HIV prevention activities and treatment options, as well as services that provide care and support based on the recipients' different backgrounds and needs (90, 91). Regarding the *Comprehensive education and information* sub-dimension, the panel of experts endorsed the importance of the *"Number of people with levels of sexual health literacy considered adequate."* An adequate level of health literacy would contribute to making informed decisions, which contribute to an increase in migrants' health and empowerment (92, 93).

Within the sub-dimension *Gender-based violence prevention, support and care,* high endorsement was found for the item *"Number of people who correctly identify gender and sexual violence."* Monitoring gender violence and reducing its structural risk factors remains a vital public health priority (94). Although indicators on gender violence and sexual violence in Portugal have been published annually since the beginning of the millennium (e.g., APAV—Portuguese Association for Victim Support—which has a specialized support unit for migrant and discrimination victims) (95), there is lack of information regarding the extent to which the population is aware of the forms gender and sexual violence can take. Sexual violence victimization has been associated with a broad range of health and risk behaviors, including posttraumatic stress disorder, depression, eating disorders, substance use, smoking, and poor self-rated health (28). In Portugal, despite the presence of organizations such as the National Observatory of Violence and Gender that conduct victimization surveys, this dimension (the correct identification of gender violence) is not properly accounted for. It should be noted that both gender and sexual violence must be carefully defined, to avoid generalizations or create an overgeneralized concept not allowing the identification of different potential forms of violence. The need for sexual and gender-based violence conceptualization is well-illustrated in a recent study in the context of European asylum reception centers, which showed a disparity between what is, or what is not considered a violent behavior among residents and professionals, the latter considering more acts as violence then the former (96). Public health policies should be adapted to the cultural and structural context, and for that comparing sexual and gender-based violence conceptualization between migrants and hosting population is crucial. The development, implementation and monitoring prevention programs in this area would benefit from a comprehensive societal conceptualization of sexual and gender-based violence considering the influences of individual, relational, community, and societal factors (96).

In addition, within the sub-dimension *Gender-based violence prevention, support and care,* the item *"Number of reports of obstetric violence"* deserves attention. Although sexual and reproductive rights are protected under Portuguese law, obstetric violence is an existing phenomenon that currently lacks a legal framework and remains difficult to quantify. The survey "Childbirth Experiences in Portugal," carried out by the Portuguese Association for Women's Rights in Pregnancy and Childbirth, collected responses of more than 3,800 women and revealed that 43.5% of the women surveyed did not have the desired delivery, however very few of them filed complaints. The proper identification of obstetric violence would contribute with a 2-fold benefit to SRH: (1) it is a form of gender-based violence that would be identified and, potentially prevented; and (2) based on women's experiences and perceptions during childbirth, as well as on the normative pattern of obstetric management it would allow the provision of a physically and mentally healthy birth (97, 98).

Inequities in the quality of care must be understood in light to the intersecting challenges migrant women face due to language difficulties, lack of familiarity with healthcare systems, and discriminatory attitudes (99).

Within the sub-dimension *"Sexual function and psychosexual counseling,"* the items with the highest endorsement were *"Number of new cases (incidence) diagnosed with sexual dysfunction,"* and *"Number of people who consider that have a healthy sexuality."* Sexual dysfunctions are a multifaceted phenomenon that can be understood as the reason that prevents individuals from experiencing satisfaction from sexual activity (29). Although there are several scales already validated among Portuguese samples (88, 100), longitudinal studies that can assess the evolution of prevalence of various sexual dysfunctions are still missing. Additionally, more knowledge is needed concerning the individual sexual well-being of the Portuguese population, using positive indicators of sexual health such as sexual satisfaction (101, 102). An assessment of subjective sexual well-being, defined as the cognitive and emotional assessment that each person makes of their sexuality (103), was applied as part of an international study—the Global Study of Sexual Attitudes and Beliefs (104) but Portugal did not participate. It would be important to replicate the study in the Portuguese population, including migrants and contribute to overcome the scarcity of data intersecting sexuality and migration.

Reproductive Health

Reproductive Health dimension embraces the WHO definition of reproductive health and rights, such as the right make a free and responsible decision on the number, spacing, and timing of their children; ability to obtain the appropriate information and means to make such a decision; and the right to decide on reproduction without threat of discrimination, coercion, and violence (105). Despite the significant reduction in the number of cases of unwanted pregnancy in the last two decades worldwide, the phenomenon continues as a significant burden globally, with ∼16 million (11%) of all births worldwide attributed to young women aged 15–19 years (106). In Portugal, the interruption of pregnancy on women's request can be seen as a way of regulating fertility in order to limit births of unwanted pregnancies (107). A study revealed that more than 95% of all interruptions of pregnancy performed in the country, were performed in hospital settings (86, 108). In 2018, 20% of all women who interrupted their pregnancy in Portugal were foreigners (i.e., 3.098 in 11.827) (86, 108, 109). Although there are several causes of unintended pregnancy, one of the most important tools that can help in preventing them is the timely use of emergency contraception and access to primary health facilities that provide family planning services. According to the WHO, all women and girls at risk of an unintended pregnancy have the right to access emergency contraception and these methods should be routinely included within all national family planning programs (109, 110). As an indicator, the number of sold emergency contraception pills can inform on the number of terminated unwanted pregnancies, but also may point to the ineffectiveness of regular contraception. Despite its relevance, currently there is no official data available in Portugal.

The items *"Number of women who comply with gynecological surveillance recommendations"* and *"Number of family planning users who were counseled, referred or treated for infertility"* had the highest level of endorsement by the expert panel within the *"Fertility care"* sub-dimension.

Ensuring universal access to SRH services is incorporated in Target 3.7 of the United Nations' Sustainable Development Goals (SDG). Refugees, migrant women and children are at particular risk of being excluded in achieving this target, since they hold a higher chance of maternal death and maternal near-miss events (111, 112). In this context, gynecological surveillance is important to prevent potential complications, with the recommended number of annual visits depending on the woman's age and the existence of previous problems. In Portugal, the relevant data can only be obtained at the aggregate level (and excludes the entire private sector). Therefore, the alternative would be to resort to population surveys. The second item with highest approval rate in this sub-dimension, concerning the identification and management of infertility, must be analyzed in conjunction with others, such as the quality of services received. Currently, public and private offers for infertility treatments are available. However, several negative beliefs and representations block access to these services. A survey on this topic of a representative sample of the Portuguese population (113) estimated that 9.8% of women aged between 25 and 69 years had already had problems with pregnancy, of which 43.4% had consultations for reasons of infertility. The number of people doing infertility treatments can serve as an orientation point of the number of people who, regardless of constraints, are referred in order to enjoy a desired pregnancy.

Within the *"Antenatal, intrapartum and postnatal care"* sub-dimension, the expert panel considered as most important to focus on *"Gestational age of women at the first consultation of Gynecology-Obstetrics"* and *"Maternal mortality rate, by cause."* The reduction in the global maternal mortality rate is part of the sustainable development goals for 2030 (24, 114). According to the WHO although the maternal mortality rate and rate of complications in childbirth in Portugal has been reduced, the global maternal mortality rate is still unacceptably high. The high number of maternal deaths in some areas of the world reflects inequity in the access to quality health services and highlights the gap between rich and poor countries. A distinction is also made between "maternal mortality" (death of women during pregnancy or within 42 days after termination of pregnancy, excluding external causes) and "late maternal mortality" (when it concerns obstetric causes, direct or indirect, after 42 days, and less than a year after termination of pregnancy). It is proposed (second most consensual item) that this indicator should be disaggregated by the main cause of death in order to better understand this multifaceted phenomenon and the areas of intervention. The recommendations of the Portuguese General Directorate of Health imply that a normal pregnancy should have at least six consultations that can identify potential risk factors and needs for intervention. However, the proportion of pregnant women who act in line with this recommendation is still unknown.

Additionally, within the *"Safe abortion and care"* sub-dimension, the importance of *"Number of terminations of pregnancy, total and at the request of the woman"* was highlighted. The situation of induced abortion has changed markedly over the past few decades, with abortion being legalized and its rates dropping in many developing countries in the world (115). The Guttmacher Institute report shows that abortion rates are similar in countries where abortion is highly restricted and where it is broadly legal (116). In Portugal, the Directorate-General for Health compiles the number of pregnancy interruptions that occur in public and private health facilities. The reports with these numbers contemplate time series and present a characterization of the women who utilized this service (117). As in other cases, it is an ambivalent indicator, especially when it comes to interruptions that occur at the request of women.

Evidence gathered by the international research collaboration ROAM (reproductive outcomes and migration) from 20 countries including Portugal shows that culturally diverse guidelines are needed to individualize antenatal care and promote optimal maternal-fetal health outcomes across cultural groups (118, 119). Further research is needed to identify and understand specific vulnerabilities and subsequent action is needed to address the intersecting inequities.

Social-Structural Factors

Social-Structural Factors dimension covers items ranging from *Cultural and social norms* a*round sexuality* to *Laws Policies, regulations, and strategies*.

The two highest endorsed items in the *"Cultural and social norms around sexuality"* sub-dimension were *"Number of complaints of female genital mutilation"* and *"Number of people who report that their partner's sexual pleasure is important for the quality of the relationship."*

In Portugal, Female Genital Mutilation (FGM) is considered an autonomous crime according to article 144 A of the Criminal Code of 2005. The applicable penalty is 2 to 10 years in prison. The currently existing numbers of FGM (64 cases in 2018 and 129 cases in 2019) are the result of cases identified by health professionals, who received training under the *"Healthy Practices"* project, which covered groups of health centers with the highest number of women at risk. Although no reliable data exist, estimates point to 6,576 women living in Portugal already subjected to FGM (120).

Regarding the second item, no reliable data exist, as sexual pleasure is under-researched and there is only one available measure addressing this sexual health dimension (115, 116). Furthermore, the existing research does not take a partner-centered approach (121). Sexual pleasure is at the heart of sexual rights advocacy (122, 123) and our results support this view. The inclusion of items related to interpersonal pleasure would allow a more complex and accurate picture on the interpersonal nature of sexual pleasure.

Within the *Gender and socioeconomic inequalities* sub-dimension, the highest endorsement was found in the items *"Rate of adherence to cervical cancer screening"* and *"Paternity leave utilization rate."* Strategies to reduce inequalities in adherence to cervical cancer screening are needed, to allow timely diagnosis and improve the sexual life of all women diagnosed after treatment. These include cultural competence in healthcare and having cervical cancer screening information linguistically and culturally adapted (124, 125). Within the scope of the National Program for Oncological Diseases of the Portuguese General Directorate of Health, data on the rate of adherence to cervical cancer screening were released. The applied measure is *"Total Number of Women Tracked/Number of Women Invited."*

The second most consensual item provides an interesting insight into parenting. It is important to know the proportion of fathers who want or have the possibility to take full paternity leave, also because the stay of both parents during the initial period promotes a healthy child development and less overload of domestic tasks for the recent mothers, thus improving their postpartum condition (126, 127). According to OECD data (128), Portugal is one of the countries in this group with the longest duration of paternity leave (21 weeks in 2015), and the share of men among parental leave users in Portugal, as well in some Nordic countries, goes up to 40% or more. Fathers-only Parental leave (formerly Paternity leave) is a relatively recent right, since it was non-existent until the year 1999. Using data from the Social Security data and the number of births available on the PORDATA portal, the Observatory of Families and Family Policies (129), found an increasing tendency with 68% of fathers using their right to paternity leave in 2019.

Research has shown that besides attitudes toward gender roles within the family sphere, the level of knowledge about the parental leave system, the vulnerability on the labor market, and non-universal eligibility are major factors explaining migrant-native differentials in parental leave use. In this sense, parental leave policies need to avoid perpetuating labor market disadvantages by limiting support for work–family reconciliation (130, 131). Further research is needed on the differences in parental leave use between different groups of parents. In the *Human rights* sub-dimension, the expert panel considered important to address the "*Number of complaints for discrimination based on gender identity and sexual orientation.*" Since 2013, the ILGA Portugal Association's Observatory of Discrimination Based on Sexual Orientation and Gender Identity—Lesbian, Gay, Bisexual, Trans, and Intersex Intervention collects, analyzes, and disseminates data on complaints of discrimination (132). However, the collected data lacks disaggregation to provide adequate assessment of this issue (133).

Studies have shown that transgender migrants and migrants who engage in sex work also face higher risk for HIV infection (89). The UNAIDS Gap Report highlights how migrants who engage in sex work face a double stigma because of their immigration status and their engagement in sex work. Adding the fact that stigma and discrimination of living with HIV amplifies their risk of experiencing violence and the barriers to accessing services (134). Of most importance for practice, is the fact that the characteristics of the country of origin and destination (such as access to healthcare, social protection, and social exclusion) influences migrants' risk of HIV infection (134). In the final sub-dimension in this category, "*Laws, policies, regulations and strategies,*" the expert panel considered *"Number of Local Support Centers for the Integration of Migrants (CLAIMs) available to the migrant population"* and *"Percentage of government spending on health, directed at SRH"* to be of highest importance.

According to a recent study, Portugal is one of the three European Union countries (together with Ireland and Spain) that propose their largest range of policies aiming at improving access to healthcare services for migrants (135). In this context, Portugal has founded CLAIMs and has also made efforts toward securing a specific budget for these relevant issues. CLAIMs were founded in 2003 and they help in *"regularization, nationality, family reunification, housing, voluntary return, work, health, education, among other issues of daily life"* (136). The Portuguese CLAIMs network includes already more than 100 centers, provides information and assistance. In the context of the second most consensual item of this sub-dimension, the percentage of government expenditure that is directed to health is available from the Portuguese Directorate-General for Budget and from the Ministry of Finance, with a proposal for a separate breakdown for the Division of Sexual, Reproductive, Child and Youth Health (DSSRIJ).

A final issue deserves attention: the answer *"no opinion"* can be interpreted as an indicator of the areas in which further intervention in terms of dissemination and training may be needed (137).

Good Practices

Finally, the most highly endorsed *Good Practices* by the expert panel were: (1) *"Existence of procedures in healthcare units that* *guarantee the informed choice in SRH*"; (2) *"Health facilities, goods, information and health services related to SRH must be accessible to all individuals and groups without discrimination and free from obstacles*"; (3) *"Existence of evidence-based SRH counseling services"* and (4) *"Existence of laws and regulations that guarantee full and equal access to SRH care."*

In the *Good Practices* dimension, apart from the low *"no opinion"* rate, there is an observed outlier concerning the *"Greater coverage of the reasons why abortion is permitted*" item. In Portugal, two referendums were needed to stablish that voluntary abortion was no longer illegal when performed up to the 10th gestational week in official or officially accredited health services since 2007, thus voluntary abortion remains a fracturing issue in Portuguese society (108, 138).

Strengths and Limitations

This study represents a contribution toward the identification of country-based relevant indicators on the SRH and rights to improve health and well-being for all (139). This research has some limitations to consider. First, although the Delphi panel was consisted of a variety of experts and stakeholders, the convenience (snowball) sampling method may influence the transferability of experts' opinions to that of the wider community of professionals that work in the relevant field and with the population of interest to this study. Even so, an effort was made in order to be the most comprehensive as possible in sampling in order to have a varied sample of panelists. Another limitation of this study is the reduced participation of stakeholders representing migrant communities. Although invitations and reminders were made to various actors in this area, this was clearly the area of intervention in which less adherence was felt. Further studies are needed to investigate and understand the preferences of migrants and their families on how relevant SRH issues should be promoted. Still another limitation linked with the sample of panelists, regards the fact that it included more specialists than stakeholders. On the other hand, a main strength of this study remains on the inclusion of a variety of experts and stakeholders, with diverse professional backgrounds and with extensive experience, underlining their potential for a strong contribution in the area.

Despite the fact that the date of the implementation of the second round (12 to 31 March, 2020) coincided with the first confinement due to COVID-19 pandemic, within the scope of the first state of emergency, declared on 19 March 20 by the council of Portuguese ministers, response rate exceeded the recommend 70% rate as necessary to maintain rigor (41).

Although the findings of this study are intended to be formative rather than definitive, the final set of items is valid and consistent with a range of important dimensions related to SRH areas, and also diverse and inclusive to enable monitoring inequalities.

Future Recommendations for Research and Action

Results highlight the importance of identifying and understanding the origin of health inequalities, inequities, and monitoring the impact on SRH and rights between ethnic/racial minorities and migrant groups. Addressing the social determinants of health inequalities and inequities holds the potential to raise awareness to design appropriate interventions both in terms of access to healthcare and quality of SRH services.

Findings can serve for inspiration to the multiple actors in the field of SRH who wish to protect and promote SRH human rights by building operational links between principles and realities.

In 2007, the ROAM international research collaboration and EURO-PERISTAT project developed an international Delphi survey to recommend migration indicators for national and international monitoring. A strong consensus was attained to include firstly country of birth and secondly length of time in the country in core perinatal health indicator sets. Specific studies were also recommended to complement routine data collection on three other indicators of migration: migration status, receiving-country language capacity, and maternal parents' place of birth as proxy for ethnicity (45). These recommendations remain up to date and should be expanded to the overall SRH issues and across life course and populations to effectively reduce SRH inequities between migrant and receiving-country populations.

The Academic Network for Sexual and Reproductive Health and Rights Policy (ANSER) is a global platform for SRH and rights policy research, education and healthcare delivery that addresses the gap between research and policy in this area. It is a good example of how SRH research findings can be translated into feasible policy and practice by engaging effectively stakeholders at different stages of the research cycle and by taking into account existing and changing political contexts and priorities (140).

Findings can serve as a starting point to awareness-raising actions on the cultural, socioeconomic, geopolitical and legal environment diversity that forms the context for people's lives in different settings and which influences SRH outcomes. They can also serve the basis for providing training to health professionals toward an improved focus on migrants' needs, and effective communication practices (141).

CONCLUSION

This study reinforces the need to address the wide variation of national contexts regarding policy measures to protect migrants' SRH and rights and ensuring their access to basic and essential services—with special emphasis on sexual education, as well as sexual and reproductive justice. The Delphi method, as performed in this study, provided avenues that can be used by the healthcare system to engage in better informed decisions and, more importantly, inclusive and integrative strategies regarding SRH equity. Given the global COVID-19 pandemic, the findings are of special importance since the existing achievements to promote equal access to healthcare and decrease the risk of healthcare-related inequities, were undermined. Results can enable the health systems to adapt to the needs of the migrant population and thus ensure effective and efficient deployment of SRH care structures and processes within the context of inclusive and integrated care. As envisaged throughout the paper, this can be achieved by using the life course approach to plan, program, implement, monitor, and evaluate the relevance of SRH indicators of the populations and across life course.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, under request.

ETHICS STATEMENT

The study was approved by the Ethics Committee of the Centro Académico de Medicina de Lisboa (CAML). All participants received an online consent form, together with the questionnaire forms, informing about the project aims and their rights (e.g., procedures, voluntary non-gratified participation, data confidentiality, dropout option with no consequences.

AUTHOR CONTRIBUTIONS

VA, OS, and AV designed the study and wrote the protocol with inputs from PC, SP, PP, AC, and FM. PC, VA, OS, and AV did the initial survey form, with the review of PC, SP, PP, AC, and FM. VA and PC were responsible for the recruitment of participants and data collection. PC performed data analysis and interpreted the results with the review of VA, OS, and AV. PC, VA, and MS-P wrote the first draft of the manuscript. All authors reviewed, contributed to the article and approved the submitted version.

FUNDING

This work contributes to the study *Fertility, Migration and Acculturation: Intersectional approach to the sexual and reproductive experiences and expectations among Cape Verdean and Portuguese Families (FEMINA project)*, granted by Fundação para a Ciência e a Tecnologia, I.P. with national funding (PTDC/SOC-SOC/30025/2017). It is also a valuable contribute for the study *Promoting citizenship and sexual rights in Immigrant Populations in Portugal: co-production of recommendations and best practices for the reduction of sexual and reproductive inequalities (ProCima project)*, granted by the Asylum, Migration and Integration Fund (PT/2018/FAMI/356). The writing of this manuscript had also the support of Fundação para a Ciência e a Tecnologia, grant number UIDB/04295/2020 and UIDP/04295/2020.

ACKNOWLEDGMENTS

We want to express our gratitude to all experts who composed the World Café as well as the experts who composed the Delphi panel, for their active, invested, and dynamic participation. The authors would also like to acknowledge Mário Augusto for his valuable contributions for the identification of the first set of key indicators through literature review.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2021.656454/full#supplementary-material

REFERENCES

1. IOM. *World Migration Report 2020*. Geneva (2019). Available online at: www.iom.int/wmr (accessed May 11, 2020).
2. World Health Organization (WHO). *Health of Migrants – the Way Forward: Report of a Global Consultation* (2010). Available online at: www.who.int/hac (accessed November 24, 2018).
3. De Schrijver L, Vander Beken T, Krahé B, Keygnaert I. Prevalence of sexual violence in migrants, applicants for international protection, and refugees in Europe: a critical interpretive synthesis of the evidence. *Int J Env Res Public Health*. (2018) 15:1979. doi: 10.20944/preprints201807.0001.v1
4. Mason-Jones AJ, Nicholson P. Structural violence and marginalisation. The sexual and reproductive health experiences of separated young people on the move. A rapid review with relevance to the European humanitarian crisis. *Public Health*. (2018) 158:156–62. doi: 10.1016/j.puhe.2018.03.009
5. United Nations Population Fund. *Gender-Based Violence in Humanitarian Settings in 2014* (2014). Available online at: https://www.unfpa.org/resources/gender-ba%0Ased-violence-humanitarian-settings
6. Bloom G, Katsuma Y, Rao KD, Makimoto S, Yin JDC, Leung GM. Next steps towards universal health coverage call for global leadership. *BMJ*. (2019) 365:l2107. doi: 10.1136/bmj.l2107
7. International Labour Organization. *ILO Global Estimates on International Migrant Workers: Results and Methodology* (2018). Available online at: https://www.ilo.org/wcmsp5/groups/public/---dgreports/---dcomm/---publ/documents/publication/wcms_652001.pdf
8. Dune T, Perz J, Mengesha Z, Ayika D. Culture Clash? Investigating constructions of sexual and reproductive health from the perspective of 1.5 generation migrants in Australia using Q methodology. *Reprod Health*. (2017) 14:50. doi: 10.1186/s12978-017-0310-9
9. Sheikh-Mohammed M, Macintyre CR, Wood NJ, Leask J, Isaacs D. Barriers to access to health care for newly resettled sub-Saharan refugees in Australia. *Med J Aust*. (2006) 185:594–7. doi: 10.5694/j.1326-5377.2006.tb00721.x
10. Vearey JO, Hui C, Wickramage K. Migration and health: current issues, governance and knowledge gaps. In: McAuliffe M, Khadria B, editors. *World Migration Report*. Geneva: International Organization for Migration (2020). p. 212–49. Available online at: https://www.un.org/sites/un2.un.org/files/wmr_2020.pdf (accessed December 13, 2020).
11. PORDATA (2019). Available online at: https://www.pordata.pt/en/Europe/Mean$+$age$+$of$+$the$+$mother$+$at$+$birth$+$of$+$first$+$child-3468 (accessed March 18, 2019).
12. SEF/GEPF. *Relatório de Imigração, Fronteiras e Asilo 2019*. Oeiras (2020).
13. Oliveira CR. *Indicadores de Integração de Imigrantes: Relatório Estatístico Anual 2020*. Lisboa: Alto Comissariado para as Migrações (2020). Available online at: https://www.om.acm.gov.pt/documents/58428/383402/Relatorio+Estatistico+Anual+2020+-+Indicadores+de+Integracao+de+Imigrantes/472e60e5-bfff-40ee-b104-5e364f4d6a63
14. Marmot M, Allen J, Goldblatt P, Herd E, Morrison J. *Build Back Fairer: the COVID-19 Marmot Review. The Pandemic, Socioeconomic and Health Inequalities in England*. London: Institute of Health Equity (2020). Available online at: http://www.instituteofhealthequity.org/resources-reports/build-back-fairer-the-covid-19-marmot-review/build-back-fairer-the-covid-19-marmot-review-executive-summary.pdf (Accessed December 28, 2020).
15. Bambra C, Gibson M, Sowden A, Wright K, Whitehead M, Petticrew M. Tackling the wider social determinants of health and health inequalities:

evidence from systematic reviews. *J Epidemiol Community Health.* (2010) 64:284–91. doi: 10.1136/jech.2008.082743

16. Whitehead M, Dahlgren G. Concepts and principles for tackling social inequities in health: levelling up Part 1 (2006). Available online at: www.euro.who.int (accessed March 25, 2021).
17. Global Health Europe. *Inequity and Inequality in Health* (2009). Available online at: https://globalhealtheurope.org/values/inequity-and-inequality-in-health/ (accessed April 1, 2021).
18. Pickett KE, Wilkinson RG. Immorality of inaction on inequality. *BMJ.* (2017) 356:j556. doi: 10.1136/bmj.j556
19. Shibre G, Zegeye B, Ahinkorah BO, Keetile M, Yaya S. Magnitude and trends in socio-economic and geographic inequality in access to birth by cesarean section in Tanzania: evidence from five rounds of Tanzania demographic and health surveys (1996-2015). *Arch Public Health.* (2020) 78:80. doi: 10.1186/s13690-020-00466-3
20. United Nations. *Reproductive Health Policies 2014.* New York, NY (2014). Available online at: http://www.unpopulation.org (accessed April 5, 2021).
21. Do M, Machado C, Santana P, Car-Reiro H, Nogueira H, Barroso R, et al. Cuidados de saúde materna e infantil a uma população de imigrantes. *Rev MigraçõesNúmero Temático Imigração e Saúde.* (2007) 1:103–27.
22. Harding S, Boroujerdi M, Santana P, Cruickshank JK. Decline in, and lack of difference between, average birth weights among African and Portuguese babies in Portugal. *Int J Epidemiol.* (2006) 35:270–6. doi: 10.1093/ije/dyi225
23. IOM. *Migration Factsheet No. 6 – The Impact of COVID-19 on Migrants* (2020). Available online at: https://www.iom.int/sites/default/files/our_work/ICP/MPR/migration_factsheet_6_covid-19_and_migrants.pdf (accessed January 19, 2021).
24. World Health Organization (WHO). *Fact Sheet: Maternal Health* (2017). Available online at: http://www.euro.who.int/__data/assets/pdf_file/0006/354921/3.1-SDG-Fact-sheet-Maternal-Health.pdf (accessed February 14, 2018).
25. World Health Organization (WHO). *Sexual and Reproductive Health Fact Sheet 2019* (2019). Available online at: https://www.afro.who.int/publications/sexual-and-reproductive-health-fact-sheet (accessed April 5, 2021).
26. Urbanos-Garrido RM, Lopez-Valcarcel BG. The influence of the economic crisis on the association between unemployment and health: an empirical analysis for Spain. *Eur J Heal Econ.* (2015) 16:175–84. doi: 10.1007/s10198-014-0563-y
27. WHO. *The Minsk Declaration*. World Health Organization (2021). Available online at: https://www.euro.who.int/en/publications/policy-documents/the-minsk-declaration (accessed March 17, 2021).
28. Jones NL, Gilman SE, Cheng TL, Drury SS, Hill C V., Geronimus AT. Life course approaches to the causes of health disparities. *Am J Public Health.* (2019) 109:S48–55. doi: 10.2105/AJPH.2018.304738
29. World Health Organization (WHO). *Sexual Health and Its Linkages to Reproductive Health: An Operational Approach.* New York, NY: World Health Organization (2017). Available online at: http://www.who.int/reproductivehealth/publications/sexual_health/sh-linkages-rh/en/ (accessed April 1, 2021).
30. WHO Regional Office for Europe. *Health at Key Stages of Life – The Life-Course Approach to Public Health*. Copenhagen: World Health Organization (2017). Available online at: https://www.euro.who.int/en/health-topics/Life-stages/healthy-ageing/activities/health-at-key-stages-of-life-the-life-course-approach-to-public-health (accessed March 17, 2021).
31. Kuruvilla S, Sadana R, Montesinos EV, Beard J, Vasdeki JF, Carvalho IA De, et al. A life-course approach to health: synergy with sustainable development goals. *Bull World Health Organ.* (2018) 96:42–50. doi: 10.2471/BLT.17.198358
32. UNHCR. *UNHCR Policy on Age, Gender and Diversity* (2018). Available online at: https://www.unhcr.org/5aa13c0c7.pdf (accessed May 11, 2020).
33. Evans R, Forte C Lo, Mcaslan Fraser E. *UNHCR's Engagement with Displaced Youth* (2013). Available online at: www.unhcr.org (accessed May 11, 2020).
34. World Health Organization. *The Global Strategy and Action Plan on Ageing and Health 2016–2020: Towards a World in Which Everyone Can Live a Long and Healthy Life*. World Health Organization (2016). Available online at: https://apps.who.int/gb/ebwha/pdf_files/WHA69/A69_R3-en.pdf?ua=1 (accessed March 25, 2021).
35. Richter LM, Daelmans B, Lombardi J, Heymann J, Boo FL, Behrman JR, et al. Investing in the foundation of sustainable development: pathways to scale up for early childhood development. *Lancet.* (2017) 389:103–118. doi: 10.1016/S0140-6736(16)31698-1
36. Cherian Oommen J. *"To Be The Change" Rediscovering The Spirit of Service in Health Care in India.* Karamsad: Charutar Arogya Mandal (2011).
37. Carpenter LM. Gendered sexuality over the life course: a conceptual framework. *Sociol Perspect.* (2010) 53:155–77. doi: 10.1525/sop.2010.53.2.155
38. Castañeda H, Holmes SM, Madrigal DS, Young M-ED, Beyeler N, Quesada J. Immigration as a social determinant of health. *Annu Rev Public Health.* (2015) 36:375–92. doi: 10.1146/annurev-publhealth-032013-182419
39. Carpenter L, DeLamater J. *Sex for Life: From Virginity to Viagra, How Sexuality Changes Throughout Our Lives*. University Press Scholarship Online (2012).
40. López N. Contextualizing lived race-gender the racialized gendered social determinants of health. In: Gómez LE, López N, editors. *Mapping "Race": Critical Approaches to Health Disparities Research.* New Brunswick, NJ: Rutgers University Press (2013). p. 179–211.
41. Freitas Â, Santana P, Oliveira MD, Almendra R, Bana E Costa JC, Bana E Costa CA. Indicators for evaluating European population health: a Delphi selection process. *BMC Public Health.* (2018) 18:1–20. doi: 10.1186/s12889-018-5463-0
42. Beattie E, Mackway-Jones K. A Delphi study to identify performance indicators for emergeny medicine. *Emerg Med J.* (2004) 21:47–50. doi: 10.1136/emj.2003.001123
43. Coleman P, Nicholl J. Consensus methods to identify a set of potential performance indicators for systems of emergency and urgent care. *J Health Serv Res Policy.* (2010) 15:12–18. doi: 10.1258/jhsrp.2009.009096
44. Zeitlin J. PERISTAT: indicators for monitoring and evaluating perinatal health in Europe. *Eur J Public Health.* (2003) 13:29–37. doi: 10.1093/eurpub/13.suppl_1.29
45. Gagnon AJ, Zimbeck M, Zeitlin J. Migration and perinatal health surveillance: an international Delphi survey. *Eur J Obstet Gynecol Reprod Biol.* (2010) 149:37–43. doi: 10.1016/j.ejogrb.2009.12.002
46. Turner S, Ollerhead E, Cook A. Identifying research priorities for public health research to address health inequalities: use of Delphi-like survey methods. *Health Res Policy Syst.* (2017) 15:87. doi: 10.1186/s12961-017-0252-2
47. Wong ST, Browne AJ, Varcoe C, Lavoie J, Fridkin A, Smye V, et al. Development of health equity indicators in primary health care organizations using a modified Delphi. *PLoS ONE.* (2014) 9:e114563. doi: 10.1371/journal.pone.0114563
48. Landeta J. Current validity of the Delphi method in social sciences. *Technol Forecast Soc Change.* (2006) 73:467–82. doi: 10.1016/j.techfore.2005.09.002
49. Linstone H, Turoff M. *The Delphi Method: Techniques and Applications*. Linstone H, Turoff M, editors. Reading, MA: Addison-Wesley (1975).
50. Powell C. The Delphi technique: myths and realities. *J Adv Nurs.* (2003) 41:376–82. doi: 10.1046/j.1365-2648.2003.02537.x
51. Adler M, Ziglio E, editors. *Gazing Into the Oracle*. Bristol, PA: Kingsley (1996). Available online at: https://www.scirp.org/(S(351jmbntvnsjt1aadkposzje))/reference/ReferencesPapers.aspx?ReferenceID=591433 (accessed March 31, 2021).
52. Hung HL, Altschuld WJ, Lee YF. Methodological and conceptual issues confronting a cross-country Delphi study of educational program evaluation. *Eval Prog Plann.* (2008) 31:191–8. doi: 10.1016/j.evalprogplan.2008.02.005
53. World Health Organization (WHO), Department of Reproductive Health and Research. *Developing Sexual Health Programmes*. World Health Organization (2012). Available online at: http://www.who.int/reproductivehealth/publications/sexual_health/rhr_hrp_10_22/en/ (accessed April 1, 2021).
54. World Health Organization (WHO). *A Multi-country Study on Gender, Sexuality and Vaginal Practices: Implications for Sexual Health: Policy Brief* (2012).
55. World Health Organization (WHO*). Brief Sexuality-Related Communication*. WHO (2015). Available online at: http://www.who.int/reproductivehealth/publications/sexual_health/sexuality-related-communication/en/ (accessed April 1, 2021).

56. Reprostat. *Reproductive Health Indicators in the European Union Project Co-financed by the European Comission Health Monitoring Programme* (2001).
57. Estrela E. *Relatório Sobre Saúde e Direitos Sexuais e Reprodutivos* (2013). Available online at: https://www.europarl.europa.eu/doceo/document/A-7-2013-0306_PT.html (accessed April 1, 2021).
58. DIHR. *AAAQ & Sexual and Reproductive Health and Rights*. The Danish Institute for Human Rights (2017). Available online at: https://www.humanrights.dk/publications/aaaq-sexual-reproductive-health-rights (accessed April 1, 2021).
59. Ezzati M, Lopez AD, Rodgers A, Vander Hoorn S, Murray CJL. Selected major risk factors and global and regional burden of disease. *Lancet.* (2002) 360:1347–1360. doi: 10.1016/S0140-6736(02)11403-6
60. Fraser EDG, Dougill AJ, Mabee WE, Reed M, McAlpine P. Bottom up and top down: analysis of participatory processes for sustainability indicator identification as a pathway to community empowerment and sustainable environmental management. *J Environ Manage.* (2006) 78:114–27. doi: 10.1016/j.jenvman.2005.04.009
61. Temmerman M, Khosla R, Say L. Sexual and reproductive health and rights: a global development, health, and human rights priority. *Lancet.* (2014) 384:e30-1. doi: 10.1016/S0140-6736(14)61190-9
62. Wang CC, Wang Y, Zhang K, Fang J, Liu W, Luo S, et al. Reproductive health indicators for China's rural areas. *Soc Sci Med.* (2003) 57:217–25. doi: 10.1016/S0277-9536(02)00341-6
63. World Health Organization (WHO), UNFPA. *Measuring Sexual Health: Conceptual and Practical Considerations and Related Indicators.* World Health Organization (2010).
64. World Health Organization (WHO), Department of Reproductive Health and Research. *Social Determinants of Sexual and Reproductive Health*. World Health Organization (2010).
65. Fehr A, Lange C, Fuchs J, Neuhauser H, Schmitz R. Journal of health monitoring | 1/2017 | health monitoring in Europe. *Eur J Heal Monit J Heal Monit.* (2018) 2:3–21. doi: 10.17886/RKI-GBE-2017-020.2
66. Bretschneider S, Marc-Aurele FJ, Wu J. "Best practices" research: a methodological guide for the perplexed. *J Public Adm Res Theory.* (2005) 15:307–23. doi: 10.1093/jopart/mui017
67. Slocum N. *Participatory Methods Toolkit: A Practitioner's Manual*. S. Steyaert King Baudouin Foundation (2003).
68. Tan S, Brown J. The World Café in Singapore: creating a learning culture through dialogue. *J Appl Behav Sci.* (2005) 41:83–90. doi: 10.1177/0021886304272851
69. Stewart A. On conversation and collective questioning: theory and practice of the World Café. *Syst Think.* (2005) 16:9–10.
70. Anderson L. Use the World Café concept to create an interactive learning environment. *Educ Prim Care.* (2011) 22:337–38. doi: 10.1080/14739879.2011.11494028
71. Grabovac I, Smith L, Yang L, Soysal P, Veronese N, Isik AT, et al. The relationship between chronic diseases and number of sexual partners: an exploratory analysis. *BMJ Sex Reprod Heal.* (2020) 46:100–07. doi: 10.1136/bmjsrh-2019-200352
72. PORDATA. *Mean Age of the Mother at Birth of First Child.* (2019). Available online at: https://www.pordata.pt/en/Europe/Mean+age+of+the+mother+at+birth+of+first+child-3468 (accessed March 18, 2019).
73. Naderifar M, Goli H, Ghaljaie F. Snowball sampling: a purposeful method of sampling in qualitative research. *Strides Dev Med Educ.* (2017) 14:1–14. doi: 10.5812/sdme.67670
74. Alarcão V, Stefanovska-Petkovska M, Virgolino A, Santos O, Ribeiro S, Costa A, et al. Fertility, migration and acculturation (FEMINA): a research protocol for studying intersectional sexual and reproductive health inequalities. *Reprod Health.* (2019) 16:140. doi: 10.1186/s12978-019-0795-5
75. Delbecq AL, Van De Ven AH, Gustafson DH. *Group Techniques for Program Planning: A Guide to Nominal Group and Delphi Processes*. Foresman Glenview: Scott (1975). Available online at: https://www.researchgate.net/publication/40930404_Group_Techniques_for_Program_Planning_A_Guide_to_Nominal_Group_and_Delphi_Processes (accessed April 3, 2021).
76. Hugé J, Le Trinh H, Hai PH, Kuilman J, Hens L. Sustainability indicators for clean development mechanism projects in Vietnam. *Environ Dev Sustain.* (2010) 12:561–71. doi: 10.1007/s10668-009-9211-6
77. Deane KHO, Ellis-Hill C, Dekker K, Davies P, Clarke CE. A Delphi survey of best practice occupational therapy for parkinson's disease in the United Kingdom. *Br J Occup Ther.* (2003) 66:247–54. doi: 10.1177/030802260306600603
78. von der Gracht HA. Consensus measurement in Delphi studies. Review and implications for future quality assurance. *Technol Forecast Soc Change.* (2012) 79:1525–36. doi: 10.1016/j.techfore.2012.04.013
79. Santos O, Lopes E, Virgolino A, Stefanovska-Petkovska M, Dinis A, Ambrósio S, et al. Defining a brief intervention for the promotion of psychological well-being among unemployed individuals through expert consensus. *Front Psychiatry.* (2018) 9:13. doi: 10.3389/fpsyt.2018.00013
80. Shah HA, Kalaian SA. Which is the best parametric statistical method for analyzing Delphi data? *J Mod Appl Stat Methods.* (2009) 8:226–32. doi: 10.22237/jmasm/1241137140
81. English JM, Kernan GL. The prediction of air travel and aircraft technology to the year 2000 using the Delphi method. *Transp Res.* (1976) 10:1–8. doi: 10.1016/0041-1647(76)90094-0
82. Kalaian S, Kasim RM. Terminating sequential delphi survey data collection. *Pract Assess Res Eval.* (2012) 17:1–10.
83. Yang YN. Testing the stability of experts' opinions between successive rounds of Delphi studies. *Paper presented at the Paper Prepared for the Annual Meeting of the American Educational Research Association.* Chicago, IL (2003).
84. OECD. *Frascati Manual 2015: Guidelines for Collecting and Reporting Data on Research and Experimental Development, The Measurement of Scientific, Technological and Innovation Activities*. OECD (2015).
85. Alarcão V, Stefanovska-Petkovska M, Virgolino A, Santos O, Costa A. Intersections of immigration and sexual/reproductive health: an umbrella literature review with a focus on health equity. *Soc Sci.* (2021) 10:63. doi: 10.3390/socsci10020063
86. DGS/INSA. *Infeção VIH e SIDA em Portugal - 2019*. Lisboa (2019).
87. World Health Organization (WHO). *Global Health Sector Strategy on Sexually Transmitted Infections 2016–2021 Towards Ending STIs.* Geneva (2016).
88. Peixoto MM, Nobre P. Prevalence and sociodemographic predictors of sexual problems in Portugal: a population-based study with women aged 18 to 79 years. *J Sex Marital Ther.* (2015) 41:169–80. doi: 10.1080/0092623X.2013.842195
89. Platt L, Grenfell P, Fletcher A, Sorhaindo A, Jolley E, Rhodes T, et al. Systematic review examining differences in HIV, sexually transmitted infections and health-related harms between migrant and non-migrant female sex workers. *Sex Transm Infect.* (2013) 89:311–9. doi: 10.1136/sextrans-2012-050491
90. Dzomba A, Tomita A, Govender K, Tanser F. Effects of migration on risky sexual behavior and HIV acquisition in South Africa: a systematic review and meta-analysis, 2000–2017. *AIDS Behav.* (2019) 23:1396–430. doi: 10.1007/s10461-018-2367-z
91. Dias S, Gama A, Tavares AM, Reigado V, Simões D, Carreiras E, et al. Are opportunities being missed? Burden of HIV, STI and TB, and unawareness of HIV among african migrants. *Int J Environ Res Public Health.* (2019) 16:2710. doi: 10.3390/ijerph16152710
92. Kreps GL, Sparks L. Meeting the health literacy needs of immigrant populations. *Patient Educ Couns.* (2008) 71:328–32. doi: 10.1016/j.pec.2008.03.001
93. Park S, Lee H, Kang M. Factors affecting health literacy among immigrants - systematic review. *Eur J Public Health.* (2018) 28:cky214.283. doi: 10.1093/eurpub/cky214.283
94. Scheer JR, Pachankis JE, Bränström R. Gender-based structural stigma and intimate partner violence across 28 countries: a population-based study of women across sexual orientation, immigration status, and socioeconomic status. *J Interpers Viol.* (2020). doi: 10.1177/0886260520976212
95. APAV. *Estatísticas APAV, Relatório Anual 2019*. Lisboa (2020).
96. Oliveira C, Oliveira Martins MDR, Dias S, Keygnaert I. Conceptualizing sexual and gender-based violence in European asylum reception centers. *Arch Public Health.* (2019) 77:1–11. doi: 10.1186/s13690-019-0351-3
97. Macintyre S. The sociology of reproduction. *Sociol Heal Illness.* (1980) 2:215–22. doi: 10.1111/1467-9566.ep10487797

98. Oakley A. The sociology of childbirth: an autobiographical journey through four decades of research. *Sociol Heal Illness.* (2016) 38:689–705. doi: 10.1111/1467-9566.12400
99. Small R, Roth C, Raval M, Shafiei T, Korfker D, Heaman M, et al. Immigrant and non-immigrant women's experiences of maternity care: a systematic and comparative review of studies in five countries. *BMC Pregn Childbirth.* (2014) 14:152. doi: 10.1186/1471-2393-14-152
100. Quinta Gomes AL, Nobre P. Prevalence of sexual problems in Portugal: results of a population-based study using a stratified sample of men aged 18 to 70 years. *J Sex Res.* (2014) 51:13–21. doi: 10.1080/00224499.2012.744953
101. Pascoal PM, Narciso IDSB, Pereira NM. What is sexual satisfaction? Thematic analysis of lay people's definitions. *J Sex Res*. (2013) 51:22–30. doi: 10.1080/00224499.2013.815149
102. Pascoal PM, Shaughnessy K, Almeida MJ. A thematic analysis of a sample of partnered lesbian, gay, and bisexual people's concepts of sexual satisfaction. *Psychol Sex.* (2019) 10:101–18. doi: 10.1080/19419899.2018.1555185
103. Laumann EO, Paik A, Glasser DB, Kang JH, Wang T, Levinson B, et al. A cross-national study of subjective sexual well-being among older women and men: findings from the global study of sexual attitudes and behaviors. *Arch Sex Behav.* (2006) 35:145–61. doi: 10.1007/s10508-005-9005-3
104. Laumann EO, Nicolosi A, Glasser DB, Paik A, Gingell C, Moreira E, et al. Sexual problems among women and men aged 40-80 y: Prevalence and correlates identified in the Global Study of Sexual Attitudes and Behaviors. *Int J Impot Res.* (2005) 17:39–57. doi: 10.1038/sj.ijir.3901250
105. WHO. *Defining Sexual Health*. World Health Organization (2006).
106. Glasier A, Cameron ST, Blithe D, Scherrer B, Mathe H, Levy D, et al. Can we identify women at risk of pregnancy despite using emergency contraception? Data from randomized trials of ulipristal acetate and levonorgestrel. *Contraception.* (2011) 84:363–7. doi: 10.1016/j.contraception.2011.02.009
107. Baptista MIR. *O Aborto como Recurso na Regulação da fecundidade: Tendências Recentes em Portugal* (2017). Available online at: https://repositorio.iscte-iul.pt/bitstream/10071/17093/1/phd_maria_rodrigues_baptista.pdf
108. Vicente L. Aborto por opção da mulher: a experiência portuguesa da implementação da Rede Nacional. *Cad Saude Publica.* (2020) 36:e00036219. doi: 10.1590/0102-311x00036219
109. Seetharaman S, Yen S, Ammerman S. Improving adolescent knowledge of emergency contraception: challenges and solutions. *Open Access J Contracept.* (2016) 7:161–73. doi: 10.2147/OAJC.S97075
110. Almeida L, Caldas J, Ayres-De-Campos D, Salcedo-Barrientos D, Dias S. Maternal healthcare in migrants: a systematic review. *Matern Child Heal J.* (2013) 17:1346–54. doi: 10.1007/s10995-012-1149-x
111. Almeida L, Caldas J. Social determinants of health in pregnancy, postpartum and early motherhood : the impact of migration. *Meth Revis Cienc Soc.* (2014) 2:154–60. doi: 10.17502/m.rcs.v2i1.43
112. Endler M, Al Haidari T, Chowdhury S, Christilaw J, El Kak F, Galimberti D, et al. Sexual and reproductive health and rights of refugee and migrant women: gynecologists' and obstetricians' responsibilities. *Int J Gynecol Obstet.* (2020) 149:113–19. doi: 10.1002/ijgo.13111
113. Carvalho JLS, Santos A. Estudo Afrodite. *Caraterização da Infertilidade em Portugal I - Estudo na Comunidade*. Porto (2009).
114. World Health Organization (WHO). *Fact Sheets on Sustainable Development Goals: Sexual and Reproductive Health*. (2017). Available online at: http://www.euro.who.int/__data/assets/pdf_file/0005/348008/Fact-sheet-SDG-SRH-FINAL-04-09-2017.pdf (accessed December 5, 2018).
115. Sedgh G, Filippi V, Owolabi OO, Singh SD, Askew I, Bankole A, et al. Insights from an expert group meeting on the definition and measurement of unsafe abortion. *Int J Gynecol Obstet.* (2016) 134:104–6. doi: 10.1016/j.ijgo.2015.11.017
116. Bearak J, Popinchalk A, Ganatra B, Moller AB, Tunçalp Ö, Beavin C, et al. Unintended pregnancy and abortion by income, region, and the legal status of abortion: estimates from a comprehensive model for 1990–2019. *Lancet Glob Health.* (2020) 8:e1152–61. doi: 10.1016/S2214-109X(20)30315-6
117. Ventura T, Lima G, Gomes MC. *Relatório dos Registos das Interrupções da GravideZ.* Lisbon (2018). Available online at: https://www.dgs.pt/portal-da-estatistica-da-saude/diretorio-de-informacao/diretorio-de-informacao/por-serie-1144918-pdf.aspx?v=%3D%3DDwAAAB%2BLCAAAAAAABAArySzItzVUy81MsTU1MDAFAHzFEfkPAAAA (accessed January 19, 2021).
118. Gagnon AJ, Zimbeck M, Zeitlin J, Alexander S, Blondel B, Buitendijk S, et al. Migration to western industrialised countries and perinatal health: a systematic review. *Soc Sci Med.* (2009) 69:934–46. doi: 10.1016/j.socscimed.2009.06.027
119. Urquia ML, Glazier RH, Blondel B, Zeitlin J, Gissler M, Macfarlane A, et al. International migration and adverse birth outcomes: role of ethnicity, region of origin and destination. *J Epidemiol Community Health.* (2010) 64:243–51. doi: 10.1136/jech.2008.083535
120. Teixeira AL, Lisboa M. Estimating the prevalence of female genital mutilation in Portugal. *Public Health.* (2016) 139:53–60. doi: 10.1016/j.puhe.2016.05.002
121. Pascoal PM, Raposo CF, Roberto MS. A transdiagnostic approach to sexual distress and sexual pleasure: a preliminary mediation study with repetitive negative thinking. *Int J Environ Res Public Health.* (2020) 17:7864. doi: 10.3390/ijerph17217864
122. Gruskin S, Yadav V, Castellanos-Usigli A, Khizanishvili G, Kismödi E. Sexual health, sexual rights and sexual pleasure: meaningfully engaging the perfect triangle. *Sex Reprod Heal Matt.* (2019) 27:29–40. doi: 10.1080/26410397.2019.1593787
123. Ford JV, Corona Vargas E, Finotelli I, Fortenberry JD, Kismödi E, Philpott A, et al. Why pleasure matters: its global relevance for sexual health, sexual rights and wellbeing. *Int J Sex Health.* (2019) 31:217–30. doi: 10.1080/19317611.2019.1654587
124. Nunes MF, Leite AH, Dias SF. Inequalities in adherence to cervical cancer screening in Portugal. *Eur J Cancer Prev.* (2020) 30:171–7. doi: 10.1097/CEJ.0000000000000612
125. Marques P, Nunes M, Antunes MDL, Heleno B, Dias S. Factors associated with cervical cancer screening participation among migrant women in Europe: a scoping review. *Int J Equity Health.* (2020) 19:160. doi: 10.1186/s12939-020-01275-4
126. Dobrotić I, Stropnik N. Gender equality and parenting-related leaves in 21 former socialist countries. *Int J Sociol Soc Policy.* (2020) 40:495–514. doi: 10.1108/IJSSP-04-2019-0065
127. Andersen SH. Paternity leave and the motherhood penalty: new causal evidence. *J Marr Fam.* (2018) 80:1125–43. doi: 10.1111/jomf.12507
128. OECD. *Parental Leave: Where Are the Fathers?* Policy Brief (2016).
129. Wall K, Correia RB, Leitão M. Portugal country note. In: Koslowski A, Dobrotić I, Kaufman G, Moss P, editors. *International Review of Leave Policies Research 2020.* Available online at: http://www.leavenetwork.org/lp_and_r_reports/ (accessed January 19, 2021).
130. Kil T, Wood J, Neels K. Parental leave uptake among migrant and native mothers: Can precarious employment trajectories account for the difference? *Ethnicities.* (2018) 18:106–41. doi: 10.1177/1468796817715292
131. Mussino E, Duvander A-Z, Ma L, Talaga L. Recours au congé parental chez les immigrés pères d'un premier enfant en Suède : la durée passée dans le pays compte-t-elle? *Population.* (2018) 73:381–401. doi: 10.3917/popu.1802.0381
132. ILGA. *State-Sponsored Homophobia Report - 2020 Global Legislation Overview Update*. ILGA (2020). Available online at: https://ilga.org/state-sponsored-homophobia-report-2020-global-legislation-overview (accessed January 19, 2021).
133. Portugal ILGA. Homofobia e Transfobia: Dados da Discriminação em Portugal 2017. (2017). Available online at: https://ilga-portugal.pt/ficheiros/pdfs/observatorio/Relatorio-Discriminacao-2017_17maio2018.pdf
134. UNAIDS. *The Gap Report* (2014). Available online at: https://www.unaids.org/en/resources/documents/2014/20140716_UNAIDS_gap_report (accessed December 14, 2020).
135. Ledoux C, Pilot E, Diaz E, Krafft T. Migrants' access to healthcare services within the European Union: a content analysis of policy documents in Ireland, Portugal and Spain. *Global Health.* (2018) 14:57. doi: 10.1186/s12992-018-0373-6
136. Presidência do Conselho de Ministros. *Portaria 203/2016, 2016-07-25 - DRE* (2016). Available online at: https://data.dre.pt/web/guest/pesquisa/-/search/75025066/details/maximized (accessed January 19, 2021).

137. De Leeuw ED, Hox J, Huisman M. Prevention and treatment of item nonresponse. *J Off Stat*. (2003) 19:153–76.
138. Oliveira Da Silva M. Reflections on the legalisation of abortion in Portugal. *Eur J Contracept Reprod Heal Care*. (2009) 14:245–48. doi: 10.1080/13625180903053740
139. Lazdane G, Rakvac I. *How to Monitor Sexual and Reproductive Health*? (2015). Available online at: http://www.europeristat.com/ (accessed January 19, 2021).
140. Ogbe E, Van Braeckel D, Temmerman M, Larsson EC, Keygnaert I, De Los Reyes Aragón W, et al. Opportunities for linking research to policy: lessons learned from implementation research in sexual and reproductive health within the ANSER network. *Heal Res Policy Syst*. (2018) 16:123. doi: 10.1186/s12961-018-0397-7
141. World Health Organization (WHO). *Guidelines for Their Generation, Interpretation and Analysis for Global Monitoring* (2006).

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.

Copyright © 2021 Candeias, Alarcão, Stefanovska-Petkovska, Santos, Virgolino, Pintassilgo, Pascoal, Costa and Machado. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

frontiers in Public Health

ORIGINAL RESEARCH
published: 14 October 2021
doi: 10.3389/fpubh.2021.616742

The Barriers and Enablers of Primary Healthcare Service Transition From Government to Community Control in Yarrabah: A Grounded Theory Study

Janya McCalman[1], Crystal Sky Jongen[1], Sandy Campbell[1,2], Ruth Fagan[3], Kingsley Pearson[3] and Suzanne Andrews[3]*

[1] Centre for Indigenous Health Equity Research, Central Queensland University, Cairns, QLD, Australia, [2] Molly Wardaguga Research Centre, Charles Darwin University, Darwin, NT, Australia, [3] Gurriny Yealamucka Health Service, Yarrabah, QLD, Australia

OPEN ACCESS

Edited by:
Alexandra P. Leader,
Eastern Virginia Medical School,
United States

Reviewed by:
Brita A. Pekarsky,
Flinders University, Australia
Josée Lavoie,
University of Manitoba, Canada

***Correspondence:**
Janya McCalman
j.mccalman@cqu.edu.au

Specialty section:
This article was submitted to
Public Health Policy,
a section of the journal
Frontiers in Public Health

Received: *13 October 2020*
Accepted: *10 September 2021*
Published: *14 October 2021*

Citation:
McCalman J, Jongen CS, Campbell S, Fagan R, Pearson K and Andrews S (2021) The Barriers and Enablers of Primary Healthcare Service Transition From Government to Community Control in Yarrabah: A Grounded Theory Study. Front. Public Health 9:616742. doi: 10.3389/fpubh.2021.616742

Introduction: Consistent with the aspirations of First Nations Australians for community control of healthcare services, 123/196 (63%) of Australia's First Nations-specific primary health care services are community-controlled. Yet despite policy commitment over 30 years, the transition of government-run First Nations' primary healthcare services to First Nations community control has been slow. This paper identifies the barriers and enablers to transitioning the delivery of primary healthcare services from Queensland Health to Gurriny Yealamucka community-controlled health service in Yarrabah.

Methods: Grounded theory methods were used to select 14 Gurriny and Queensland Health (QH) personnel involved in the transition for interview and to analyse these interview transcripts and 88 Gurriny organisational documents.

Results: Barriers and enablers to transition were identified at three levels: those internal factors within Gurriny, external factors directly related to the government handover, and broader structural and policy factors outside the control of either Gurriny or QH. Barriers at the Gurriny organisational level were an internal lack of experience and capacity, and varying levels of community confidence; enablers were leadership stability and capacity, community mandate, relationships with partner organisations, and ability to provide service continuity. Barriers in Gurriny's relationship with QH were a lack of certainty, transparency and prioritisation of the transition process; systemic racism; difficulties obtaining and maintaining the necessary workforce; limited resources including insufficient, unstable and inappropriate funding support; and problems with information sharing; enablers were performance frameworks to keep transition progress on track. Barriers in broad policy environment were an unsupportive Queensland government policy environment; government bureaucracy; and delays, conflicts and divisions; enablers were high-level government support and commitment.

Conclusions: The evaluation of Yarrabah's transition process suggests that future such transitions will require planning and commitment to a long-term, multi-faceted and complex process, encompassing the required level of authorisation and resourcing. This case example of a transition from government to community control of PHC highlighted the ongoing power issues that are faced every day by community-controlled organisations that co-exist with mainstream health systems within a colonial power structure.

Keywords: community control, self-determination, governance, transition, Indigenous

INTRODUCTION

First Nations peoples globally value their right to "retain their Indigenous values and traditions, ways of life and their languages and cultures, and to do so in a contemporary context" [(1), p. 156]. They have sought this autonomy despite government "policies of dispossession, marginalisation, assimilation and integration," and related experiences of discrimination, prejudice and indifference [(2), p. 10]. As clearly cited in the Uluru Statement from the Heart by Aboriginal and Torres Strait Islander (hereafter respectfully termed First Nations) Australians, "Our Aboriginal and Torres Strait Islander tribes were the first sovereign Nations of the Australian continent … and possessed it under our own laws and customs.… This sovereignty is a spiritual notion.… With substantive … structural reform, we believe this ancient sovereignty can shine through" [(3), p. iv]. Only with self-determination will First Nations peoples be able to fully overcome the legacy of Australia's colonisation and dispossession including current disadvantage (4, 5).

Self-determination (or community control) of an organisation is achieved when it attains real power to make decisions through community boards and management, such as how to utilise resources (6). The first Aboriginal Community-Controlled Healthcare Organisation (ACCHO) was established in 1971 in the context of ongoing resistance by First Nations peoples to widespread systemic racism, ongoing processes of colonisation and dispossession (6–8), and a dearth of government support and funding (9, 10). In Australia, 123/196 (63%) of First Nations primary healthcare services are currently community controlled, with the remaining 63 (32%) being government-run and 12 (6%) non-government operated (11).

Most of the current ACCHOs were established as community controlled from the start; a minority were transitioned from previously government-run services. Transition requires the devolution of power and authority by the state or territory government over First Nations' core institutions, goals and identity, as well as strengthening of the capacity of a First Nations community controlled organisation to renegotiate bureaucratic, legal and policy arrangements with the state (2). This research paper examines the barriers and enablers encountered throughout one attempt to attain self-determination through the first transition of a government-run primary healthcare (PHC) service to First Nations community control in Queensland. The healthcare service transition was negotiated in Australia's largest discrete First Nations community and one of its most disadvantaged—Yarrabah.

BACKGROUND

As well as through the 196 First Nations PHCs across Australia, PHC services are available to First Nations Australians through mainstream services such as the Commonwealth government subsidised, privately owned general practise PHCs, or state funded and provided hospitals (12). However, barriers have been documented relating to the accessibility, affordability, cultural acceptability and appropriateness of mainstream PHC to First Nations peoples' health needs (12–15). For example, a recent systematic review found that "Aboriginal people fare worse than non-Aboriginal people when accessing usual (mainstream) healthcare services" [(16), p. 314]; with mainstream health services and standard, non-tailored care not being responsive to community health needs (17). In Queensland, a report by the Anti-Discrimination Commission and Aboriginal and Islander Health Council found that government-run mainstream hospital and healthcare services were "not taking [their] responsibilities to Close the Indigenous Health Gap seriously" [(18), p. 14], and identified the structural conditions for institutional (or systemic) racism. Systemic racism occurs when in-built discrimination "systematically reflect[s] and produce[s] racial inequalities…" [(19), p. 438]. Such barriers result in later presentation at PHC services and at hospitals with more advanced and complex health issues than those of non-Aboriginal Australians, thereby contributing to an increased burden of disease and reduced quality of life (13, 20).

The ACCHO sector provides an important expression of the principle of self determination as "a proven mechanism for Aboriginal people to take responsibility over their own health matters" (6, 9, 21). ACCHOs are incorporated organisations initiated and governed by First Nations community members. They deliver holistic and culturally appropriate health services to the community (22). They are funded by both state and Commonwealth governments, using multiple funding models. ACCHOs address many of the healthcare access barriers because services and programs are grounded in local values and culture (23), adopt the First Nations concept of holistic health that encompasses social, political and cultural determinants

---

Abbreviations: PHC, Primary healthcare; ACCHO, Aboriginal Community Controlled Healthcare Service; Gurriny, Gurriny Yealamucka Health Service; QH, Queensland Health; CIHER, Centre for Indigenous Health Equity Research; CQU, Central Queensland University; CHHHS, Cairns and Hinterland Hospital and Health Service; QAIHC, Queensland Aboriginal and Islander Health Council; DOGIT, Deed of Grant in Trust; SEIFA, Socio-Economic Indexes for Areas; NATSIHA, Northern Aboriginal and Torres Strait Islander Health Agency; NACCHO, National Aboriginal Community Controlled Health Organisation.

of health (8, 9, 21), and are accountable to the interests, needs, values, vision and concerns of community members (2, 21, 23). They address affordability barriers by providing free primary healthcare, and accessibility barriers by providing transport, outreach and childcare support services (23). Non-Indigenous people are also cared for in these clinics, but First Nations people represent 82% of all clients (11). Through their cultural-centredness, and comprehensive and flexible approach to primary healthcare, ACCHOs are similar to Indigenous health services internationally (24).

The transition of state or territory run PHC services to First Nations community control is a complex process (25). For the past 30 years, Commonwealth and state governments in Australia have provided a funding and policy commitment to community control (26). For example, the National Aboriginal and Torres Strait Islander Health Plan 2013–2023 promotes a "robust, strong, vibrant and effective community controlled health sector" in which "individuals and community actively engage in decision making and control" [(27), p. 7]. Since the mid-late 90's this trend has included the support of the Queensland and Northern Territory health authorities for transferring PHC services delivered in First Nations communities to community control (25, 26). But despite policy commitment and significant investments in health reforms, there have been few successful transitions (28). For example, Northern Territory reforms to promote community control over PHC governance and service delivery produced the transfer of only one clinic during the period 2011–14, and no further proposals being accepted by the government (8).

Past evaluations of documented examples, particularly in regional and remote Northern Territory, Queensland and South Australian communities, have ascertained a range of enablers to successful transitions. These included a recognition by governments that their dominant governance arrangements required institutional change, including: the presence of niche alternative practises within government departments that provide a template for change; effective authorisation and sustained commitment through a continuity of leadership from ministers and senior government officials; and explicit measures to address systemic racism (8). As well, transitions required adequate time, funding and capacity (5, 26, 28). However, many more evaluations document the barriers to transition efforts (28). For example, barriers to transitioning the regional ACCHOs, Miwatj Health Aboriginal Corporation in the Northern Territory and Apunipima Cape York Health Council in Queensland included: poor coordination and role clarity between state/territory and ACCHO providers and between funding agencies and ACCHOs; short-term funding contracts; challenges associated with regionalising governance; accountability for effective care, access and responsiveness to communities; two-way accountability with funders; and a need for increased funding to cover rural/remote costs and improve needs-based equity (16). These barriers limited the success of transition to community controlled governance of only one each of their several regional clinics (the Yirrkala and Mossman clinics, respectively) (28). Elucidating these barriers and enablers allows government and community stakeholders to streamline processes and avoid the repetition of costly and damaging practises that hinder such efforts (29).

To that end, this paper presents the results of the recent evaluation of the successful transition to community control of PHC in Yarrabah from Queensland Health to Gurriny Yealamucka Health Service (hereafter Gurriny) (30). It documents the enablers and barriers to the transition process so that other communities aspiring to transition, and government partners wanting to support them, can improve future transitions. The research question was: What were the enablers that supported the transition of the delivery of PHC services to First Nations community control in Yarrabah, and what were the barriers to this transition?

METHODS

Research Approach

We applied the Indigenous research and data sovereignty principles of ownership, control, access and possession (OCAP®) within the research (31). The research was contracted to Gurriny by Queensland Health (QH). "Ownership" was enacted through Gurriny control of the research funding, governance and research partnership with Central Queensland University (CQU)'s Centre for Indigenous Health Equity Research (CIHER) through a Research Services Agreement, and their oversight throughout. "Control" was asserted through a steering committee established to guide the research, that was chaired and coordinated by Gurriny, and included representatives from the Aboriginal and Torres Strait Islander branch of QH, Cairns and Hinterland Hospital and Health Service (CHHHS), Queensland Aboriginal and Islander Health Council (QAIHC), Gurriny, and the CIHER research team. Seven of the nine members were First Nations people. Data from participant interviews were secured on a CQUniversity data management server but "access" to the aggregated findings was provided by CIHER researchers to Gurriny staff through plain English reports and presentations of the findings. "Possession" was enabled through Gurriny ownership of the final report and co-authorship of this paper (31). Further details of the approach and methods are provided in a companion paper on the processes and strategies of transition (25).

The Provision of Primary Healthcare to the Yarrabah Community

Yarrabah is a discrete First Nations community in Far North Queensland, 52 km south east of Cairns (**Figure 1**). In 1892, an Anglican Mission was founded on the traditional lands of the Gunggandji people, and subsequent state government policies resulted in the forcible relocation of First Nations and some South Sea Islander peoples to Yarrabah. The community is now self-governing under a Deed of Grant in Trust (DOGIT) land tenure status.

Yarrabah is now Australia's largest First Nations community. The 2016 census records the community as home to 2,559 First Nations' residents (33); however, Gurriny's regular client list of 3,600 in 2016 suggests that this is a significant undercount. Yarrabah was ranked amongst Australia's local government areas

FIGURE 1 | Yarrabah. Source: Bentleys (32).

with the most extreme concentration of social and economic disadvantage (34). Associated with this disadvantage, Yarrabah experiences a high burden of chronic disease.

Until 2014, primary healthcare and emergency hospital services were provided in Yarrabah by the Queensland government-run Yarrabah Primary Health Care Centre. The centre was operated by one of Queensland Health (QH)'s Cairns and Hinterland Hospital and Health Service (CHHHS) with healthcare services offered by medical, nursing and health worker staff, and visiting community and allied health providers (35).

The first stage of the transition journey to establishing a community-controlled health organisation in Yarrabah was triggered by community dissatisfaction with the healthcare services provided by CHHHS in Yarrabah (see **Figure 2**). This led the Yarrabah Aboriginal Council to form a health committee in 1989, which was incorporated in 1991. A feasibility study in 1997 led to a renaming as the Yarrabah Health Council, and again in 2000 as Gurriny Yealamucka Health Service. The second stage (2005–2014) entailed preparing for transition to full community control of PHC in Yarrabah, with commitment to this end articulated through a Deed of Commitment between Gurriny and Commonwealth, Queensland and local government partners (2005) to achieve better health outcomes for Yarrabah. The four partners committed to implementing community control over the planning, prioritisation and management of PHC service delivery to the community of Yarrabah, and affirmed the essential requirements of community control as: (1) community identification of needs, aspirations and priorities; (2) a representative organisation based on good governance and best practise; and (3) a baseline document (Health Strategic Plan) for resource allocation (25). When the Deed of Commitment was signed in 2005, transition partners agreed upon the transition date of 2008, or 2010 at the latest.

In June 2014, the 28-year process was completed when Yarrabah became the first community in Queensland to transition PHC services from Queensland Health to First Nations' community control. Like other ACCHOs, Gurriny was funded through administratively complex funding arrangements through the Commonwealth government's Indigenous Australian's Health Program, primary health networks and Medical Benefits Schedule, QH program and grant funding, and other sundry funding such as research grants and philanthropic funds (36). The processes undertaken in these two stages are described more fully in Jongen et al. (25).

Data Collection and Analysis

Interviews were held with people involved in the transition of PHC to community control in Yarrabah. A purposive sampling technique was initially used, with information-rich participants identified by senior managers at Gurriny and QH. These and self-identified participants were invited to participate in interviews that focused on their experiences of the transition, including enablers and barriers. A broad interview schedule (provided as a **Supplementary Material**) guided the interviews. As data collection progressed, theoretical sampling processes were used to identify further potential participants with diverse perspectives and ability to explore issues that had emerged from the initial data analysis.

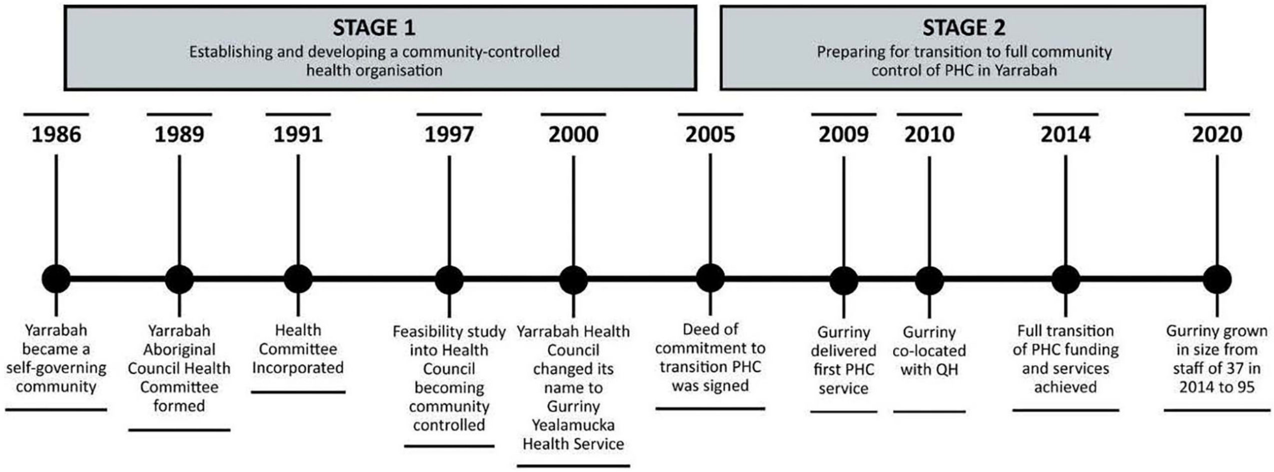


FIGURE 2 | Stages of transition.

Fourteen people were interviewed, eight of whom were First Nations' people. They included current Gurriny staff members (6, 3 of whom had previously worked for QH), ex-Gurriny staff members (2), current QH staff members (1), ex-QH staff members (3 in addition to those who were re-employed at Gurriny), other Yarrabah community members (1) and one other (1). With participant consent and at a place of their choice, face-to-face or telephone interviews were undertaken by SC and JM; interviews were audio-recorded and transcribed [for further details, see (25)]. Transcripts were given back for checking to those participants who requested them.

Eighty-eight historical organisational documents, dated from 2005, were provided by Gurriny as a point-in-time record of the transition, with augmentation of data from interviews of the retrospective viewpoints of those involved. The most common types of documents were progress or status reports ($n = 19$, 22%), published or internal reports ($n = 11$, 13%), plans ($n = 10$, 12%) and communication briefs ($n = 9$, 10%).

Data Analysis

The interview transcripts and Gurriny organisational documents were analysed using grounded theory methods. As described in Jongen et al. (25), the transcripts and documents were imported into NVIVO qualitative software and analysed using the constant comparison methods of grounded theory. Open-coding was conducted iteratively upon receipt of the transcripts and documents to identify actions and interactions (37). Codes that were associated in meaning were then grouped under higher order categories (38). These were integrated to determine the context, strategies implemented, and the barriers to and enablers of implementation (38). The strategies are described in Jongen et al. (25).

RESULTS

The enablers and barriers to transition are the factors that supported and/or hindered Gurriny's journey towards achieving community control of PHC during the two stages of transition. Three types of enablers and barriers were identified: internal factors within Gurriny's control, external factors directly related to the CHHHS handover which occurred largely outside of the control of Gurriny, and broader structural and policy factors outside the control of either Gurriny or CHHHS.

Internal barriers were Gurriny's lack of experience and capacity, and varying levels of community confidence. Internal Gurriny enablers were its leadership stability and capacity, community mandate, relationships with partner organisations, and ability to provide service continuity. External barriers were a lack of certainty, transparency and prioritisation of the transition process by CHHHS; systemic racism; difficulties obtaining and maintaining the necessary workforce; limited resources including insufficient, unstable or inappropriate funding support; and problems with information sharing. External enablers were CHHHS performance frameworks to keep transition progress on track. Broad structural barriers included an unsupportive Queensland government policy environment; government bureaucracy; and delays, conflicts and divisions. Broad structural enablers were high-level Commonwealth and QH government support and commitment, and funding (**Table 1**).

Internal Gurriny Barriers

The two internal barriers were Gurriny's lack of experience and capacity and a lack of confidence by some community members in Gurriny's capacity to run an effective PHC service.

Gurriny's Lack of Experience and Capacity

By 2005, when the Deed of Commitment was signed by Gurriny, the Yarrabah Aboriginal Shire Council, QH and the Commonwealth Department of Health, Gurriny was still a small organisation, employing only 10 staff members. At the time, Gurriny Board members and some key senior managers had limited experience in health and some lacked financial expertise. There was a perception that the burden of transition was borne

TABLE 1 | Key barriers and enablers of the transition to community control in Yarrabah.

Level	Barrier	Enabler
Internal Gurriny factors	Gurriny's lack of experience and capacity Lack of community confidence	Gurriny's leadership stability and capacity Community mandate Relationships with partner organisations Ability to provide service continuity
Relationships with CHHHS	A lack of certainty, transparency and prioritisation of the transition process by QH Systemic racism Difficulties obtaining and maintaining the necessary workforce Limited resources including insufficient, unstable or inappropriate funding support Problems with information sharing	Performance frameworks to keep transition progress on track
Broader structural and policy environments	An unsupportive Queensland government policy environment Unresponsive government bureaucracy Delays, conflicts and divisions	High-level Commonwealth and QH government support and commitment Funding

by Gurriny which had little funding, power or experience to enact the expected tasks. A Gurriny staff member noted:

> *"the problem that we had all the way through, was that we were just a small organisation and we didn't have the capacity to just churn out all these things that Queensland Health were expecting us to churn out. And they were trying to measure us on our ability to provide that documentation or provide that evidence."*

Lack of Community Confidence

Early in the transition process, some community members were concerned that the transition would incur a potential reduction in service availability and quality. Some local community members also expressed a lack of confidence in Gurriny's capacity to be in control of Yarrabah's health care. Some CHHHS staff were also community members, and they felt that they were already delivering quality services and achieving results, and that Yarrabah did not need community control. Community desire for community control was taken personally as it was related to direct criticisms of the work of CHHHS. A former-CHHHS/current Gurriny staff member reflected:

> *"I could not see community control working. I was like, '… but we have everything. Why are we changing?"'*

Internal Gurriny Enablers

The four internal enablers were Gurriny's: leadership stability and capacity, community mandate, relationships with partner organisations, and ability to provide service continuity.

Gurriny's Leadership Stability and Capacity

Gurriny's leadership was strong, stable and determined through the lengthy transition process. Gurriny had a reasonably stable Board throughout stage two of the transition years, which meant that experience and knowledge was retained. A Gurriny manager said:

> *"I think that was really important having that consistent leadership at the Board level."*

Senior managers and Board Directors played critical leadership roles in the oversight, guidance, planning and negotiating of transition processes with government, and built organisational capacity over a long timeframe. For example, all Board members partook in capacity building opportunities and an ex-officio Board member was recruited to bring financial expertise. Gurriny also had a dedicated Transition Manager, who was funded by the Commonwealth and responsible for transition coordination and program monitoring and reporting. Despite significant challenges, Gurriny leaders and staff demonstrated the leadership qualities of perseverance and determination to the extent that they were willing to do whatever was necessary to make the transition happen. Another Gurriny manager said:

> *"All the way along, we did just keep chugging along, making the organisation better and smarter.... We use[d] the deadline like with the 2010 Deed of Commitment. We all tried to use those deadlines to hold people to account, but we never thought that once we got to that deadline we'd just give up."*

Community Mandate

The transition of PHC in Yarrabah to community control was driven by the dissatisfaction of community members about existing CHHHS healthcare service provision. A former Gurriny employee said:

> "*there was quite a lot of people in Yarrabah… we had these great big ideas to develop community control because people weren't happy with the current services that was going on there.*"

Relationships With Partner Organisations

From the first stage of the transition journey, the implementation of evidence-informed programs and services was facilitated through research collaborations; researchers also evaluated their effects. The evaluations demonstrated to community and government stakeholders that Gurriny had built capacity, thereby enhancing confidence and trust, and helping to secure funding for workforce and leadership capacity development, employment of staff, and further expansion of programs and services.

Several of Gurriny's key alliances during the second stage of transition were within the ACCHO sector. The Queensland Aboriginal and Islander Health Council (QAIHC), the peak body for the Queensland state ACCHO, mentored Gurriny through high level strategic negotiations with state and Commonwealth governments and provided consultancy expertise. Gurriny was also able to share organisational knowledge, experience and resources with Apunipima Cape York Health Council (located in nearby Cairns) related to their simultaneous transition processes. They collaborated to develop community and research engagement strategies and shared in contracting various consultants to complete required planning and assessment tasks. Gurriny also established strategic partnerships through membership with the Northern Aboriginal and Torres Strait Islander Health Agency (NATSIHA) and National Aboriginal Community Controlled Health Organisation (NACCHO) to leverage knowledge and support to progress the transition. A Gurriny manager said:

> *"we leaned a lot on expertise that might've came our way from QAIHC and from Apunipima – just to tap into other work that they'd already done, or talking to different people. And that was kinda how we got through it."*

Finally, partnerships and alliances with consultancy services provided strategic, legal and operational guidance to progress the transition. For example, an Organisational Capacity Review, developed by consultancy firm Bentley's, identified core strategy areas requiring improvement as: workforce planning and development, the service delivery model, information technology, finance/funding modelling, and legal issues and governance (32). A Gurriny manager recalled:

> *"Bentley's came in and done a twelve-month review on Gurriny. From the Board right down to service delivery, to IT and workforce, all of that... And out of that were some really good stuff because there were some things that we did lack. And I thought, 'wow, I didn't realise that."'*

The Bentley's Organisational Capacity Review helped to create clarity and certainty concerning government expectations and requirements, was incorporated within Gurriny's plans, strategies and actions to progress transition, and enabled Gurriny to take appropriate action.

Ability to Provide Service Continuity

Planning processes to develop a health services plan and delivery model were focused most intensively in the years 2006–2008. In 2007, external consultants provided a Proposed Service Delivery Framework for Gurriny, including recommendations about services that should be included, and the integration of clinical services with previously established social and emotional well-being programs. Gurriny and CHHHS also developed a Yarrabah Health Services Plan (2008) based on the assessment and mapping of Yarrabah's health service needs and options; this became a guiding document for service provision. Later in the transition process, health program planning occurred annually. Despite workforce supply challenges, Gurriny achieved its staffing requirements and was able to provide continuous services during transition.

External Barriers in the Relationship With CHHHS

The five barriers that were beyond the control of Gurriny but were apparent in its relationship with CHHHS were a lack of certainty, transparency and prioritisation in the transition process; systemic racism; difficulties obtaining and maintaining the necessary workforce; limited resources including insufficient, unstable or inappropriate funding support; and problems with information sharing.

Lack of Certainty, Transparency, and Prioritisation of the Transition Process

Being the first Queensland transition of PHC from government to community control, there was a general lack of clarity and understanding amongst all involved parties about the process, and a lack of expert knowledge and guidance or frameworks to guide the transition. Leaders within CHHHS did not have the relevant expertise, experience or resources to oversee the process, and many stakeholders were not aware of which legislation and government policies and procedure could affect transition. Frequent changes in these laws, policies and procedures meant that policy was developed as the transition occurred. A former CHHHS manager said:

> *"we had this unprecedented industrial arrangement where we then had to question how staff would transition from one service to another… it was the first time it had happened, so policy was kind of being developed as it was happening."*

A Gurriny manager also reflected:

> *"I don't know even if the politicians even understood that they might have some legislation or some policies that are actually gonna stop or impact on what they're saying they want done."*

Furthermore, the transition process comprised one small component amongst other competing priorities of the CHHHS portfolio, which resulted in its low prioritisation. There was no dedicated leadership within CHHHS to oversee and progress the transition. A former CHHHS manager recalled:

> *"…the first failing. This is a multi-million-dollar procurement over a significant period of time. And in any other procurement of this size, you would actually have allocated a person managing that... So it was one of those things that got managed when it came up. When there was a need for it to come up, it came up and the rest of the time, to be honest, it wasn't something that we had somebody who made it their full-time priority."*

Systemic Racism

Systemic racism manifested from the start of the transition process in the form of resistance, negative reactions and a lack of support from some CHHHS staff, and through the inherent power imbalance of the two organisations and risk-averse processes of CHHHS. A former Gurriny manager said:

> *"originally when we finished the Feasibility Study report [1998] and we gave a copy to the state government, and the state government services was upset. So all the [QH] nursing staff was really upset and they sort of rebelled. The government itself didn't accept the Feasibility Study report."*

This resistance and reluctance to relinquish control continued throughout the transition. Presenting as a catch-22 situation, it was based in the (not unfounded) concern about Gurriny's relative lack of experience and capacity to manage the complex operations of the large PHC service to provide quality care to ameliorate the considerable burden of disease in the community. A former CHHHS manager also noted:

> *"you had many people playing the politics of 'this is community driven and led.' Like I agree in the principle, but if you're going to give it to people that actually understand health and have some skills and knowledge I think. 'Cause there's risks behind that if you don't.'"*

From 2009, this reluctance to let go of control contributed to a shift in commitment from community control to an explicit focus on service integration through co-location of Gurriny and CHHHS. Gurriny struggled to secure CHHHS commitment to a plan and timeline for full transition. A Gurriny manager noted:

> *"I did feel a little bit that Queensland Health... weren't that willing to let go."*

The marked power imbalance that existed between QH/CHHHS and Gurriny was also seen as a barrier to an equitable partnership between the two organisations and a significant source of systemic racism. A former CHHHS manager perceived:

> *"We held the power in this relationship. There's no questioning that."*

The inequality in power was evident, for example, in the risk-averse service Operating Deed (2016) which set out the legal relationship between the two services. The Deed and Lease agreements, which were written to protect QH's reputation and funding, demonstrated a lack of trust by CHHHS in Gurriny's capacity to take control of PHC services. The Operating Deed was described by a Gurriny staff member as "*risk-averse, protective, hand-holding, unilateral, paternalistic and overbearing.*" For example, despite only 20% of the services being funded by QH (the balance being funded through Commonwealth grants and Medicare), the Operating Deed required Gurriny to account to CHHHS with data and reports for every aspect of service delivery. A former manager from Gurriny shared this perspective:

> *"The Deed of Operations... was incredibly one-sided, judgmental and demanding from the Queensland Health side. And absolutely, when you considered they were providing less than twenty per cent of our funds, they were wanting all the data set, all of the knowledge... when in fact, when you look at the amount of money that was coming into Yarrabah at that time for the Health Services, Gurriny got a tiny drop in the ocean of that."*

Through these documents and other indications, participants inferred an implicit message on the part of CHHHS that they expected Gurriny to fail, and that CHHHS would need to reassert control. In parallel, the willingness of QH/CHHHS staff to support Gurriny in building the required knowledge and capacity varied. A Gurriny manager said:

> *"the hidden message underneath that was, 'we're gonna keep tentacles involved in this because they're probably gonna fall over and we'll have to step back in.'"*

As a result, Gurriny levels of reciprocal trust in the goodwill of CHHHS fell. For example, a Joint Working Group was reported as difficult to progress due to the "*risk... that QH will railroad (Gurriny's) work according to their needs.*" Gurriny documents also reported that a joint planning, monitoring and reporting framework was put on hold because required support from CHHHS was not provided; the Transition Risk Management Plan couldn't progress due to lack of cooperation from CHHHS; a Transition Implementation Plan that was supposed to be jointly developed faltered due to lack of CHHHS involvement; and a review and design framework to support joint accreditation was difficult to progress in collaboration with CHHHS.

Difficulties Obtaining and Maintaining the Necessary Workforce

The transition of CHHHS staff across to Gurriny entailed an unprecedented industrial dilemma and major challenge in the transition process. As public servants, CHHHS staff experienced better employment conditions than most of the private sector workforce, and some CHHHS staff were concerned about the potential that they might lose their jobs, accrued benefits and leave entitlements. Differences in organisational cultures and values, models of care, and staff award wage and entitlement systems between government and non-government systems meant that not all positions in CHHHS were to transition to equivalent roles. Also, a strong resistance from CHHHS staff contributed to their unwillingness to work for Gurriny.

At the 11th h and without consultation with Gurriny, CHHHS/QH decided to offer redundancies to their Yarrabah staff members. However, the conditions of the redundancy offer meant that those who accepted would need to wait for 3 months before they could apply for available positions at Gurriny. Gurriny responded by temporarily employing people for that 3 month period to enable CHHHS staff to apply, which impacted on their capacity for service continuity and achievement of a smooth transition. A Gurriny manager recalled:

> *"it was a real pain but we worked out if they were made redundant... they had to not work for three months... so what*

> *we could do is, we would only put on staff for three months to fill positions, to keep the wheels chugging along and then we would advertise the permanent positions and if a Queensland Health staff was interested in applying, they were welcome to apply. There was no guarantee they'd get the job but we would hold off on recruiting permanent positions until they were eligible to apply. Which is what we ended up doing."*

Although Gurriny managers supported the transition of CHHHS staff through engagement and providing the opportunity to apply for positions, ultimately, only two former CHHHS staff out of Gurriny's staff complement at the time of 37 transitioned across to Gurriny.

Limited Resources Including Insufficient, Unstable or Inappropriate Funding Support

There were three funding phases relevant to the transition: (1) the pre-transition operating costs, which were borne by CHHHS; (2) the costs of the transition process itself; and (3) the ongoing operating costs post-transition which were borne by Gurriny. The method for calculating the funding to be transferred was a key barrier to the smooth transition of PHC responsibilities in Yarrabah.

There was a lack of clarity by CHHHS pre-transition about the what the costs of delivering PHC services to Yarrabah entailed. Related to this, the type of services and amount of funding that would be transitioned from CHHHS to Gurriny were unclear throughout much stage two of the transition process. There was no assessment to inform the funding decisions of community needs and demand for services, the actual services Gurriny would deliver, the cost of Gurriny's model of care, or potential service delivery improvements.

Transition costs included the costs of infrastructure, accreditation, recruitment, systems, developing pathways and models of care. Limitations in the availability of resources to progress organisational development, and uncertain or unstable funding hindered Gurriny's workforce and organisational growth throughout the process. For example, Gurriny was required by Commonwealth and state governments to complete various planning processes that necessitated the engagement of external consultants, and frequently no additional funding was allocated for these efforts.

The costing method used by CHHHS to determine the funding they would transfer upon final transition was driven by their (non-stated) vested interest in managing a cost neutral transition of PHC services (i.e., they would continue to contribute the same funds as they had previously expended - regardless of actual cost of service delivery or growth). The budget was based on actual expenditure which was lower than the operational budget (presumably due to unfilled positions, and the delivery of less service provision than that planned/budgeted for). Furthermore, payment was to be provided after service provision. Funding was coming directly from the CHHHS budget and they had competing needs and priorities. This funding approach was not anticipated by Gurriny and was considered by participants from both services to be inappropriate. A Gurriny manager said:

> *"If someone could come back and say, 'well actually… you are delivering more care than was anticipated when we gave you this small amount of money. This is actually what it costs, and this is what you should be funded for to deliver that care…' because Queensland Health said, 'well these are the positions: four nurses and a number of Health Workers. You will deliver this, this and this.' But we're doing triple that amount of work on whatever that budget is."*

A former CHHHS manager explained:

> *"There was a very strong drive from the (hospital and health service) … that we weren't going to give Gurriny any more money than we actually would save by not providing that service… and we commissioned that audit…. So I suspect that a conservative approach may have been taken."*

Furthermore, the final funding amount was decided almost immediately prior to the official handover. This meant that Gurriny was compelled to plan service delivery without any clarity about the available level of funding for those services. A former CHHHS manager said:

> *"Gurriny didn't know how much money they were getting, they didn't know what services they could offer, so they couldn't have positions in place, ready to fill, to go into a transparent recruitment process."*

After the official handover of funding and services in June 2014, funding issues continued to plague Gurriny's capacity to provide healthcare. For the first 3 months, CHHHS did not pay the allocated funding to Gurriny or respond to the invoices sent. This meant Gurriny did not have the required funding for 11 positions. Furthermore, once funding commenced, CHHHS paid at the end rather than the beginning of the month. These issues created a significant financial burden and compromised the solvency of Gurriny in the first year following transition. A Gurriny manager said:

> *"Queensland Health … didn't pay their first monthly remittance for those positions until the September of that year. So Gurriny was almost pushed to bankruptcy because they had once again, [acted in] good faith and employed people, but the funds weren't there because Queensland Health didn't pay."*

Problems With Information Sharing

Issues related to the sharing of client information between Gurriny and CHHHS were a significant and persistent barrier to successful service collaboration over many years. For example, the CHHHS CEO agreed to share medical records while the services were co-located. A Gurriny staff member recalled:

> *"it was constant head-butting right up til twenty thirteen when… (the) CEO of Cairns Hospital at the time said, 'enough is enough. We cannot go on with these separate records. We must have one record because we continue to compromise patient care.'"*

However, following this decision, the Nurses Union advised CHHHS nurses to only use paper records. To this day, Gurriny's client information system is shared with CHHHS emergency department doctors in Yarrabah, but CHHHS emergency department nurses do not share their client data.

Enablers in the Relationship With CHHHS

The two external enablers in the relationship with CHHHS were funding and performance frameworks to keep transition progress on track.

Performance Frameworks to Keep Transition Progress on Track

A package of performance frameworks was prepared by Queensland Health and used throughout the transition process to keep Gurriny and CHHHS on track in key action areas of the transition. These included a Strategic Policy Framework for Transition, a Readiness Assessment Framework, Industrial Relations Guidelines, Information Management Guidelines, Joint Communication and Engagement Guidelines, Evaluation Guidelines, and Funding Guidelines. The performance frameworks helped Gurriny to assess its implementation of core strategies across all areas of its operation and to demonstrate organisational capacity to operate a complex PHC service. This was a requirement of government stakeholders and necessary for building trust and securing ongoing support for transition.

Broad Structural Barriers

The three broad structural barriers were an unsupportive policy environment; government bureaucracy; and delays, conflicts and divisions.

Unsupportive Policy Environment

The lack of dedicated CHHHS leadership and resources (discussed above) largely resulted from the difficult funding and policy environment that shaped the QH organisational context, capacity and priorities at the time of transition. The transition process occurred during a conservative state government term. Funding cuts compelled QH to reduce staff contingents and CHHHS was under considerable pressure to allocate all resources to frontline service delivery. It was very unlikely in this environment that it would have been achievable to acquire a dedicated position to manage the transition. A former CHHHS manager noted:

> *"I don't… in retrospect think that we would actually have been given approval to have somebody dedicated to work on this. It was a very difficult time to get administrative staff employed because of the philosophy of the Newman government and the caps that it had set on employment. And the head-count reduction… it was trying to achieve."*

Unresponsive Government Bureaucracy

A lack of capacity for reflexive, innovative and creative responses in government bureaucracy stalled and complicated decision making, hampering effective collaboration between Gurriny and CHHHS. Decisions went to and fro between sub-committees and lawyers to the extent that those involved could no longer make sense of the process. This unresponsive bureaucratic system not only disempowered Gurriny, but also diminished the decision-making ability of managers and leaders in CHHHS to seek and implement creative solutions. A Gurriny manager reflected:

> *"I think sometimes when people get into middle management or upper level management in bureaucracies… they just can't make decisions, so they deflect that decision across to a sub-committee that'll look at it for six to twelve months and it drifts into the ether and gets lost in translation."*

There was a disconnect between implementation on the ground and the support and policy directives coming from top levels of government. Another Gurriny manager reflected:

> *"One of the barriers was that it seemed at like the really high levels of government… they seemed to support this idea but when you got down to the bureaucrats who were supposed to do it… they then didn't know how to do it."*

Delays, Conflicts, and Divisions

Delays in the transition process were noteworthy and a significant hindrance. Prior to setting an official handover date in 2014, delays and setbacks in the transition process were so frequent that many participants considered that it was not getting anywhere. The transition was conditional on Gurriny's completion of ever-shifting deliverables, and was made even more difficult by limited resources and cooperation. The sheer quantity of work the transition required [see (25)] also contributed to the delays. A former CHHHS manager said:

> *"The transition for Yarrabah just had been coming for a very long time and it just dragged on and it dragged on and it dragged on, and it got to a point that many staff believed it would never happen."*

Conflicts and divisions experienced between Gurriny and CHHHS were particularly evident during the years of co-location (2010-14) when the relationships between Gurriny and CHHHS staff were fractured. There was a distinct separation between the two organisations, with no apparent collaboration, despite being co-located in the same building. A Gurriny manager reflected:

> *"I think what had happened was that when we co-located back in 2010… they tried to merge teams and they had two sets of Line Managers and you know, it was just really unpleasant. It was actually creating some little fires and there was lots of assumptions and toxic kind of team dynamics and things going on here all the time."*

Broad Structural Enablers

The two key structural enablers identified were funding and the support and commitment to transition at high levels of government.

Funding

State and Commonwealth government funding commitments were essential to transition. By 2005, when the Deed of Commitment was signed, the receipt of sustained Commonwealth government funding for the first Transition Officer position and four permanent social and emotional well-being positions was a defining moment for Gurriny, helping to create stability and sustainability in the organisation. A community research partner said:

> *"by 2005, the Commonwealth Office of Aboriginal Health actually came to the party and offered the first four permanent positions for Gurriny."*

High-Level Government Support and Commitment

Many of the enablers of the transition were at least partly a result of the broader state and Commonwealth government structural and policy systems. The support and good will of high-level QH and the Commonwealth Department of Health and Ageing bureaucrats towards the transition, evidenced by their signing of the Deed of Commitment, was identified as an enabler. Many high-level CEO to CEO meetings, committee meetings, Transition Steering Committee meetings, and clinical leadership meetings were held between Gurriny and CHHHS to support the transition and address operational issues. These pertained to issues regarding information systems, the signing of a memorandum of understanding between doctors, the use of a consent form, and the sharing of medical record systems. Despite protracted delays in dealing with many issues, key individual leaders within QH/CHHHS demonstrated courage in resolving them through taking charge of situations that had reached an impasse.

In 2014, an 11th-h Ministerial directive for setting and publicly declaring a clear transition date clinched the commitment, investment of resources, and accountability from government stakeholders, and resulted in rapid progress in the transition process. A Gurriny manager said:

> *"It was … the public declaration of a date that actually spurred everybody into action."*

Once transition had been achieved, Gurriny leaders implemented systems to enable delivery of a more comprehensive primary healthcare service to the people of Yarrabah.

DISCUSSION

This paper sought to identify the barriers that hindered and enablers that supported the transition of PHC services to First Nations' community control through Gurriny in Yarrabah. The experiences of Gurriny demonstrate that, as in other PHC transitions in other parts of Queensland and the Northern Territory, there are a range of factors which can both hinder and support the process (28, 39). In Yarrabah, these occurred at three levels: factors within Gurriny itself, those that were directly related to the handover from CHHHS, and broader structural and policy factors outside the control of Gurriny or CHHHS. The enablers of transition, extrapolated from the case of Yarrabah's transition, are depicted in **Figure 3**.

Internal Gurriny organisational barriers and enablers played a role in the transition. The mandate from the community to pursue self-determination regarding their own healthcare and other services drove the transition to community control, although work was required by Gurriny to strengthen community confidence. Strong organisational leadership and good governance were critical enablers of community control, with ongoing capacity development undertaken at the community, organisational and clinical levels (40). The dedication and perseverance of Gurriny leaders in efforts to achieve the vision of community control also contributed to the achievement of the 2014 transition. Partnerships and collaborations with research, ACCHO and other organisations also strengthened the capacity of Gurriny (39).

Most of the barriers to transition were directly related to the state government handover of control from CHHHS to Gurriny. As in examples of other transition processes, whilst the act of transitioning to community control was based on Commonwealth and state governments' commitment to decolonising health service provision, key among the challenges was the imbalanced power relationship and the reluctance of CHHHS to relinquish control (8). This reluctance to cede control to First Nations organisations is founded on a history of conflict and resentment between government health departments and ACCHOs (41)—and comprises an example of systemic racism [(28), p. 58]. In Yarrabah, as for other transitions, there were perceptions that CHHHS did not acknowledge the cultural legitimacy of Gurriny, imposing unreasonably high levels of accountability, micro management and reporting [(28), p. 58].

The transition encompassed a challenging cross-cultural process between two very different organisations with different values and priorities (28). CHHHS had concerns about Gurriny's capacity and, as in other transitions of PHC to community control, Gurriny did considerable work to address government doubts (28). Some government stakeholders perceived the transition to the ACCHO as an implicit criticism of existing services, especially relating to access and appropriateness (28). Factors such as a lack of trust arising from the different interests of partners across government and community sectors, the cross-cultural nature of relationships and a perception of underlying racism influenced different organisational perceptions of priorities, meaning, values, interests, goals, choices, expectations and timelines. There was a "mutual perception of failure to maintain commitment and a sense of significant pressure on established relationships and mutual trust" (28).

In part, the CHHHS resistance to community control came from the concerns of Yarrabah-based QH staff about the practicalities of transitioning their employment arrangements to Gurriny. For many QH staff, there was a sense of ownership over the clinic and lack of comfort about working for Gurriny. As in other transitions, government staff concerns included fears of losing job security, wages, and salary-related benefits, and possible fears about marginalisation and changes in accountability relationships (28). Industrial relations issues around transferring existing employees were complex. The

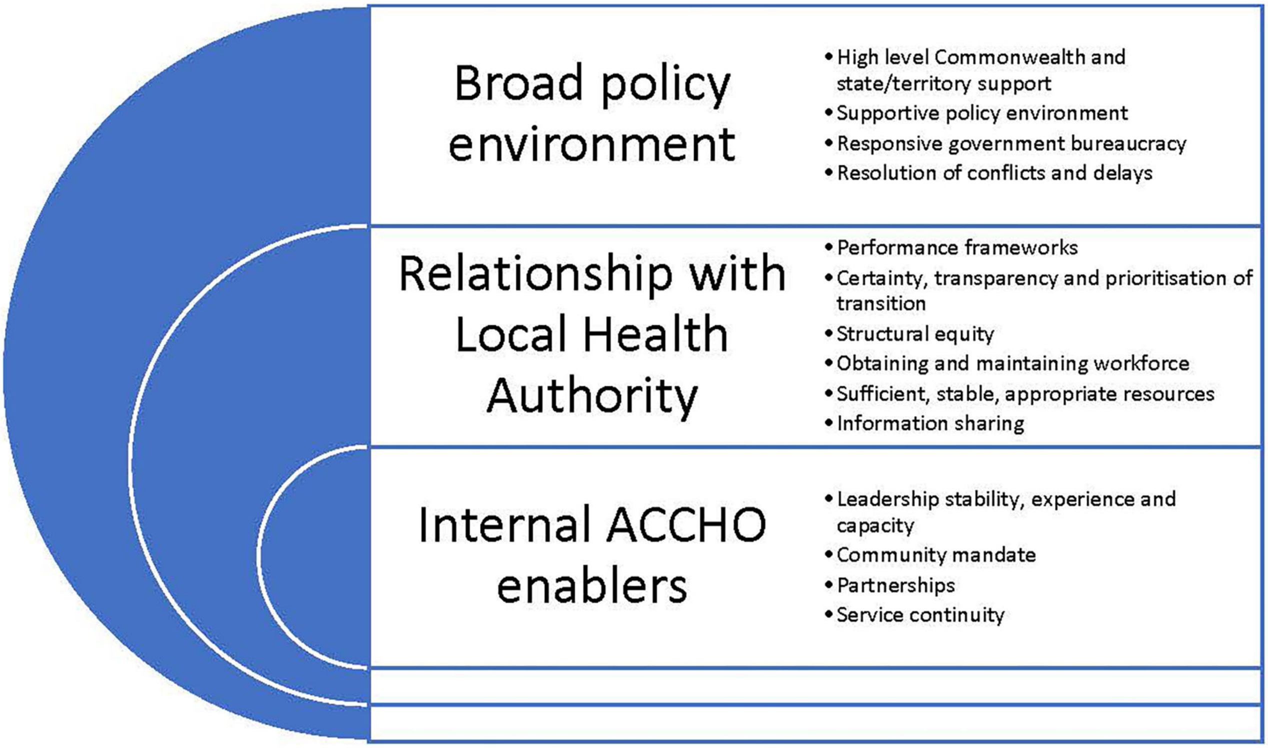


FIGURE 3 | The enablers of transition.

perceptions of staff in other PHC services experiencing transition were bluntly articulated thus: "the opposition of local non-Indigenous government staff was deeply rooted in disbelief in the capability of Aboriginal people and a distrust of Aboriginal organisations: 'I'll say blunt on record… that they don't want to work for a black organisation"' (28).

Inadequate resources, inflexible funding arrangements and the absence of clear agreements about funding amounts, conditions or timeframes were key barriers to a smooth transition of PHC responsibilities in Yarrabah (28, 40). Due to the poorer health status of the Yarrabah population, it was reasonable to expect greater (at least double the average per capita) government expenditure than for non-First Nations Australians in order to achieve equity of access (17). However, the 11th-h funding package was based neither on current or projected health need, demand for services, or equity (13). Instead, the funding allocation at transition was based on historical expenditure and designed to be cost neutral for CHHHS. Furthermore, while funds pooling was an explicit intention of transition, there was no substantial work from government towards achieving those aims. It became apparent that if transition was to make a significant difference to health outcomes, additional investment from the Commonwealth and Queensland governments would be required.

A package of performance management framework documents was developed by Queensland Health to assess the competencies and capabilities of Gurriny's governing board and its organisational readiness, and was applied as a condition of approval of transfer. Similar to other PHC transitions, standards were high, with Gurriny staff noting that CHHHS services were not similarly scrutinised (28). As in other transitions, it was "perceived by some in the ACCHO sector to be an extension of a generally excessive risk intolerance displayed by both levels of government" and represented a deficit approach to community control—a complicated process intended to mask the real underlying thought that "blackfellas can't run these things" (28). This type of risk intolerance in funding Aboriginal organisations meets the definition of systemic racism, as it has a differential application and impact in First Nations communities and organisations (8).

Despite continuing government policy commitments to community control, there was nevertheless "no enduring basis for accountability by governments for … transferring PHC provision to community-controlled healthcare providers" (26). Whilst Commonwealth funding support was a critical early enabler for the transition, there was insufficient recognition of the need for adequate resourcing of the change process itself [(28), p. 6]. Broad changes of national and state elected governments, restructuring of QH and reallocation of health service delivery responsibility to regional Hospital and Health Services in 2011-2012, led to frequent shifts in government priorities, policies and personnel, and high-level government prioritisation of transition was withdrawn as the transition progressed. The focus was shifted from community control to "service integration" (40).

As for PHC transition processes in other communities, the broader structural enablers in Yarrabah included a strong early commitment, policy support, and goodwill from governments towards the vision of community control (8, 28).

There has been policy commitment to community control in Australia for more than 30 years (8). But transitions of PHC to community control in Yarrabah and other communities have been "difficult and complex, and progress has been slow" (28). Despite consistent policy support, there was a failure at the outset to negotiate and secure the required level of authorisation and commitment to transitioning PHC services to community control (7), and an apparent reluctance of government health administrators to engage effectively with and transfer control to Aboriginal communities (8). Implementing community control takes time, and sufficient support and resources are required to navigate the process successfully (25, 26). Efforts to transfer services to community control have generally been conducted under tight timelines, which are inappropriate for the needed long-term commitment (26). The slowness of progress in the case of Yarrabah created a perception that there was limited government commitment to expected timelines and processes. As in other PHC transitions, these challenges manifested in delays, divisions and conflicts and a lack of certainty and clarity in the process and created opportunities for divisiveness and blame allocation.

Queensland Health now has a reform agenda to embed health equity across the health system and address the legacy of systemic racism described in this case of PHC transition. For the first time, a legislative requirement was passed by the Queensland Parliament in August 2020 which embeds a requirement for each Hospital and Health Service (HHS) to redesign and reorient local health systems to better listen to and support First Nations Queenslanders, address historical and ongoing economic and social injustices, and recognise First Nations sovereignty and right to self-determination (42). As part of this agenda, a relocation of authority and control from governments back to Indigenous organisations is needed to provide governance of First Nations peoples' right to self-determination that originates from their inalienable connexions to lands, waters and the natural world (31). The preference of First Nations people to access ACCHOs over mainstream PHC services supports this agenda (43, 44). For example, one study of patient access to one urban and five regional Queensland ACCHOs found that First Nations people preferred using the ACCHOs over mainstream PHC services (43). Furthermore, ACCHOs in Queensland have achieved very high access rates, with 60–100% of First Nations people who live close to ACCHOs accessing their services (44). Such control over essential services is recognised as an underlying social determinant of health and a health intervention in its own right (45, 46).

LIMITATIONS

This research is based on the perspectives of 14 participants who retrospectively recalled the enablers and barriers 4 years after the actual transition occurred, and the analysis of 88 point-in time documents. Although participants were selected based on their roles in the transition and/or unique perspectives, efforts to interview QH staff were met with limited success. Of the 12 current or former QH staff invited to interview, only 3 accepted. In contrast, of the 12 current or former Gurriny staff members invited, 8 accepted. Similarly, our analysis of historical organisational documents was based on documents provided by Gurriny. Similar documents from Queensland Health could not be assessed because acquiring access to Queensland Health documents required additional ethical approval which was not possible within the time limits of the research.

CONCLUSIONS

The implementation of community control in Australia requires commitment at three levels: by the local community organisation, in the relationship with the government health authority, and at the broader Commonwealth and state structural and policy level. The transition of PHC to community control in Yarrabah took 28 years. It was complicated by the ACCHO's lack of experience and capacity, wavering community confidence; the local government authority's lack of certainty, transparency and prioritisation of the transition process; systemic racism; difficulties obtaining and maintaining the necessary workforce; limited resources including insufficient, unstable or inappropriate funding support; problems with information sharing; and the broad structural and policy barriers of an unsupportive policy environment; government bureaucracy, delays, conflicts and divisions. Enablers were community-controlled leadership stability and capacity, community mandate, relationships with partner organisations, ability to provide service continuity, CHHHS performance frameworks to keep the transition process on track, and Commonwealth and Queensland government funding and high level support and commitment. This case example of a transition from government to community control of PHC highlighted the ongoing power issues that are faced every day by community-controlled organisations that co-exist with mainstream health systems within a colonial power structure.

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.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Ethics Approval was received from Far North Queensland Human Research Ethics Committee (HREC/18/QCH/95 HREC/18 Project ID: 41295). The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

JM contributed significantly to the conception and design of the research project and wrote the paper. CJ was responsible for completing the grounded theory analysis of organisational documents and participant interviews, and for writing the first draught of the results. SC was the project manager and completed the ethics applications. CJ, SC, and JM all contributed to the development of interview questions and SC and JM conducted interviews with participants. RF, KP, and SA were all members of the reference committee, helped to source documents and recruit participants, provided guidance on the evaluation, and provided feedback on the final manuscript. All authors read and approved the final manuscript.

FUNDING

The evaluation of transition to community control in Yarrabah was funded by Queensland Health. QH was represented on the evaluation advisory committee, but played no role in the design of the study or collection, analysis and interpretation of data or in the writing of the manuscript.

ACKNOWLEDGEMENTS

The authors acknowledge all participants for their willingness and time to be interviewed and the Reference Committee for guiding this project. Reference Committee members included: Venessa Curnow (Director, Aboriginal and Torres Strait Islander Health Unit, CHHHS, Queensland Health), Vanda Simpson (A/Manager, Aboriginal and Torres Strait Islander Health Branch, Strategy, Policy and Planning Division, QH), Gregory Richards (Aboriginal and Torres Strait Islander Health Branch, QH), Sue Andrews (CEO, Gurriny), Ruth Fagan (Business Manager, Gurriny), Karen Dennien (Director of Operations, Gurriny), Kingsley Pearson (Senior Medical Officer, Gurriny), Chris Doran (Health Economist, CQU), Sandra Campbell (Research Project Manager, CQU), Janya McCalman (Senior Research Fellow, CQU). We also acknowledge Professor Roxanne Bainbridge from the Gungarri/Kunja nations and Professorial Research Fellow at Central Queensland University for her involvement and guidance in the early development of the evaluation. We also wish to acknowledge Ros Calder for editing the manuscript.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2021.616742/full#supplementary-material

REFERENCES

1. Hawkes DC. Indigenous peoples: self-government and intergovernmental relations. *Int Soc Sci J.* (2001) 53:304. doi: 10.1111/1468-2451.00304
2. Behrendt L, Jorgensen M, Vivian A. *Self-determination: Background Concepts Scoping paper 1*. Melbourne, VIC: Victorian Department of Health & Human Services (2016).
3. Mayor T. *Finding the Heart of the Nation: The Journey of the Uluru Statement towards Voice, Treaty and Truth* (2019) Richmond, VIC: Hardie Grant Books.
4. Calma T, Chaney F. *'Never as Simple as Black and White: Turning Good Intention into Results' National Press Club (NPC) Canberra* (2007). Commissioner Calma's speech is Available online at: http://www.humanrights.gov.au/about/media/speeches/social_justice/2007/pressclub2007.html. Fred Chaney's speech is available at: http://www.reconciliation.org.au/downloads/156/ChaneyPressClub4July(smalltype)2007.pdf (accessed July 4, 2007).
5. Sherwood J, Edwards T. Decolonisation: a critical step for improving Aboriginal health. *Contemp Nurse.* (2006) 22:178–90. doi: 10.5172/conu.2006.22.2.178
6. Bartlett B, Boffa J. Aboriginal community controlled comprehensive primary health care: the Central Australian Aboriginal Congress. *Aust J Primary Health.* (2001) 7:74–82. doi: 10.1071/PY0105039
7. Durey A, Thompson SC, Wood M. Time to bring down the twin towers in poor Aboriginal hospital care: addressing institutional racism and misunderstandings in communication. *Intern Med J.* (2012) 42:17–22. doi: 10.1111/j.1445-5994.2011.02628.x
8. Dwyer J, Tenbensel T, Lavoie J, Martini A, Brown C, Devitt J, et al. Public administration reform for aboriginal affairs: an institutionalist analysis. *Aust J Public Admin.* (2020) 79:550–66. doi: 10.1111/1467-8500.12422
9. Rosewarne C, Vaarzon-Morel P, Bell S, Carter E, Liddle M, Liddle J. The historical context of developing an aboriginal community-controlled health service: a social history of the first ten years of the central australian aboriginal congress. *Health History.* (2007) 9:114–43. doi: 10.2307/40111578
10. Weightman M. The role of Aboriginal community controlled health services in Indigenous health. *Aust Med Stud J.* (2013) 4. Available online at: http://www.amsj.org/archives/3012
11. Australian Institute of Health and Welfare. *Online Services Report*. Canberra, ACT: Australian Government (2021).
12. Davy C, Harfield S, McArthur A, Munn Z, Brown A. Access to primary health care services for Indigenous peoples: a framework synthesis. *Int J Equity Health.* (2016) 15:163. doi: 10.1186/s12939-016-0450-5
13. Alford K. *Economic Value of Aboriginal Community Controlled Health Services: Executive Summary* (2014). NACCHOP Club. Available online at: https://nacchocommunique.files.wordpress.com/2014/04/naccho_14_econimic-value-exec-summary_020414.pdf (accessed September 27, 2021).
14. Bainbridge R, McCalman J, Clifford A, Tsey K. *Cultural Competency in the Delivery of Health Services for Indigenous People*. Canberra, ACT: Closing the Gap Clearinghouse; Australian Institute of Health and Welfare and Australian Institute of Family Studies (2015).
15. Clifford A, McCalman J, Bainbridge R, Tsey K. Interventions to improve cultural competency in health care for Indigenous peoples of Australia, New Zealand, Canada and the USA: a systematic review. *Int J Qual Health Care.* (2015) 27:89–98. doi: 10.1093/intqhc/mzv010
16. Gwynne K, Jeffries T, Lincoln M. Improving the efficacy of healthcare services for Aboriginal Australians. *Aust Health Rev Public Aust Hosp Assoc.* (2018) 43:314–22. doi: 10.1071/AH17142
17. Shannon C, Longbottom H. *Capacity Development in Aboriginal and Torres Strait Islander Health Service Delivery: Case Studies*. Canberra, ACT: Department of Health and Ageing (2004).
18. Marrie A. *Addressing Institutional Barriers to Health Equity for Aboriginal and Torres Strait Islander people in Queensland's Public Hospital and Health Services*. Cairns, QLD: Anti-Discrimination Commission, Queensland (2017).
19. Jones JM, Carter RT. Racism and white racial identity: merging realities. In: Bowser BP, Hunt RG, editors. *Impacts of racism on White Americans*. 2nd ed. Thousand Oaks, CA: Sage Publications, Inc. (1996). p. 1–23.

20. Department of the Prime Minister and Cabinet. *Closing the Gap Report 2019* (2019). Available online at: https://ctgreport.niaa.gov.au/ (accessed September 27, 2021).
21. National Aboriginal Community Controlled Health Organisation (NACCHO). NACCHO *10 Point Plan 2013–2030*. Canberra, ACT: NACCHO (2013).
22. National Aboriginal Community Controlled Health Organisation (2011). Available online at: https://www.naccho.org.au/acchos (accessed September 27, 2021).
23. Campbell MA, Hunt J, Scrimgeour DJ, Davey M, Jones V. Contribution of aboriginal community-controlled health services to improving aboriginal health: an evidence review. *Aust Health Rev.* (2018) 42:218–26. doi: 10.1071/AH16149
24. Pearson O, Schwartzkopff K, Dawson A, Hagger C, Karagi A, Davy C, et al. Aboriginal community controlled health organisations address health equity through action on the social determinants of health of Aboriginal and Torres Strait Islander peoples in Australia. *BMC Public Health.* (2020) 20:1859. doi: 10.1186/s12889-020-09943-4
25. Jongen C, Campbell S, McCalman J, Fagan R, Pearson K, Andrews S. Transitioning to aboriginal community control of primary health care: the process and strategies of one community-controlled health organisation in Queensland. *BMC Family Pract.* (2020) 21:230. doi: 10.1186/s12875-020-01300-z
26. Lavoie JG, Dwyer J. Implementing Indigenous community control in health care: lessons from Canada. *Aust Health Rev.* (2016) 40:453. doi: 10.1071/AH14101
27. Commonwealth of Australia. *National Aboriginal and Torres Strait Islander Health Plan 2013-2023*. Canberra: Commonwealth of Australia (2013).
28. Dwyer J, Martini A, Brown C, Tilton E, Devitt J, Myott P, et al. *The Road Is Made by Walking: Towards a better primary health care system for Australia's First Peoples: Summary Report. T. L. Institute* (2015). Available online at: https://www.lowitja.org.au/page/services/resources/health-policy-and-systems/health-policy/The-road-is-made-by-walking-Report (accessed September 27, 2021).
29. Tsey K, McCalman J, Bainbridge R, Brown C. *Improving Indigenous Community Governance Through Strengthening Indigenous and Government Organisational Capacity*. Canberra, ACT: Closing the gap Clearing House (2012).
30. McCalman J, Jones G. *Gurriny Yealamucka Health Service Evaluation Report 2014–15*. Cairns, QLD: James Cook University (2015).
31. Kukutai T, Taylor J. Data sovereignty for Indigenous peoples: current practice and future needs. In: TKaJ Taylor, editor. *Indigenous Data Sovereignty: Toward an Agenda*. Australia National University (2016). p. 1–24.
32. Bentleys. *Gurriny Yealamucka Health Service: Organisational Capacity Review 2014*. Brisbane, QLD: Bentleys Queensland (2014).
33. Australian Bureau of Statistics. 2016 *Census Quickstats*. Yarrabah (2018). Available online at: https://quickstats.censusdata.abs.gov.au/census_services/getproduct/census/2016/quickstat/SSC33236 (accessed September 27, 2021).
34. Australian Bureau of Statistics. *Australian Aboriginal and Torres Strait Islander Health Survey: First Results, Australia, 2012-13* (2014). Available online at: http://www.abs.gov.au/ausstats/abs@.nsf/Lookup/4727.0.55.001Chapter1002012-13 (accessed September 27, 2021).
35. Queensland Government. *Queensland Health Organisational Structure*. Queensland Government (2021). Available online at https://www.health.qld.gov.au/system-governance/health-system/managing/org-structure (accessed September 27, 2021).
36. The Centre of Research Excellence in Aboriginal Chronic Disease Knowledge Translation and Exchange (CREATE). *Aboriginal Community Controlled Health Organisations in practice: Sharing ways of working from the ACCHO sector* (2020). Available online at: https://create.sahmri.org/wp-content/uploads/2020/02/funding.pdf (accessed July 22, 2021).
37. Charmaz K. *Constructing Grounded Theory: A Practical Guide Through Qualitative Analysis*. Thousand Oaks, CA: SAGE (2014).
38. Strauss AL, Corbin JM. *Basics of Qualitative Research: Techniques and Procedures for Developing Grounded Theory*. 2nd ed. Thousand Oaks, CA: Sage Publications (1998).
39. McCalman J, Tsey K, Reilly L, Connolly B, Fagan R, Earles W, et al. Taking control of health: gurriny's story of organisational change. *Third Sector Rev.* (2010) 16:29–49. doi: 10.3316/IELAPA.201008108
40. Queensland Health. *Transition to Aboriginal and Torres Strait Islander Community Control of Health in Queensland: A Draft Strategic Policy Framework*. Brisbane: Queensland Government (2012).
41. Bartlett B, Boffa J. The impact of Aboriginal community controlled health service advocacy on Aboriginal health policy. *Aust J Prim Health.* (2005) 11:53–61. doi: 10.1071/PY05022
42. Queensland Health and Queensland Aboriginal and Islander Health Council (QAIHC). *Making Tracks Towards Health Equity with Aboriginal and Torres Strait Islander Peoples-Working together to Achieve Life Expectancy Parity by 2031*. Discussion paper: a shared conversation Brisbane: State of Queensland and QAIHC (2021).
43. Panaretto KS, Dellit A, Hollins A, Wason G, Sidhom C, Chilcott K, et al. Understanding patient access patterns for primary health-care services for Aboriginal and Islander people in Queensland: a geospatial mapping approach. *Aust J Prim Health.* (2017) 23:37. doi: 10.1071/PY15115
44. Panaretto KS, Wenitong M, Button S, Ring IT. Aboriginal community controlled health services: leading the way in primary care. *Med J Aust.* (2014) 200:649–52. doi: 10.5694/mja13.00005
45. Coombe LL. The challenges of change management in Aboriginal community-controlled health organisations. Are there learnings for Cape York health reform? [Research Support, Non-U.S. Gov't Review]. *Austr Health Rev.* (2008) 32:639–47. doi: 10.1071/AH080639
46. Loppie Reading C, Wien F. *Health Inequalities and Social Determinants of Aboriginal Peoples' Health*. Prince George, BC: National Collaborating Centre for Aboriginal Health (2009).

Conflict of Interest: RF, KP, and SA are managers at Gurriny.

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: All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

Copyright © 2021 McCalman, Jongen, Campbell, Fagan, Pearson and Andrews. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

frontiers in Psychology

ORIGINAL RESEARCH
published: 16 December 2021
doi: 10.3389/fpsyg.2021.771028

Ingroup Bias in Healthcare Contexts: Israeli-Jewish Perceptions of Arab and Jewish Doctors

Elliot Graham[1]*, *Samer Halabi*[1] *and Arie Nadler*[2]

[1]*Department of Behavioral Sciences, Academic College of Tel-Aviv-Yaffo, Yaffo, Israel,* [2]*Department of Psychological sciences, Tel Aviv University, Tel Aviv, Israel*

The influence of group membership on perceptions of outgroup members has been extensively studied in various contexts. This research has indicated a strong tendency for ingroup bias – preferring the ingroup over the outgroup. We seek to further expand on the growing literature regarding the effects of group membership within healthcare contexts. Focusing on the Arab-Jewish context in Israel, the present study explored the influence of group membership on Israeli-Jewish participants' evaluations when exposed to potential malpractice. Specifically, participants ($n = 165$) read a description of an Israeli-Jewish or Israeli-Arab physician who was either culpable or non-culpable of malpractice. Consistent with our predictions, findings generally indicated more negative evaluations of the Israeli-Arab physician, regardless of objective culpability. We conclude by discussing the study's limitations and implications.

Keywords: ethnic bias, group membership, ingroup bias, physician ethnicity, ingroup favoritism, physician bias

OPEN ACCESS

Edited by:
Guido Veronese,
University of Milano-Bicocca, Italy

Reviewed by:
Ian Lustick,
University of Pennsylvania,
United States
Irene Razpurker-Apfeld,
Zefat Academic College, Israel

Correspondence:
Elliot Graham
grahamjelliot@gmail.com

Specialty section:
This article was submitted to
Health Psychology,
a section of the journal
Frontiers in Psychology

Received: *05 September 2021*
Accepted: *08 November 2021*
Published: *16 December 2021*

Citation:
Graham E, Halabi S and Nadler A (2021) Ingroup Bias in Healthcare Contexts: Israeli-Jewish Perceptions of Arab and Jewish Doctors.
Front. Psychol. 12:771028.
doi: 10.3389/fpsyg.2021.771028

INTRODUCTION

We live in a globalized world where we are continuously exposed to other groups, ethnicities, and cultures. Examining the effects of individuals' membership in groups that interact daily has therefore become increasingly relevant. Indeed, extensive research has been conducted to explore the important effects of group membership in intergroup relations across multiple life spheres (Allport, 1954; Tajfel, 1978; Pettigrew and Tropp, 2008). Israel, 20% of whose citizens belong to the Arab minority, provides an ideal context for studying how group membership can impact intergroup relations between that minority and the Jewish majority. In what follows, we focus on a specific and understudied setting: healthcare. Specifically, we investigated how a physician's group membership may influence Israeli-Jews' evaluations of that physician following potential malpractice.

Israel's public health system is a microcosm of its diverse social environment. In particular, there has recently been significant growth in the number of Israeli-Arabs employed in the medical and paramedical professions. Israeli-Arabs represent 19.2% of medical students and 14.7% of physicians (Averbuch and Avni, 2018; Israel Central Bureau of Statistics, 2018). Daily intergroup contact – such as Israeli-Jewish patients treated by Israeli-Arab physicians and *vice-versa* – may influence patients' perceptions of the specific medical processes, as well as general attitudes toward the provider and their group. Indeed, several qualitative studies conducted within the Israeli health system have reported on the way conflicted relations between Arabs and Jews in Israel affect attitudes toward outgroup caregivers, such as the preference for physician–patient concordance (or ingroup matchup; Popper-Giveon and Keshet, 2018; Popper-Giveon, 2019). In fact, an example of the conflicted relations within a medical setting could be observed when a member of the Israeli parliament was reported to have backed Jewish women who requested to be separated from Arab women while staying in maternity wards, a request that some hospitals were reported to have agreed to Siegel-Itzkovich and Solomon (2016). The present research focuses primarily on understanding the influence of the above relationship by exploring the effects of a physician's group membership on the perceptions of Israeli-Jewish participants regarding the extent of a physician's hypothetical culpability following potential malpractice.

Physicians usually enjoy high social status in addition to being the most respected profession among both groups of Israeli citizens (Haruti-Sover, 2017; Kadan et al., 2019). As such, it would be important to explore how Jewish members of this high-status group react to an Arab physician compared to Jewish physician. Indeed, research has indicated that members of high-status groups may recognize and inhibit obvious forms of discrimination, and yet be more affected by implicit negative stereotypes that are often remain subconscious (Stone and Moskowitz, 2011). Consequently, they may attribute more blame to outgroup than to ingroup members (Halabi et al., 2015). Pursuing this line of research in healthcare system, specifically in Israel, is most important as group membership is most salient and may trigger negative social perceptions that may affect judgments, evaluations, and behavior of potential patients.

Throughout life, individuals associate themselves and others with those who share a similar social identity – a social grouping defined as an *ingroup* – while those who differ are considered *outgroup* members (*we* and *them*; Tajfel, 1978; Abrams and Hogg, 1998). This recognition of others as in- or outgroup members has a robust, general effect on the perceptions and evaluations of others by displaying a preference for ingroup over outgroup members – or *ingroup bias* (Turner et al., 1979; Tajfel and Turner, 1986; Brown, 2019). Consequently, people tend to process information more deeply about their ingroup as opposed to any outgroup (Van Bavel et al., 2008). Additionally, people generally recall more unfavorable information about outgroup than about ingroup members (Howard and Rothbart, 1980). Furthermore, individuals evaluate ingroup members more positively and allocate resources more generously to ingroup than to outgroup members (Guimond, 2000; see Dovidio and Gaertner, 2010, for a review), especially when the intergroup context is salient, as postulated by social identity theory (Tajfel and Turner, 1986).

Importantly, people tend to attribute culpability for adverse outcomes differently for in- and outgroup members. For example, a study on linguistic intergroup bias (Maass et al., 1989) revealed a tendency to describe positive ingroup and negative outgroup behaviors in abstract terms, while describing negative ingroup and positive outgroup behaviors in concrete terms. For example, undesirable actions by outgroup members were perceived in a way that presumed intentionality and dispositional origin more than identical behaviors of ingroup members. Additional research showed that people more strongly attribute positive behaviors and successful outcomes to internal, stable characteristics of ingroup than outgroup members, whereas they ascribe the causes of adverse outcomes more strongly to the personalities of outgroup than ingroup members (Pettigrew, 1979; Hewstone, 1990).

In the current study, we extend these findings and explore the influence of group membership while focusing on the Israeli healthcare context. Specifically, we investigate the interethnic effects that may influence the evaluations of Israeli-Jewish participants regarding possible malpractice by an Israeli-Jewish vs. Israeli-Arab physician. Extensive research has indicated to the complicated relationships between Arabs and Jews in Israel (e.g., Enos and Gidron, 2016; Suleiman et al., 2018). Due to both historical, cultural, and contemporary political factors, the relationship between Israeli-Jews and Israeli-Arabs has been one of antagonism and conflict, creating a climate of suspicion, mistrust, and threat that has posed real difficulties to the engagement in shared living (Bar-Tal et al., 2010). Furthermore, these pervasive tensions between the two groups have contributed to the preservation and reinforcement of the ongoing discrimination and exclusion of Arabs in Israel (Smooha, 2014). At the same time, signs of rapprochement can be seen as well, as observed in the reduction in radicalization and improved attitudes between the groups observed from 2017 to 2019 (Smooha, 2020).

Studies conducted on the effects of intergroup relations in Israel have found that certain cues serve to categorize others as outgroup members, increasing biases in the process. One such study found that while positive imagined interaction with Arab-Muslim group members reduced intergroup anxiety among Jewish participants, the effect was significantly weakened when the Arab-Muslim member was perceived to display identifying Muslim religious symbols (Razpurker-Apfeld and Shamoa-Nir, 2020a). Another study conducted by Razpurker-Apfeld and Shamoa-Nir (2020b) presented similar results in regard with regard to motor cues, with arm extension found to increase intergroup anxiety and prejudice. These findings are consistent with those of a similar study on the effects of aural cues such as accents on enhancing ingroup biases (Jiang et al., 2020).

This tension may manifest itself within the Israeli healthcare system, where Israeli-Arabs and Jews interact daily as patients and healthcare providers. Indeed, the influence of the complicated relations between Jews and Arabs in Israel on the Israeli medical healthcare system has been observed in a studies that explored both physician–patient and physician–nurse interactions (Popper-Giveon et al., 2014; Keshet and Popper-Giveon, 2017; Popper-Giveon and Keshet, 2018). For example, Popper-Giveon et al. (2014) found that while Israeli-Arab physicians supportive of integration did not necessarily display a preference for patient concordance, both Israeli-Arab and Jewish patients would often rather be treated by physicians from their own ethnic group (see also Popper-Giveon, 2019). Further studies conducted in medical clinics in Israel found that positive intergroup contact could help reduce intergroup prejudice and improve attitudes both in a natural setting and in those based on positive information (Weiss, 2020, 2021). In light of these studies, we must also consider the positive effects found of patient-physician concordance on general health outcomes, as well as the likelihood of survival and correct treatment (Alsan et al., 2019; Greenwood et al., 2020).

The Present Study

In the current study, we focused on Jews, the majority group in Israel, and explored their response to a hypothetical case in which a physician, presented as an Israeli-Jew or Israeli-Arab, was responsible for malpractice that caused severe damage to a patient. Based on a previous study that indicated the influence of group membership on perceptions regarding culpability (Maass et al., 1989), we focused on variables that would represent the effects of group membership on the physician-patient relationship following a negative outcome. In particular, we assessed participants' emotions toward the physician, their trust in the physician, and the severity of punishment that they believed the physician deserved. We predicted that participants would display more negative attitudes toward an Arab compared to a Jewish physician across all our dependent variables and, importantly, regardless of the physician's culpability.

Considering the negative influence of group membership on perceived culpability when blame is not clear, a situation where blame can be more readily attributed should present an even "clearer" context; in which more negative reactions are made toward the culpable outgroup members (Maass et al., 1989; Schruijer et al., 1994). This is particularly so where contact is brief and impersonal, as in a hospital emergency room, with brief contact found to increase the effect of in-group biases (Forbes, 2004; Füchtbauer et al., 2013). Therefore, we expected to find a significant effect for Physician's Group Membership (Israeli-Jew vs. Israeli-Arab) × Culpability (High vs. Low) interactions regarding emotions expressed toward the physician, degree of trust in the physician, and the severity of punishment considered appropriate for the physician. Further, based on previous research that focused on the role of group-based emotions play in fueling and maintaining intergroup conflicts (Halperin, 2016) and on the role of trust in establishing positive intergroup relations (Bar-Tal et al., 2017), we also tested whether more positive emotions and more trust in the Israeli-Arab physician would mediate attributing, similar to the Israeli-Jewish physician, less severe punishment for the Israeli-Arab physician even when the physician is perceived as responsible for a potential malpractice.

MATERIALS AND METHODS

Participants and Design

Following approval from the institutional review board, a total of 163 Israeli-Jews (102 women) were recruited to participate in the study on voluntarily basis. Participants were recruited through advertisements in various social media platforms, using a convenience sampling method. We published a call for participants in a study involving real-life incidents to examine laypersons' information processing regarding the world of medicine. A link was provided within the post directing candidates to our questionnaire, which was conducted on Qualtrics. Participants then filled out an informed consent form with our contact information for any further inquiries. Data of all participants were retained and used for the analysis resulting in zero exclusion.

Ranging in age from 18 to 72 ($M = 28.16$; $SD = 7.82$), the participants were randomly assigned to one of four conditions within a 2 (Phynosician's Group Membership: Israeli-Jewish vs. Israeli-Arab) × 2 (Culpability: High vs. Low) between-subjects design. The goal was to recruit as many participants as possible up to an n of 128 (based on *a priori* statistical power analysis using G*Power version 3.1; Faul et al., 2009) to test all direct, interactive, and simple effects within a 2 × 2 factorial design with 80% power and a medium effect size. *Post-hoc* sensitivity analysis (Faul et al., 2009) showed a small effect size, $f = 0.25$ (Cohen, 1988). Thus, we had sufficient power to test our key predictions.

Procedure

The participants were informed they were participating in a study on how people perceived and reacted to real-life incidents involving social interactions. Following a general description of the study, they were asked to read a paragraph from an article published in a well-known newspaper. The paragraph supposedly included a description of a real-life event involving a physician and patient. Then, following the procedure employed by Halabi et al. (2017), the paragraph described a male physician called Alon, a common and exclusively Israeli-Jewish name, for the first group of participants ($n = 68$). For the other group of participants ($n = 95$), the male physician described in the paragraph was named Mahmoud, a common and exclusively Arab name[1]. Following this group membership manipulation, the participants were asked to state the physician's name and mother tongue. These items were designed to test the effectiveness of our group membership manipulation.

Next, the physician's culpability was manipulated. Specifically, participants were asked to read a brief paragraph including a description of a "real-life incident" that occurred in an Israeli hospital. In this incident, Yossi, a Jewish patient experienced irreversible damage following a severe allergic reaction to a presurgical sedative. Then, for approximately half of the

---

[1] It should be noted that the two conditions of physician group membership vary in size, 68 participants in the Israeli-Jewish condition vs. 95 participants in the Israeli-Arab condition. However, we believe that this did not affect our randomization process, as the experiment was conducted *via* the Qualtrics online platform. Indeed, as in all sound experimental studies, the random assignment of participants to the different conditions is the hallmark of a "true experiment." While random assignment can be accomplished by complete randomization, in which half of participants are randomly assigned to the control and half to the experimental condition, it can also be achieved by simple randomization, in which for each participant that volunteers for the experiment, there is a 50/50 chance that she/he ends up in the control or in the experimental condition – regardless of how large either sample already is. Simple randomization, as administered in the current study, may have caused the unequal sample sizes, yet it may have notably reduced the potential risk for selection bias (see Kahan et al., 2015) as the experimental software takes care of the randomization process.

participants ($n = 83$), the paragraph described the Jewish patient as being under stress and, when questioned by the physician, as failing to report any medicinal allergies, representing the low-culpability condition. In contrast, in the high-culpability condition, participants ($n = 80$) were presented with a case where the on-call physician was under stress in the emergency room and consequently did not inquire about any patient allergies.[2]

To examine the effectiveness of our culpability manipulation, we asked the participants to rate their agreement on whether the physician was culpable in the incident on a five-point scale ranging from 1 (*highly*) to 5 (*not at all*). Specifically, participants were asked, "To what extent is the physician culpable in the case you just read?"

Next, participants were asked to indicate their feelings and reactions regarding what they had just read. The following were our dependent variables: (a) emotions toward the physician, (b) trust in the physician, and (c) the severity of punishment that participants thought the physician deserved.

Emotions Toward the Physician

Emotions toward the physician have been linked to patients' positive affective states and a greater satisfaction with their physician (Hall et al., 2002; Worthington and Scherer, 2004). Alternatively, it was found that a lesser degree of outwardly expressed positive emotion by patients could lead to more negative caretaker dispositions (Ogrodniczuk et al., 2008). Participants were asked to rate on a five-point scale their emotions, toward the physician using four items adapted from Ogrodniczuk et al. (2008)[3]: (1) "To what degree do you feel sympathy toward the physician involved?"; (2) "To what degree do you feel admiration toward the physician involved?"; (3) "To what degree would you be willing to forgive the physician involved?"; and (4) "To what degree do you feel anger toward the physician involved?" (reverse-coded; Cronbach's alpha = 0.72). The items were averaged, with higher scores reflecting more positive emotions toward the physician.

Trust in the Physician

Trust has been found to play a significant role in positive patient-physician relationships in addition to being linked to patient satisfaction and adherence to treatment (Pearson and Raeke, 2000; Hall et al., 2001). Trust in the context of the patient-physician relationship is a multilayered construct that includes the patient's belief in a physician's competence and their confidence that their best interest is the primary consideration (Becker, 1996). Using a five-point scale, participants were asked to rate scale their trust in the physician *via* four items: (1) "To what degree would you trust this physician if he were treating you?"; (2) "To what degree would you recommend this physician to your friends?"; (3) "Compared to other physicians, would you positively evaluate the physician involved?"; and (4) "To what degree would you be willing for this physician to be your own?" (Cronbach's alpha = 0.88). The items were averaged, with higher scores reflecting higher trust.

Severity of Punishment

Based on previous studies that investigated the relationship between group membership and recommended severity of punishment (Halabi et al., 2015), participants were asked to rate on a five-point scale the severity of the punishment they thought the physician deserved *via* three items: (1) "Do you believe that the physician's license should be revoked?"; (2) "Do you believe that the physician should be criminally charged?"; and (3) "If you were the patient involved, would you have sued the physician?" (Cronbach's alpha = 0.75). The items were averaged, with higher scores reflecting a more severe punishment.

RESULTS

Preliminary analysis showed no significant effects for participants' gender; thus, this factor was not considered in the subsequent analysis. We then tested the effect of perceived culpability and the physician's group membership manipulations on the participants' emotions toward the physician, the extent of their trust in the physician, and the severity of punishment they believed that the physician deserved.

Manipulation Checks

To test the effectiveness of the physician's perceived culpability manipulation, a 2 (Physician's Group membership: Israeli-Jew vs. Israeli-Arab) × 2 (Culpability: High vs. Low) ANOVA was conducted on participants' level of agreement with the statement that the physician was culpable for the incident. This analysis revealed only the predicted main effect for the culpability manipulation, $F(1,164) = 41.25$, $p < 0.001$, $\eta^2 = 0.160$. Supportive of the intended manipulation, participants in the high-culpability condition ($M = 1.95$, $SD = 0.953$) perceived the physician as significantly more responsible than in the low-culpability condition ($M = 3.02$, $SD = 1.33$). No significant effects were found for physician group membership ($p = 0.873$) or for the interaction effect ($p = 0.090$).

For the group membership manipulation, all the participants correctly identified the physician's name and mother tongue according to the experimental condition.

---

[2] This culpability manipulation was piloted prior to the main study. Forty Israeli-Jewish participants were randomly recruited *via* Facebook to test the consistency and validity of our manipulation. As in the main study, participants were presented with a "news report." For approximately half of them, the physician could be perceived as more culpable ($n = 22$), while the other half received a report where the physician's culpability was low ($n = 20$). The results demonstrated that the manipulation was correctly understood ($M = 2.37$, $SD = 0.92$ versus $M = 1.57$, $SD = 0.56$; $t(30) = 3.32$, $p = 0.017$, $d = 1.05$).

[3] It should be mentioned that the original measure used by Ogrodniczuk et al. (2008) was not a self-reported scale and focused on caretakers' reactions toward the patient, thus differing from our study which may have influenced results. Furthermore, the original measure used broad terms for positive and negative emotions. In the current study, the emotions scale, adapted from Ogrodniczuk et al. (2008) focused on assessing emotions that previous studies on reactions to receiving help found to be indicative in describing the emotional state of participants.

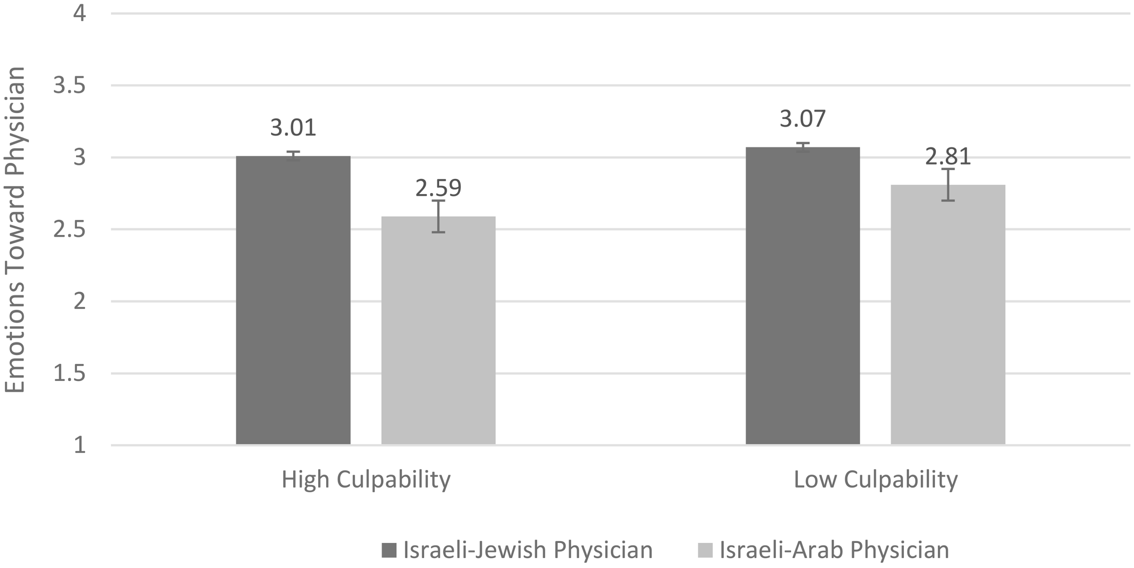


FIGURE 1 | Emotions toward Physician as a Function of Physician's Ethnicity and Culpability.

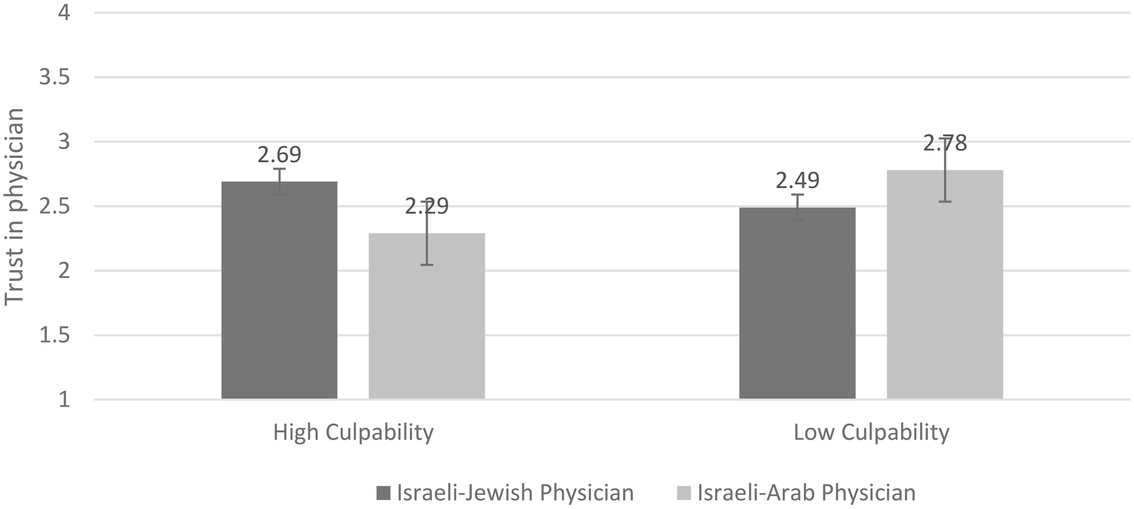


FIGURE 2 | Trust in Physician as a Function of Physician's Ethnicity and Culpability.

Ratings of the Physician

With respect to our main outcomes of interest, we conducted a 2 (Physician's Group membership: Israeli-Jew vs. Israeli-Arab) × 2 (Culpability: High vs. Low) between-subjects ANOVAs separately for indexes of emotions toward the physician, trust in the physician, and severity of punishment. Means and SDs of dependent variables across the study conditions are presented in **Table 1**.

For *emotions toward the physician*, the ANOVA demonstrated a significant main effect for ethnicity, $F(1,162) = 6.29$, $p = 0.01$, $\eta^2 = 0.04$. Participants reported more positive emotions toward the Israeli-Jewish physician than the Israeli-Arab physician, $Ms = 3.04$ ($SD = 0.86$) vs. 2.7 ($SD = 0.81$). The main effect of culpability was not significant, $F(1,162) = 1.15$, $p = 0.28$, $\eta^2 = 0.01$. Additionally, the Physician's group membership × Culpability (see **Figure 1**) interaction was not significant, $F(1,162) = 0.25$, $p = 0.61$, $\eta^2 = 0.002$.

A 2 (Physician's Group Membership) × 2 (Culpability) ANOVA on *trust in the physician* revealed only the significant two-way interaction, $F(1,162) = 5.66$, $p = 0.019$, $\eta^2 = 0.034$. Further analysis showed that participants in the high-culpability condition reported slightly higher trust in the Israeli-Jewish physician than in the Arab physician, $Ms = 2.69$ ($SD = 0.89$) vs. 2.29 ($SD = 0.79$), respectively, $t(159) = 1.92$, $p = 0.06$, $d = 0.47$. However, the comparable difference in the low-culpability condition did

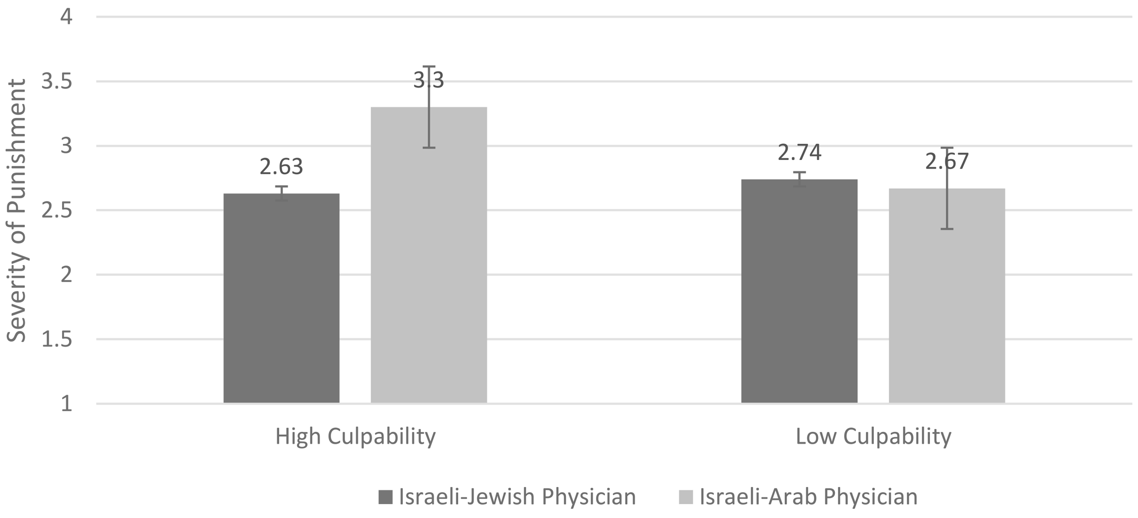


FIGURE 3 | Severity of Punishment as a function of Physician's Ethnicity and Culpability.

TABLE 1 | Means (SDs) of emotions towards the Physician, Trust in the Physician, and Severity of Punishment as a Function of Physician's Ethnicity and Culpability.

Physician's Ethnicity	**Experimental condition**			
	Israeli-Jewish		**Israeli-Arab**	
Culpability:				
	High (n =32)	Low (n =36)	High (n=48)	Low (n =47)
Dependent measures:				
Emotions towards the Physician	3.01 (0.82)	3.07 (0.91)	2.59 (0.78)	2.81 (0.84)
Trust in the Physician	2.69 (0.89)	2.49 (1.06)	2.29 (0.79)	2.78 (0.89)
Severity of Punishment	2.63 (0.97)	2.74 (1.14)	3.30 (0.87)	2.67 (0.80)

not approach significance, $Ms = 2.49$ ($SD = 1.08$) vs. 2.78 ($SD = 0.89$), respectively; $t(159) = 1.43$, $p = 0.15$, $d = 0.96$. Looked at differently, *post hoc* analysis showed that in the Israeli-Arab physician condition, participants reported significantly higher trust in the physician in the low-culpability than in the high-culpability condition, $Ms = 2.78$ ($SD = 0.89$) vs. 2.29 ($SD = 0.79$), respectively; $t(159) = 2.60$, $p = 0.01$, $d = 0.57$. However, in the Israeli-Jewish physician condition, the degree of trust did not differ between the high- and low-culpability conditions, $Ms = 2.69$ ($SD = 0.89$) vs. 2.49 ($SD = 1.06$), respectively; $t(159) = 0.92$, $p = 0.36$, $d = 0.22$ (see **Figure 2**).

Finally, a 2 (Physician's Group Membership) × 2 (Culpability) ANOVA on *severity of punishment* revealed a significant main effect for ethnicity, $F(1,162) = 4.07$, $p = 0.04$, $\eta^2 = 0.025$. As expected, participants attributed more severe punishment to the Israeli-Arab physician than to the Israeli-Jewish physician, $Ms = 2.99$ ($SD = 0.89$) vs. 2.69 ($SD = 1.05$), $d = 0.31$. Furthermore, as predicted, a significant two-way Group Membership of Physician × Culpability interaction effect was found, $F(1,162) = 6.07$, $p = 0.01$, $\eta^2 = 0.04$. Further analysis showed that participants in the high-culpability condition attributed significantly more severe punishment to the Israeli-Arab than to the Israeli-Jewish physician, $Ms = 3.30$ ($SD = 0.877$) vs. 2.63 ($SD = 0.970$), respectively; $t(159) = 3.12$, $p = 0.002$, $d = 0.72$. However, in the low-culpability condition, no such significant difference was found, $Ms = 2.67$ ($SD = 0.8$) vs. 2.74 ($SD = 1.14$), respectively; $t(159) = 0.33$, $p = 0.75$, $d = 0.1$. Interestingly, when observing the participants' perceptions regarding the Israeli-Jewish physician, no statistically significant differences were found between the high- and low-culpability conditions in severity of punishment, $Ms = 2.63$ ($SD = 0.97$) vs. 2.74 ($SD = 1.14$), respectively; $t(159) = 0.46$, $p = 0.64$, $d = 0.1$. Furthermore, note that, no statistical difference was found between the high-culpability-Israeli-Jewish physician and low-culpability-Israeli-Arab physician conditions, $Ms = 2.63$ ($SD = 0.970$) vs. 2.67 ($SD = 0.8$), respectively; $t(159) = 0.39$, $p = 0.69$, $d = 0.04$ (see **Figure 3**).

DISCUSSION

The present study examined the effects of group membership on perceptions of Israeli Jews in the understudied context of the healthcare system. Specifically, we explored Israeli-Jewish evaluations of male physicians presented as either Israeli-Arab or Israeli-Jewish. Our findings illuminate the crucial role that a physician's ethnicity may play in eliciting positive or negative emotions of ingroup vs. outgroup members toward them, as well as perceptions regarding the severity of punishment that the physician deserved following potential malpractice. Furthermore, the current findings presented that participants were more likely to be influenced by the Israeli-Arab physician's group membership in ratings of trust in the physician when the physician was presented as more responsible for the malpractice. Specifically, Israeli-Jewish participants reported more negative emotions toward the Israeli-Arab physician than toward the Israeli-Jewish physician. Furthermore, when the physician was presented as more responsible for potential malpractice the participants reported a higher degree of trust and attributed the less severe punishment toward the Israeli-Jewish physician when compared to the Israeli-Arab physician. Importantly, the severity of punishment attributed to the physician in the high-culpability condition was mediated by emotions reported toward the physician and by extent of trust that participants displayed toward him.

Our results show how objective culpability and ethnicity intertwine, with participants significantly more likely to display more negative emotions toward Israeli-Arab physicians and assign them more punishment than Israeli-Jewish ones, when presented as culpable for a medical malpractice. The current findings indicate the harmful effects that group membership can have on an individual's emotions toward an outgroup physician. Additionally, when considering our findings on the influence of group membership on the severity of punishment, the significance of the above becomes clearer, further emphasizing how the needs of punishment and positive emotions such as forgiveness can often be intertwined (Worthington et al., 2005).

As predicted, the influence of group membership on the desire for punishment and the capacity for trust was significantly more evident in cases where the Israeli-Arab physician's culpability was high, while no such effect was found where the culpability of the physician (of whatever ethnicity) was low. These results emphasize the impact that culpability can have in exposing and enhancing ingroup bias – specifically, the harmful role that an Israeli-Arab physician's culpability plays in Israeli-Jewish group members' trust and desire for punishment. When questioning how these influences may impact an Israeli-Arab physician, we can begin by reiterating the impact that a patient's trust in a physician has on the quality of their interaction (Pearson and Raeke, 2000; Hall et al., 2001). We also might speculate how this trust could potentially display itself regarding an Israeli-Arab's employer once an unfortunate incident occurs. Additionally, we can consider how the observed group bias impacts Israeli-Jews' perceptions regarding the severity of punishment that Israeli-Arab and -Jewish physicians deserve following medical malpractice, potentially affecting a vast number of legal cases.

Looked at differently, the current findings provide an interesting perspective in understanding prejudice and stereotyping when outgroup members of higher social status are involved. In particular, Israeli-Jewish participants reacted more negatively to Israeli-Arab physicians only when they were to blame for medical malpractice. This suggests that when the outgroup member, even with high social status, behaves in a way that "reminds" the ingroup observer of the other's status affiliation, interpersonal perceptions are likely to be affected by this affiliation more than by the higher status (Halabi and Nadler, 2021). However, this important theoretical extension needs to be further examined in future research.

Crucially, one of our most significant findings was the lack of observable difference, across all variables, between the high- and low-culpability Israeli-Jewish physicians. Additionally, we found no observable differences between the low-culpability Israeli-Arab physician and high-culpability Israeli-Jewish physician. This lack of observed differences between the Israeli-Jewish physicians could be problematic in terms of their ability to learn from their errors, affecting the system as a whole. Conversely, the results regarding the lack of observable differences between the low-culpability Israeli-Arab physician and the high culpability Israeli-Jewish physician, in conjunction with the significant difference between the Israeli-Arab physicians across all variables, suggest that Israeli-Jews are more critical of Israeli-Arab physicians, expecting less, and once a mistake is made, retroactively assuming that they have been generally less professional. These results strongly emphasize, in a medical context, that group membership can influence perceptions, such as people's capacity to judge situations objectively and perceive culpability when members of their ingroup are involved (Hewstone, 1990; Holmberg and Kyvsgaard, 2003).

Several possible explanations may be provided for the uneven results in several of our variables. A number of our participants may have felt weaker identification with their ingroup, thus influencing their sense of group homogeneity, a substantial ingredient in reducing the negative influence of group membership (Wilder, 1978; Dietz-Uhler and Murrell, 1998; Hutchison et al., 2006). Future studies could benefit from expanding upon the possible moderating influence of group identification on group membership bias within a medical setting. Additionally, the results showed no significant difference between the Israeli-Jewish and Israeli-Arab physicians in the low-culpability condition. It is conceivable that, by inquiring into the nature of the patient's allergies, the Israeli-Arab physician appeared to be showing more concern, thus reducing the effects of intergroup bias (Giannakakis and Fritsche, 2010).

LIMITATIONS AND FUTURE DIRECTIONS

The first limitation of this study is that whereas its theoretical implications present an opportunity to understand the relationship between Israeli-Jews and their Israeli-Arab physicians, due to the absence of Israeli-Arab participants, we are unable to conclude that all types of ingroup membership lead to discrimination against outgroup physicians. Future studies should attempt to build an additional sample consisting of Israeli-Arabs.

Additionally, the nature of this study, conducted on a general population sample, prevents us from observing the nature of group membership bias within an actual emergency care medical environment, such as the one presented in our study. While studies such as this allow for greater degree of control, prior research conducted regarding ingroup bias toward Arabs in Israel indicated multiple benefits for studies conducted using natural experiment settings, such as naturally occurring groups, professional judgement, and higher stakes (Shayo and Zussman, 2011; Grossman et al., 2016; Bar and Zussman, 2020). Further research may greatly benefit from a similar study, with a more gender-balanced sample, held in a hospital setting with a patient-focused sample, as well with hospital administrations that may be also affected by ethnicity and perceived physician culpability. Such a study may represent the population more appropriately and strengthen the validity of our theoretical and applied findings. Furthermore, while our results yielded a statistically significant difference regarding observable discrimination towards Israeli-Arab physicians, seeing as our results generally clustered around the midway point we must consider that this difference may not always be as perceivable in real life scenarios.

Finally, our study only attempted to observe how group membership could create a bias in perceptions of the physician-patient relationship. For that reason, we believe that future studies conducted in the field should attempt to find specific methods to reduce the impact of group membership bias on the physician-patient relationship. For example, considering the positive effects found for physician-patient concordance within other medical settings, examining the impact of concordance within an emergency room using a natural experiment may help underscore the need for increasing physician diversity. Other studies may focus on how moderating factors such as spreading positive information in the media may help reduce bias within a framework such as that presented here (Ortiz and Harwood, 2007; Weiss, 2020).

CONCLUSION

Using an experimental design, we built upon previous evidence for the existence of group membership bias in the healthcare context. Our study persuasively presents the existence of such bias against Israel-Arab physicians within the Israeli health system, implying that this effect can probably be found within actual physician-patient relationships, especially if the physician is suspected of malpractice.

These harmful influences – resulting from ethnic ingroup bias – could have grave real-world consequences, and as observed in our study, they can influence emotions toward the physician, trust in the physician, and the severity of punishment in cases of suspected malpractice. These could dramatically affect an Israeli-Arab physician's career path and the quality of physician–patient interaction (Pearson and Raeke, 2000; Baicker et al., 2007; Shurtz, 2013). This impact is crucial considering the current rise in the number of Israeli-Arab physicians employed throughout the Israeli healthcare system.

We believe that the path to reducing the harmful influences of group membership begins with their deeper understanding. Hopefully, the information provided in this study serves as a significant step in the healthcare context.

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 Institutional Review Board. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

EG was responsible on data collection and data analysis and writing research report. SH was responsible on designing the study, and involved in data analysis and writing. AN was involved in supervising and writing research report. All authors contributed to the article and approved the submitted version.

FUNDING

This research was partially supported by a research grant from the Israeli Science Foundation (grant no. 1288/16) awarded to SH and AN.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2021.771028/full#supplementary-material

REFERENCES

Abrams, D., and Hogg, M. (1998). *Social identifications*. London: Routledge.

Allport, G. W. (1954). *The Nature of Prejudice* Addison-Wesley.

Alsan, M., Garrick, O., and Graziani, G. (2019). Does diversity matter for health? Experimental evidence from Oakland. *Am. Econ. Rev.* 109, 4071–4111. doi: 10.1257/aer.20181446

Averbuch, E., and Avni, S. (2018). Inequality in health and coping with it. Jerusalem: Israeli Ministry of Health [Hebrew].

Baicker, K., Fisher, E. S., and Chandra, A. (2007). Malpractice liability costs and the practice of medicine in the Medicare program. *Health Aff.* 26, 841–852. doi: 10.1377/hlthaff.26.3.841

Bar, R., and Zussman, A. (2020). Identity and bias: insights from driving tests. *Econ. J.* 130, 1–23. doi: 10.1093/ej/uez048

Bar-Tal, D., Halperin, E., and Oren, N. (2010). Socio–psychological barriers to peace making: The case of the Israeli Jewish society. *Soc. Issues Policy Rev.* 4, 63–109. doi: 10.1111/j.1751-2409.2010.01018.x

Bar-Tal, D., Raviv, A., Shapira, P., and Kahn, D. (2017). "Lay psychology of trust/distrust and beyond in the context of an intractable conflict: The case of Israeli Jews," in *The Role of Trust in Conflict Resolution: The Israeli–Palestinian Case and beyond*. eds. I. Alon and D. Bar-Tal (Cham, Switzerland: Springer), 197–213.

Becker, L. C. (1996). Trust as noncognitive security about motives. *Ethics* 107, 43–61. doi: 10.1086/233696

Brown, R. (2019). The social identity approach: appraising the Tajfellian legacy. *Br. J. Soc. Psychol.* 59, 5–25. doi: 10.1111/bjso.12349

Cohen, J. (1988). *Statistical Power Analysis for the Behavioral Sciences. 2nd Edn.* Lawrence Erlbaum Associates.

Dietz-Uhler, B., and Murrell, A. (1998). Effects of social identity and threat on self-esteem and group attributions. *Group Dyn. Theory Res. Pract.* 2, 24–35. doi: 10.1037/1089-2699.2.1.24

Dovidio, J. F., and Gaertner, S. L. (2010). "Intergroup bias," in *Handbook of Social Psychology*. eds. S. T. Fiske, D. T. Gilbert and G. Lindzey (New York: John Wiley and Sons), 1084–1121.

Enos, R. D., and Gidron, N. (2016). Intergroup behavioral strategies as contextually determined: experimental evidence from Israel. *J. Polit.* 78, 851–867. doi: 10.1086/685545

Faul, F., Erdfelder, E., Buchner, A., and Lang, A. G. (2009). Statistical power analyses using G* power 3.1: tests for correlation and regression analyses. *Behav. Res. Methods* 41, 1149–1160. doi: 10.3758/BRM.41.4.1149

Forbes, H. D. (2004). "Ethnic conflict and the contact hypothesis," in *The Psychology of Ethnic and Cultural Conflict*. eds. Y.-T. Lee, C. McCauley, F. Moghaddam and S. Worchel (Westport, Connecticut: Praeger Publishers/Greenwood Publishing Group), 69–88.

Füchtbauer, L. M., Nørgaard, B., and Mogensen, C. B. (2013). Emergency department physicians spend only 25% of their working time on direct patient care. *Dan. Med. J.* 60:A4558

Giannakakis, A. E., and Fritsche, I. (2010). Social identities, group norms, and threat: On the malleability of ingroup bias. *Personal. Soc. Psychol. Bull.* 37, 82–93. doi: 10.1177/0146167210386120

Greenwood, B. N., Hardeman, R. R., Huang, L., and Sojourner, A. (2020). Physician–patient racial concordance and disparities in birthing mortality for newborns. *Proc. Natl. Acad. Sci.* 117, 21194–21200. doi: 10.1073/pnas.1913405117

Grossman, G., Gazal-Ayal, O., Pimentel, S. D., and Weinstein, J. M. (2016). Descriptive representation and judicial outcomes in multiethnic societies. *Am. J. Polit. Sci.* 60, 44–69. doi: 10.1111/ajps.12187

Guimond, S. (2000). Group socialization and prejudice: The social transmission of intergroup attitudes and beliefs. *Eur. J. Soc. Psychol.* 30, 335–354. doi: 10.1002/(SICI)1099-0992(200005/06)30:3<335::AID-EJSP994>3.0.CO;2-V

Halabi, S., Dovidio, J. F., and Nadler, A. (2017). When intergroup apology is not enough: seeking help and reactions to receiving help among members of low status groups. *Eur. J. Soc. Psychol.* 48, 118–133. doi: 10.1002/ejsp.2309

Halabi, S., Statman, Y., and Dovidio, J. F. (2015). Attributions of responsibility and punishment for ingroup and outgroup members: The role of just world beliefs. *Group Process Intergroup Relat.* 18, 104–115. doi: 10.1177/1368430214546067

Hall, M. A., Dugan, E., Zheng, B., and Mishra, A. K. (2001). Trust in physicians and medical institutions: what is it, can it be measured, and does it matter? *Milbank Q.* 79, 613–639. doi: 10.1111/1468-0009.00223

Hall, J. A., Horgan, T. G., Stein, T. S., and Roter, D. L. (2002). Liking in the physician–patient relationship. *Patient Educ. Couns.* 48, 69–77. doi: 10.1016/S0738-3991(02)00071-X

Halperin, E. (2016). *Emotions in Conflict: Inhibitors and Facilitators of Peace Making*. New York: Routledge.

Haruti-Sover, T. (2017). Which professions are most valued in the eyes of the Israeli public? The status of teachers surpassed that of career soldiers. The Marker. Available at: https://www.themarker.com/career/1.4367043 (Accessed October 5, 2021).

Hewstone, M. (1990). The 'ultimate attribution error'? A review of the literature on intergroup causal attribution. *Eur. J. Soc. Psychol.* 20, 311–335. doi: 10.1002/ejsp.2420200404

Holmberg, L., and Kyvsgaard, B. (2003). Are immigrants and their descendants discriminated against in the Danish criminal justice system? *J. Scand. Stud. Criminol. Crime Prev.* 4, 125–142. doi: 10.1080/14043850310020027

Howard, J. W., and Rothbart, M. (1980). Social categorization and memory for ingroup and out-group behavior. *J. Pers. Soc. Psychol.* 38, 301–310. doi: 10.1037/0022-3514.38.2.301

Hutchison, P., Jetten, J., Christian, J., and Haycraft, E. (2006). Protecting threatened identity: sticking with the group by emphasizing ingroup heterogeneity. *Personal. Soc. Psychol. Bull.* 32, 1620–1632. doi: 10.1177/0146167206292235

Israel Central Bureau of Statistics (2018). Medical studies at universities, 1999/00-2016/17. [Hebrew]. Available at: https://www.cbs.gov.il/he/mediarelease/DocLib/2018/200/06_18_200b.pdf (Accessed October 15, 2021).

Jiang, X., Gossack-Keenan, K., and Pell, M. D. (2020). To believe or not to believe? How voice and accent information in speech alter listener impressions of trust. *Q. J. Exp. Psychol.* 73, 55–79. doi: 10.1177/1747021819865833

Kadan, S., Bekerman, Z., and Roer-Strier, D. (2019). Career choice in the context of political conflict: The case of Palestinians social workers in Israel. *Intercult. Educ.* 30, 31–50. doi: 10.1080/14675986.2018.1495425

Kahan, B. C., Rehal, S., and Cro, S. (2015). Risk of selection bias in randomised trials. *Trials* 16:405. doi: 10.1186/s13063-015-0920-x

Keshet, Y., and Popper-Giveon, A. (2017). Race-based experiences of ethnic minority health professionals: Arab physicians and nurses in Israeli public healthcare organizations. *Ethn. Health* 23, 442–459. doi: 10.1080/13557858.2017.1280131

Maass, A., Salvi, D., Arcuri, L., and Semin, G. R. (1989). Language use in intergroup contexts: The linguistic intergroup bias. *J. Pers. Soc. Psychol.* 57, 981–993. doi: 10.1037/0022-3514.57.6.981

Ogrodniczuk, J. S., Piper, W. E., and Joyce, A. S. (2008). Alexithymia and therapist reactions to the patient: expression of positive emotion as a mediator. *Psychiatry* 71, 257–265. doi: 10.1521/psyc.2008.71.3.257

Ortiz, M., and Harwood, J. (2007). A social cognitive theory approach to the effects of mediated intergroup contact on intergroup attitudes. *J. Broadcast. Electron. Media* 51, 615–631. doi: 10.1080/08838150701626487

Pearson, S. D., and Raeke, L. H. (2000). Patients' trust in physicians: many theories, few measures, and little data. *J. Gen. Intern. Med.* 15, 509–513. doi: 10.1046/j.1525-1497.2000.11002.x

Pettigrew, T. F. (1979). The ultimate attribution error: extending Allport's cognitive analysis of prejudice. *Personal. Soc. Psychol. Bull.* 5, 461–476. doi: 10.1177/014616727900500407

Pettigrew, T. F., and Tropp, L. R. (2008). How does intergroup contact reduce prejudice? Meta-analytic tests of three mediators. *Eur. J. Soc. Psychol.* 38, 922–934. doi: 10.1002/ejsp.504

Popper-Giveon, A. (2019). Preferring patient–physician concordance: The ambiguity of implicit ethnic bias. *Ethn. Health* 26, 1065–1081. doi: 10.1080/13557858.2019.1620180

Popper-Giveon, A., and Keshet, Y. (2018). The secret drama at the patient's bedside—refusal of treatment because of the practitioner's ethnic identity: The medical staff's point of view. *Qual. Health Res.* 28, 711–720. doi: 10.1177/1049732318755676

Popper-Giveon, A., Liberman, I., and Keshet, Y. (2014). Workforce ethnic diversity and culturally competent health care: The case of Arab physicians in Israel. *Ethn. Health* 19, 645–658. doi: 10.1080/13557858.2014.893563

Razpurker-Apfeld, I., and Shamoa-Nir, L. (2020a). Is an outgroup welcome with open arms? Approach and avoidance motor activations and outgroup prejudice. *J. Exp. Psychol. Appl.* 27, 417–429. doi: 10.1037/xap0000334

Razpurker-Apfeld, I., and Shamoa-Nir, L. (2020b). Imagined contact with strongly identified outgroup members: do religious trappings make the man? *Asian J. Soc. Psychol.* 23, 384–396. doi: 10.1111/ajsp.12412

Schruijer, S., Blanz, M., Mummendey, A., Tedeschi, J., Banfai, B., Dittmar, H., et al. (1994). The group-serving bias in evaluating and explaining harmful behavior. *J. Soc. Psychol.* 134, 47–53. doi: 10.1080/00224545.1994.9710882

Shayo, M., and Zussman, A. (2011). Judicial ingroup bias in the shadow of terrorism. *Q. J. Econ.* 126, 1447–1484. doi: 10.1093/qje/qjr022

Shurtz, I. (2013). The impact of medical errors on physician behavior: evidence from malpractice litigation. *J. Health Econ.* 32, 331–340. doi: 10.1016/j.jhealeco.2012.11.011

Siegel-Itzkovich, J., and Solomon, B.A. (2016). Found: Smotrich supports hospital room segregation between Jews and Arabs. The Jerusalem Post. Available at: https://www.jpost.com/israel-news/report-various-israeli-hospitals-separate-pregnant-jewish-arab-on-request-450271 (Accessed October 13. 2021).

Smooha, S. (2014). Index of Arab-Jewish Relations in Israel 2012. The Israel Democracy Institute (R.A.) and University of Haifa.

Smooha, S. (2020). Still Playing by the Roles: Index of Arab-Jewish Relations in Israel 2019. University of Haifa.

Stone, J., and Moskowitz, G. B. (2011). Non-conscious bias in medical decision making: what can be done to reduce it? *Med. Educ.* 45, 768–776. doi: 10.1111/j.1365-2923.2011.04026.x

Suleiman, R., Yahya, R., Decety, J., and Shamay-Tsoory, S. (2018). The impact of implicitly and explicitly primed ingroup–outgroup categorization on the evaluation of others pain: The case of the Jewish–Arab conflict. *Motiv. Emot.* 42, 438–445. doi: 10.1007/s11031-018-9677-3

Tajfel, H. (1978). *Differentiation between Social Groups: Studies in the Social Psychology of Intergroup Relations* Academic Press.

Tajfel, H., and Turner, J. C. (1986). "The social identity theory of intergroup behavior," in *Psychology of Intergroup Relations*. eds. S. Worchel and W. G. Austin (Chicago: Hall), 7–24.

Turner, J. C., Brown, R. J., and Tajfel, H. (1979). Social comparison and group interest in ingroup favouritism. *Eur. J. Soc. Psychol.* 9, 187–204. doi: 10.1002/ejsp.2420090207

Van Bavel, J. J., Packer, D. J., and Cunningham, W. A. (2008). The neural substrates of ingroup bias. *Psychol. Sci.* 19, 1131–1139. doi: 10.1111/j.1467-9280.2008.02214.x

Weiss, C. (2020). "Representation in healthcare institutions promotes intergroup tolerance: evidence from the COVID-19 crises in Israel and the US." APSA [Preprint]. doi: 10.33774/apsa-2020-6j6z3

Weiss, C. M. (2021). Diversity in health care institutions reduces Israeli patients' prejudice toward Arabs. *Proc. Natl. Acad. Sci.* 118:e2022634118. doi: 10.1073/pnas.2022634118

Wilder, D. A. (1978). Homogeneity of jurors. *Law Hum. Behav.* 2, 363–376. doi: 10.1007/BF01038988

Worthington, E. L. Jr., and Scherer, M. (2004). Forgiveness is an emotion-focused coping strategy that can reduce health risks and promote health resilience: theory, review, and hypotheses. *Psychol. Health* 19, 385–405. doi: 10.1080/0887044042000196674

Worthington, E. L. Jr., van Oyen Witvliet, C., Lerner, A. J., and Scherer, M. (2005). Forgiveness in health research and medical practice. *Exp. Dermatol.* 1, 169–176. doi: 10.1016/j.explore.2005.02.012

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: All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

Copyright © 2021 Graham, Halabi and Nadler. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

frontiers
in Public Health

ORIGINAL RESEARCH
published: 24 January 2022
doi: 10.3389/fpubh.2021.798222

Health Literacy and Migrant Communities in Primary Health Care

Patrícia Medina[1], **Ana Catarina Maia**[1,2] **and Andreia Costa**[1,3]*

[1] Nursing Research, Innovation and Development Centre of Lisbon (CIDNUR), Nursing School of Lisbon (ESEL), Lisboa, Portugal, [2] NOVA National School of Public Health, Public Health Research Centre, Universidade NOVA de Lisboa; Comprehensive Health Research Centre (CHRC), Lisboa, Portugal, [3] Instituto de Saúde Ambiental (ISAMB), Faculdade de Medicina, Universidade de Lisboa, Lisboa, Portugal

Introduction: The promotion of health literacy of the population in a situation of migration, in the community, is a fundamental field of intervention in health promotion, for the reduction of inequalities in access to health care services. It is increasingly necessary to make health care services more equitable for migrant populations. The aim of the study was to characterize the level of health literacy of the population in a migrant situation, attending a primary health care unit in the Lisbon region, to identify priority areas for community intervention that will become the focus of intervention and contribute to the increase in the health literacy levels in this population.

Methods: A cross-sectional study was carried out by applying the Health Literacy Survey (ILS-PT) to a sample of the population in a situation of migration, found by 27 participants.

Results: The general health literacy index of the sample is inadequate (21.23 points). An analysis of the sub-indexes revealed that 75% of the participants had difficulties related to information about health care and 80% had difficulties in the field of health promotion.

Conclusions: Problematic and inadequate levels of health literacy was significantly frequent among migrant population. So that enhancing health literacy among migrant is essential to reduce health inequalities to achieve better health outcomes and contribute to defense of human rights of this vulnerable population.

Keywords: health literacy, health promotion, migrants, equity, primary health care

OPEN ACCESS

Edited by:
Aldo Rosano,
National Institute for the Analysis of Public Policy, Italy

Reviewed by:
Katielle Silva,
University of Lisbon, Portugal
Erika Masanet Ripoll,
University of Valencia, Spain

***Correspondence:**
Andreia Costa
andreia.costa@esel.pt

Specialty section:
This article was submitted to Life-Course Epidemiology and Social Inequalities in Health, a section of the journal Frontiers in Public Health

Received: *19 October 2021*
Accepted: *16 December 2021*
Published: *24 January 2022*

Citation:
Medina P, Maia AC and Costa A (2022) Health Literacy and Migrant Communities in Primary Health Care. Front. Public Health 9:798222. doi: 10.3389/fpubh.2021.798222

INTRODUCTION

There are currently about 244 million migrants worldwide (1). A migrant is understood to be any person who moves or crosses an international border far from their place of habitual residence, regardless of their legal status, voluntary movement, causes of movement or length of stay (2).

In Portugal, data released indicate that, for the third consecutive year, there was an increase in the foreign population residing in Portugal, with an increase of 13, 9% compared to 2017 data, making a total of 480.3 thousand foreigners (3). The migrant population in Portugal tends to be concentrated in coastal regions, with the Metropolitan Area of Lisbon being the geographical area of the country with the highest percentage of foreign population (68.9%) (3).

The migratory process affects and is affected by the social determinants of health, as well as by the conditions in which the migratory process takes place, influencing the health situation, when leaving the countries of origin and when arriving in the countries of destination, influencing the process of integration in societies. Some studies developed in the European context and in Portugal shows the phenomenon "healthy migrant effect," that is, the tendency of self-perception of a good health status upon arrival in the host countries. (4, 5). However, it is observed that throughout the time of stay in the host country, migrants tend to report a decline in their health status with reference to chronic diseases and disability (5, 6). In fact, studies conducted in Portugal on the health of migrants express the situation of greater vulnerability of certain groups to the development of chronic diseases and health problems with impact on the social determinants of health (5, 7). In fact, in some situations, the migration process can have a negative impact on the health status of the people involved, reinforcing their condition of vulnerability. This is the case of children and unaccompanied minors, women, elderly, people with disabilities, irregular migrants, refugees, asylum seekers, and migrants who have been subjected to human trafficking, so it is essential to understand their vulnerability and the impact it has on their health condition (8). Migrant health must be seen as a unified agenda, involving global health and universal health coverage, sustainable development goals and social determinants of health (9). In this sense, interventions must be designed to reduce inequities in health and increase the resilience of people in a situation of migration. Promoting health equity through universal health coverage with the broad participation of primary health care, ensures that improving the health of migrants is an essential part of achieving the Sustainable Development Goals addressed in the 2030 Agenda for Sustainable Development, adopted by all United Nations Member States in 2015. In this line of ideas, the design and implementation of interventions aimed at promoting the health and well-being of migrants, should have as an outcome the issue of equity on the agenda of health care providers, researchers in health sciences and in policies health care (10). Equity in health is of crucial importance for the defense of human rights in migrant populations, in view of the situation of increased vulnerability and their specific needs of this population at community and individual levels (10, 11). Aspects that for legal reasons, reduce access to health care and health information; and the environmental and integration issues of host countries, as well as the barriers related to communication, all have an impact on health equity (11).

Thus, promoting Health Literacy is a fundamental strategy for empowering citizens and promoting equity in access to health care, as described by the Portuguese National Health Plan: 2012–2016 revision and extension to 2020 (12), reducing its characteristic vulnerability associated with the migration process. In the Portuguese context, the Strategic Plan for Migration (13) (2015), structured by the Government of Portugal, highlights the universal access of migrant populations, regardless of their regularization status. On the other hand, the plan states that measures for the integration of migrants in Portugal should be structured and comprehensive for all health services, as universal access to health care by migrants and a better and more effective use of health services by migrant populations have been seen as "key indicators of social inclusion" and integration in the host country (14, 15). Initially, the concept of Health Literacy was defined by the World Health Organization as the set of cognitive and social skills that determine the motivation and ability of individuals to access, understand and use information to promote and maintain good health (16).

In the specific case of this study, the Integrated Conceptual Model of Health Literacy was used, in which health literacy is related to literacy and implies the knowledge, motivation and skills of the person to access, understand, evaluate and apply the health information in order to make judgments and make decisions on a daily basis relating to healthcare, disease prevention and health promotion in order to maintain or improve the quality of life over the course of life (17).

The health Literacy promotion process according to the present model requires four types of key competences: access, which refers to the ability to seek, find and obtain information about health; understand: referring to the ability to understand the health information that was obtained; evaluate: competence related to the ability to interpret, filter, judge and evaluate the health information that was accessed; apply: ability to communicate and use information that affect the decisions aimed at maintaining or improving health (17).

The population in a situation of migration is a naturally more vulnerable group, either because of the increased risk associated with the migration process itself and the conditions in which it occurs, or because of the situational conditions of integration in the host country, as access to health care and health information, that are determining factors for improving their health status (18).

Low health literacy and poor citizen autonomy toward health services are identified as "threats to equity and access to health care" by the National Health Plan 2012–2016 (19). This Plan proposes that literacy promotion actions be carried out, focused on measures to promote health and disease prevention, which can be developed and implemented by specialist community nurses (19).

The present study aims to describe the level of health literacy of a sample of migrant people who attend primary health care in a unit in the Lisbon region, to identify priority areas of community intervention initiated by specialist community nurses to increase literacy in health in these populations.

METHODS

Study Design

This was a cross-sectional study conducted on a primary healthcare service, in Lisbon region, between 30th September and 16th October 2019. Ethical issues were validated by an independent commission before the data collection and was also obtained the approval from the executive director from the primary health structure.

Study Participants

The population studied were persons that were in migration situation, and that resorted to a primary healthcare service in Lisbon region. A convenience sample of eligible participants was invited to participate in the study. Migrants were invited to participate through personal invitation when attending the primary healthcare service to receive nursing care. Were defined the following eligibility criteria: being over 18 years old; finding themselves in a situation of migration; have an oral proficiency of the Portuguese language; having accepted to participate in the study, after being explained by the researchers the contents of the study information sheet and after signing the written informed consent. Were included 27 participants in this study.

Data Collection Procedure

Participants who met the eligibility criteria were invited to respond to the questionnaires during the time they remained at the health unit (before or after the appointments) and to return the completed questionnaires directly to the researchers. When distributing the data collection instrument, in the temporal episodes in which the participants were in the health unit, the researchers obtained and clarified the informed consent, addressing topics such as the objectives of the study, the rights of the participants and the methodology of data collection. The informed consent form was collected immediately before the participants filled out the questionnaires, and the anonymous coding of the questionnaires was carried out. All participants gave their informed consent to participate in this study and anonymity was assured.

The non-repetition of participation was guaranteed (since the questionnaires were anonymous), through the confirmation of a non-existence of a previous administrative contact record during the period of data collection For all the people who participated, they were offered help in explaining the questions formulated in the instrument, as well as help filling out the form.

Measurements

Demographic Characteristics

Socio-demographic indicators assessed included age (date of birth), sex (female, male), birthplace (country of birth); educational level (up to the second cycle of primary education; up to the third cycle of primary education; high school; university education); employment status (working professionally; not working professionally); monthly income (≤500€; 501€−1,000€; 1,001€−1,500€; 1501€−2,000€).

Health Literacy Level

The validated version of Health Literacy Survey Portugal (ILS-PT) was used to measure migrant's health literacy. The questionnaire was developed by Professor Rita Espanha and her team (20). Permission to use the instrument was obtained from the authors. The ILS-PT consists of several modules. Module 1 (20), concerns the translation of the European Health Literacy Survey Questionnaire (HLS-EU-Q) (21)); it consists of 47 questions comprising four indexes. For its calculation, it is necessary to assign to each answer a certain classification, based on the difficulty of performing a certain health-related task (1- very difficult; 2- difficult; 3- easy; 4- very easy). The general health literacy index (comprising 47 items) GEN-HL (General Health Literacy Index) and three other indexes were calculated: the health care index (HC-HL—Health Care Literacy Index), with 16 items, the disease prevention index (DP-HL—Disease Prevention Literacy Index), with also 16 items and the health promotion index (HP-HL—Health Promotion Literacy Index), with 15 items (14). The indexes were calculated using a metric ranging from 0 to 50, where 0 represents the minimum of HL and 50 the maximum possible. The indexes were calculated according to the following formula: Index = (mean-1) × (50/3), where the mean refers to the mean of the answers of the items of each participant; value 1 is the minimum possible value of the mean and value 3 is the maximum value of the mean in the scale of difficulty in answering the questions asked (22).

In the case of the sample in question, the 27 questionnaires are valid, as they all have 100% response in the HLS items. The four indexes developed (GEN-HL, HP-HL, DP-HL and HP-HL) characterize the level of HL, according to the score obtained, as follows: from 0 to 25 points, corresponds to an inadequate level of HL; from 26 to 33 points, corresponds to a problematic HL level; from 34 to 42 points, corresponds to a HL sufficient level; and finally, from 43 to 50 points, corresponds to an excellent level of HL.

Statistical Analysis

Analysis was conducted by using a software Version 25.0 SPSS 25 (The Statistical Package for the Social Sciences software) (Version 25.0., SPSS Inc., Chicago, IL). The sample of this study was characterized in terms of their sociodemographic characteristics, as well as by the rating of the different dimensions of Health Literacy, gotten according to the responses obtained through the ILS-PT, namely in healthcare, disease prevention and health promotion indexes. In this research, only descriptive statistics were used. Descriptive statistics and frequencies were performed to describe the variables under evaluation.

RESULTS

Sociodemographic Characteristics

Table 1 shows the sociodemographic distribution of the participants. The average age of the participants was 31.6 years, with the predominant age group between 25 and 34 years of age (55.6%). The sample consisted of 85.2% women and 14.8% men. Regarding birthplace (and as birthplace, it is understood as the country of birth, as the ILS-PT refers), the most represented nationalities are the Brazil's people with 22.2%, Sao Tome's people with 18.5%, Angola's people with 18.5% and Guinea people with 14.8%. In terms of education level, degrees up to 2nd cycle (37%) and secondary education (33.3%) predominate, with higher education (18.5%) being in a minority. Regarding professional activity, 70.4% of the participants were professionally active and 29.6% were not active (which included participants with no professional activity, the unemployed, students or retired people). Finally, concerning to monthly income, about 59.3% of the participants have an income in their household which is

TABLE 1 | Sociodemographic characteristics.

Variables	***n* (%)**
Age (years)*	
18–24	3 (11.1%)
25–34	15 (55.6%)
35–54	9 (33.3%)
Sex	
Female	23 (85.2)
Male	4 (14.8)
Birthplace	
Angola	5 (18.5)
Guiné-Bissau	4 (14.8)
Cabo Verde	1 (3.7)
São Tomé e Príncipe	5 (18.5)
Guiné-Conacri	1 (3.7)
Brasil	6 (22.2)
Venezuela	1 (3.7)
Roménia	2 (7.4)
Serra Leoa	1 (3.7)
China	1 (3.7)
Education Level	10 (37)
up to the second cycle of primary education	3 (11.1)
up to the third cycle of primary education	9 (33.3)
High school	5 (18.5)
Higher education	
Employment status	
Working professionally	19 (70.4)
Not working professionally	8 (29.6)
Monthly income	
≤500€	8 (29.6)
501€–1,000€	16 (59.3)
1001€–1,500€	2 (7.4)
1501€–2,000€	1 (3.7)

* *The mean age of the participants is 31.6 years.*

equal to or <€1000 and about 29.6% of the participants have a monthly income equal to or less 500€.

Health Literacy Profile Assessment

Supplementary Table 2 shows the distribution of answers to the questions of module 1 by the matrix of the 12 sub-dimensions based on the HL Integrated Conceptual Model. Within the scope of the items related to health care (from question 1 to question 16), more than 50% of the participants refers that is "very difficult" or "difficult": (a) find information about symptoms of illness that concern them (59.3%); (b) find information about treatments of illnesses that concern them (63%); (c) knowing what to do in case of a medical emergency (51.9%); (c) understand the leaflets that come with the medications (85.2%); (d) understand what to do in a medical emergency (59.3%); (e) judge the advantages and disadvantages of treatment options (88.9%); (f) assess the need for a second medical opinion (96.3%). In the same health care index, more than 50% of the participants considered it to be "easy" or "very easy": (a) knowing where to get professional help when they are sick (88.9%); (b) understanding what their doctors tells them (55.6%), although the asymmetry is not significant against the opposite categories (44.4%); (c) understand the instructions from the doctor or pharmacist about taking a medication that was received (85.2%); (d) assess how the information given by your doctor applies to their clinical condition (55.6%); (e) assess whether information about diseases spread in the media is reliable (66.7%); (c) use doctor's information to decide about their illness (70.4%); (d) follow the instructions about the prescribed medication (92.6%); (e) call an ambulance in an emergency situation (55.6%); (f) follow the instructions of their doctor or pharmacist (92.6%).

When analyzing the items that refer to disease prevention (from question 17 to question 31), more than 50% of the participants reports that is "very difficult" or "difficult": (a) finding information on how to manage mental health problems such as stress or depression (96.3%); (b) find information about vaccines and medical exams they should have to do (88.9%); (c) judge when to go to the doctor for a check-up or a general health exam (81.5%); (d) evaluate which vaccines they may need (92.6%); (e) evaluate which health screenings they have to do (88.9%).

In the same index on disease prevention, more than 50% of the participants consider that is "easy" or "very easy": (a) finding information on how to manage unhealthy behaviors, such as smoking, lack of physical activity and excessive alcohol consumption (81.5 %); (b) find information on how to avoid or control situations such as overweight, high blood pressure and high cholesterol (88.9%); (c) understand health warnings about unhealthy behaviors such as smoking, low of physical activity and excessive alcohol consumption (92.6%); (d) understand the need of to be vaccinated (81.5%); (e) understand why they need to be seen by doctors in routine medical check-ups (63.0%); (e) assess how reliable health warnings are, such as smoking, lack of physical activity and excessive alcohol consumption (88.9%); (f) assess whether the information transmitted in the media about health risks is reliable (81.5%); (g) decide whether they should get the flu vaccine or not (59.3%); (h) deciding how to protect themselves from illnesses based on advice from family and friends (92.6%); (i) decide how to protect themselves from diseases based on information transmitted by the media (88.9%).

Finally, regarding health promotion, almost the total index of items were classified as "very difficult" or "difficult" (above 50%) by the participants, namely: (a) find information on activities that are beneficial to your mental well-being (92.6%); (b) find information about how their neighborhood can be more health-friendly (96.3%); (c) finding information about policy changes that can be addressed as health issues (96.3%); (d) find information about the efforts to promote their health at work (92.6%); (e) understand information on how to keep their mind healthy (92.6%); (f) assess how the place where they live affects their health and well-being (88.9%); (g) assess how the conditions of their home help them to stay healthy (66.7%); (h) assess which everyday behavior is related to their health (63.0%); (i) make decisions that improve their health (63.0%); (j) join a gym or a sport if they want to (81.5%); (k) change the living conditions that affect their health and well-being (92.6%); (l) participate in

TABLE 2 | Health literacy indexes.

Health literacy domain	Mean of HL index (SD)	Levels of health literacy (%)			
		Inadequate	Problematic	Sufficient	Excellent
General Health Literacy	21.2 (5.0)	85.2	11.1	3.7	0.0
Healthcare	25.3 (6.4)	55.6	37.0	7.4	0.0
Disease prevention	25.0 (4.2)	66.7	25.9	7.4	0.0
Health promotion	13.8 (7.0)	88.9	7.4	3.7	0.0

TABLE 3 | Health literacy indexes according to age, sex, and education level.

		Health literacy domain			
		General health literacy (GEN_HL)	Healthcare (HC_HL)	Disease prevention (DP_HL)	Health promotion (HP_HL)
		Mean	Mean	Mean	Mean
Age (years)	18–24	19.0	22.9	22.9	11.1
	25–34	20.5	24.9	24.5	12.3
	35–54	23.1	26.5	26.4	17.0
Sex	Male	26.5	31.3	27.5	21.3
	Female	20.3	24.2	24.5	12.4
Education level	Up to the second cycle of primary education	19.4	23.3	24.8	10.8
	Up to the third cycle of primary education	21.6	26.7	21.8	16.3
	High school	23.4	28.1	25.3	16.6
	Higher education	20.6	23.1	26.6	12.9

actions that improve health and well-being in their community (92.6%). In the same index the participants classified as "easy" or "very easy" the following items: (a) find information on healthy activities such as exercise, healthy food, and nutrition; (b) understanding health advice given by family and friends (96.3%); (c) understand information present in food packaging (59.3%); (d) understanding information in the media on to get healthier (100%).

Levels of Health Literacy

Table 2 shows the values for the General Health Literacy Index (GEN-HL), the Health Care Literacy Index (HC-HL); the Disease Prevention Literacy Index (DP-HL) and the Health Promotion Literacy Index (HP-HL). The results show that the participants have on average, an inadequate level of health literacy in general ($\overline{x} = 21.2$) (GEN-HL). Furthermore, 85.2% of the participants have an inadequate level health literacy; 11.1% have a problematic level and only 3.7% have a sufficient level. Regarding the Health Care Index (HC-HL), the participants maintain an average inadequate level of health care literacy ($\overline{x} = 25.3$). Concerning this index, about 55.6% of the participants have an inadequate level, 37% have a problematic level and 7.4% have a sufficient level. About the Disease Prevention Index, it is shown that the participants have an average inadequate level ($\overline{x} = 25.02$). About 66.7% of the participants have an inadequate level of health literacy in disease prevention; 25.9% have a problematic level and 7.4% have a sufficient level. Finally, in the Health Promotion Index (HP-HL) the participants have an average inadequate level of literacy related to disease prevention ($\overline{x} = 13.8$). The distribution of the participants in this index, shows that 88.9% have an inadequate level; 7.4% have a problematic level and 3.7% have a sufficient level.

Health Literacy Indexes According to age, sex, and Education Level

As shown in **Table 3**, when we perform a comparative analysis of the means of all indices in terms of age, we find that it is in the higher age classes, namely between 35 and 54 years of age, that the mean score of the indices of general health literacy ($\overline{x} = 23.\ 1$), health care-related health literacy ($\overline{x} = 26.5$), disease prevention ($\overline{x} = 26.4$) and health promotion ($\overline{x} = 17.0$) is highest. On the other hand, in relation to sex, we observe that men show higher mean values for general health literacy ($\overline{x} = 26.5$), health care literacy ($\overline{x} = 31.3$), health literacy related to disease prevention ($\overline{x}$

$= 27.5$) and health promotion ($\bar{x} = 12.4$) than women. In turn, with regard to educational level, the data show that migrants with secondary education have the highest mean values in the indices of general health literacy ($\bar{x} = 23.4$), health care-related health literacy ($\bar{x} = 28.1$), health literacy related to disease prevention ($\bar{x} = 25.3$), and health literacy related to health promotion ($\bar{x} = 16.6$).

DISCUSSION

The people in a migration situation, suffers from a higher vulnerability that is a result of the migration process, and that vulnerability it is increased by the low levels of health literacy. The evaluation of the health literacy indexes in our sample confirms this of which the respondents are examples. The results that we have achieved, confirm that, the level is inadequate in 85.2% participants of our study. In fact, the data from this study are in line with studies carried out in recent years which point to reduced levels of health literacy in the migrant population (23)., however, it is important to mention that, so far, there are few studies in Portugal on health literacy and migrant populations, and knowledge in this area is still limited and needs further studies (23). Given the reduced levels of health literacy of the migrant population, adverse events such as less optimized health behaviors, worse health status and limited access to health care can occur, leading to a situation of vulnerability, and health inequalities (24, 25).

Regarding health care, the results demonstrate that migrants have difficulties either in obtaining health information or in accessing health services, which translates into an inadequate level of health literacy in this index (56%). In fact, many adult migrants do not have access to appropriate health information, and often face difficulties in managing health issues. They are confronted to health staff with inadequate cultural skills and have difficulties managing and understanding the highly complex health systems (26). Additionally, access to health care can be hampered by different barriers, such as the inability to provide guidance in the health system, previous experiences in the health system, existing expectations regarding care, language and cultural barriers, financial resources and beliefs associated with health (27).

On the other hand, the study data demonstrate the existence of inadequate levels, regarding to literacy related to disease prevention and health promotion. In fact, a recent study developed in Europe in the context of a systematic review of the literature on the health literacy of the migrant population in Europe, demonstrated that the reduced level of health literacy among migrants affects the self-management of chronic illness; it provides low levels of adherence to therapeutic interventions resulting from language barriers in understanding the verbal and written information produced by health professionals (teaching, reading information leaflets and drug labels); and increases the difficulties in accessing health services (24).

In this line of ideas, the World Health Organization recognizes as barriers to accessing health care: nationality and legal status in view of remaining in the country, linguistic and cultural differences, administrative barriers, the inability of the health system to adjust in the different countries and the lack of information about health services (28).

In addition, the data show that the highest mean scores on the General Health Literacy, Health Literacy and Health Care, Health Literacy and Disease Prevention, and Health Literacy and Health Promotion indices are presented by male migrants and by migrants with secondary level education. Indeed, it has been identified in some studies that there are differences between men and women in the level of health literacy, with men tending to have higher levels of health literacy than women (29). In fact, it is important to consider future studies regarding the differences between men's and women's health literacy levels. Indeed, the identification of the role of migrant women as family leaders in the health and disease process is essential and should be valued, as women may be the essential link in the dissemination of health information and modification of health management related habits in their families and communities (29, 30). On the other hand, women are subject, throughout their life cycle, to health transition processes that make them more vulnerable, such as issues concerning reproductive and maternal health, for example, limitations in access to contraception, cultural aspects that influence the management of sexual and reproductive health, the barriers in certain regions of the globe to a safe childbirth and pospartum (30), therefore improved knowledge is needed on how these experiences influence or impact Health Literacy in women.

In our study the data obtained show that migrants with a secondary education level have better Health Literacy than migrants with higher education, however, it is important to note that such an observation may be related to the small sample size, so its interpretation should be careful.

Indeed, regarding educational level, some studies point out its significant impact on the level of Health Literacy in Migrant Populations (25, 31), and higher educational levels may be more associated with higher levels of Health Literacy, and therefore in the way people manage their health (31). On the other hand, it is important for future research to reflect on aspects regarding the methodology and organization of the education system in the migrants' countries of origin, as well as the classification system inherent to the educational level, and how these factors impact the Health Literacy levels of migrant populations (25, 31). Additionally, the mentioned aspects, together with the lack of a culturally congruent care model, which concerns an understanding of the social importance and cultural influence on people's health beliefs and behaviors, based on an awareness by the healthcare professionals, influences the existence of equity in access to health care of migrant populations (25, 32).

Having evidence available on the result of the intervention of nurses in the community, combined with the evaluation of the levels of health literacy of the target populations, will allow interventions to be personalized, which will allow us to provide more efficient care to each community, with higher quality, and based on a model of intercultural care that enhances equity in health and the defense of human rights of migrant populations (32).

The World Health Organization also recognized that the Health Literacy is a social determinant of health, a critical one (33). The increasing abilities obtained by higher levels of health literacy, can be achieved by supporting the migrant communities to better understand the health environment that they are inserted. The community specialized nurse can be the facilitating agent of the change.

It is essential to foster and strengthen research on health literacy in the field of nursing, as nurses are key agents in health promotion and health education (34). Community participation is a fundamental strategy, motivated by the empowerment and counseling of the migrant population, to motivate a change in behavior and in paradigm, from the passive group to the active group responsible for decisions that affect their health status.

CONCLUSIONS

This study with an exploratory approach is innovative and highlights the assessment of the level of health literacy in migrants attending primary health care in Portugal. The study showed that many participants have inadequate levels of health literacy in general, and inadequate levels of health literacy related to health care, disease prevention and health promotion. On the other hand, it highlights the importance of promoting equity in access to health information, contributing to the reduction of inequity in access to health by migrant populations.

The present study has some limitations inherent to the small sample size, so that the reading of the results obtained will have to be careful contextualized and not generalized for the migrant population in general. On the other hand, the limited time for data collection contributed to limit the recruitment of participants. More studies need to be conducted to better understand both the health literacy levels of migrant populations, and the factors associated with different levels of literacy as well, and their impact on health equity.

However, our study, although limited by the small number of participants, shows that there are some differences in the mean scores of the health literacy indices under analysis, for example, regarding age, sex and educational level. It is therefore urgent to identify the most vulnerable groups among migrant populations in terms of health literacy, namely women and those with a lower level of education, in order to design and implement actions that correspond to the different needs identified.

Based on the knowledge of the health literacy level of migrant populations, the implementation of interventions culturally adapted to the context and knowledge/experience of migrant populations, will strengthen health equity for migrant populations and is essential to reduce health inequalities. New studies on Health Literacy of migrant populations are needed to know how this populations access, understand, and uses health information to be able to increase the self-manage of their health. In this context, the present study has a higher relevance for the clinical practice of nurses regarding the strategies mobilized to increase health literacy in migrant populations.

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 Regional Health Administration of Lisbon and Vale do Tejo (Process 052/CES/INV/2019). The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

PM and AC conception and design of the study and performed and interpreted the statistical analysis. PM responsible for data collection. AM drafted the manuscript and revised it. All authors read and approved the final manuscript.

FUNDING

The research was funded by the CIDNUR.

ACKNOWLEDGMENTS

The authors would like to thank all migrants for taking part in the study, as well as the team of Professor Sørensen and Professor Rita Espanha for making the instruments available.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2021.798222/full#supplementary-material

REFERENCES

1. International Organization for Migration. *Global Migration Indicators 2018- Insights from the Global Migration*. Berlin. (2018). Available online at: www.migrationdataportal.org (accessed December 2, 2019).
2. International Organization for Migration. *Who is a migrant?* (2019). Available online at: https://www.iom.int/who-is-a-migrant (accessed December 2, 2019).
3. Serviço de Estrangeiros e Fronteiras. *Relatório de Imigração, Fronteiras e Asilo 2018*. Oeiras. (2019).
4. Kennedy S, Kidd MP, McDonald JT, Biddle N. The healthy immigrant effect: patterns and evidence from four countries. *J Int Migrat Integrat.* (2015) 16:317–32. doi: 10.1007/S12134-014-0340-X
5. Dias S, Gama A, Martins MO. Health status and preventative behaviors of immigrants by gender and origin: a portuguese cross-sectional study. *Nurs Health Sci.* (2013) 15:309–17.doi: 10.1111/nhs.12032
6. Moniz AM. *Healthy Immigrant Effect em Portugal: Estudo Sobre os Imigrantes Extra UE Presentes no Inquérito Nacional de Saúde 2014.* Master Thesis UNL-ENSP (2018).

7. Matos I, Alarcão V, Lopes E, Oiko C, Carreira M. Estudo SAIMI—Saúde e Acesso aos Serviços de Saúde dos Imigrantes Do Subcontinente Indiano em Lisboa: Que Recomendações Para Cuidados de Saúde Equitativos e Culturalmente Adaptados ? *Acta Médica Portuguesa.* (2015) 15:64–77.
8. United Nations. *New York Declaration for Refugees and Migrants*. New York, NY: United Nations General Assembly (2016).
9. Gushulak B. Health, health systems and global health- thematic discussion paper. *Int Organiz Migrat*. (2017) 18:987.doi: 10.1186/s12889-018-5932-5
10. Yadee J, Bangpan M, Thavorn K. Assessing evidence of interventions. *Int J Equity Health.* (2019) 19:970. doi: 10.1186/S12939-019-0970-X
11. Hahn K, Steinhauser J, Goetz K. Equity in health care: a qualitative study with refugees, health care professionals, and administrators in one region in Germany. *BioMed Res Int.* (2020) 20:7389. doi: 10.1155/2020/4647389
12. Direção-Geral da S. *Plano Nacional de Saúde 2012-2016: Revisão e Extensão a 2020. Lisboa.* (2015) Available online at: https://pns.dgs.pt/pns-Revisao-E-Extensao-a-2020/ (accessed February 12, 2019).
13. Alto Comissariado Para as Migrações. *Plano Estratégico Para as Migrações.* Lisboa: ACM. (2015).
14. Fonseca ML, Silva S. *Saúde e Imigração: Utentes e Serviços na Área de Influência Do Centro de Saúde da Graça*. Lisboa: Alto Comissariado Para a Imigração e Diálogo Intercultural. (2010).
15. Entidade Reguladora da Saúde. *Acesso a Cuidados de Saúde por Imigrantes*. Porto: Entidade Reguladora da Saúde. (2015).
16. World Health Organization. *Health Promotion Glossary*. Geneva: WHO (1998).
17. Sørensen K, Van den Broucke S, Fullam J, Doyle G, Pelikan J, Slonska Z, Brand H. Health literacy and public health: a systematic review and integration of definitions and models. *BMC Public Health.* (2012) 12:1–13. doi: 10.1186/1471-2458-12-80
18. World Health Organization. *Migration and Health: Key Issues—Public Health Aspects of Migration in Europe*. Regional Office for Europe. Geneva: WHO. (2019).
19. Direção-Geral da Saúde. *Plano Nacional de Saúde 2012-2016: Versão Resumo*. Lisboa. 2013. Available online at: https://pns.dgs.pt/pns-2012-2016 (accessed December 2, 2019).
20. Espanha R, Ávila P. Health Literacy Survey Portugal: A Contribution for the Knowledge on Health and Communications. *Procedia Computer Science.* (2016) 100:1033–1041.doi: 10.1016/j.Procs.2016.09.277
21. Sørensen K, Van den Broucke S, Fullam J, Doyle G, Pelikan J, Slonska Z, Brand H. Measuring health literacy in populations: illuminating the design and development process of the european health literacy survey questionnaire (HLS-EU-Q). *BMC Public Health.* (2013) 13:1–10. doi: 10.1186/1471-2458-13-948
22. HLS-EU Consortium. *Comparative Report on Health Literacy in Eigth EU Member States: The European Health Literacy Project 2009-2012*. Maastricht: HLS-EU Consortium. (2012).
23. Ackerman Rau S, Sakarya S, Abel T. When to see a doctor for common health problems: distribution patterns of functional health literacy across migrant populations in Switzerland. *Int J Public Health.* (2014) 59:967–74.doi: 10.1007/S00038-014-0583-5
24. Ward M, Kristiansen M, Sørensen K. Migrant health literacy in the European Union: a systematic literature review. *Health Educ J.* (2019) 78:81–95. doi: 10.1177/0017896918792700
25. Wångdahl J, Lytsy P, Mårtensson l, Westerling R. Health literacy among refugees in sweden—a cross-sectional study. *BMC Public Health.* (2014) 14:1030. doi: 10.1186/1471-2458-14-1030
26. Soto-Mas F, Schmitt CL, Jacobson HE, Myers OB. A cardiovascular health intervention for spanish speakers: the health literacy and ESL curriculum. *J Commun Health.* (2018) 43:717–724. doi: 10.1007/S10900-018-0475-3
27. O'Donnell CA, Burns N, Mair FS, Dowrick C, Clissmann C, van den Muijsenbergh M, et al. Reducing the health care burden for marginalized migrants: the potential role for primary care in Europe. *Health Policy.* (2016) 120:495–508. doi: 10.1016/j.Healthpol.2016.03.012
28. World Health Organization. *Promoting the Health of Refugees and Migrants. Draft Global Action Plan 2019-2023*. Geneva. (2019). Available online at: https://www.who.int/Publications/I/Item/Promoting-the-Health-of-Refugees-and-Migrants-Draft-Global-Action-Plan-201\penalty-\@M9-2023 (accessed June 4, 2019).
29. Aldin A, Chakraverty D, Baumeister A, Monsef I, Noyes J, Jakob T, et al. Gender differences in health literacy of migrants: a synthesis of qualitative evidence. *Cochrane Database of Systematic Rev.* (2019) 19:330. doi: 10.1002/14651858.CD013302
30. Gele AA, Pettersen KS, Torheim LE, Kumar B. Health literacy: the missing link in improving the health of Somali immigrant women in Oslo. *BMC Public Health.* (2016) 16:1134. doi: 10.1186/s12889-016-3790-6
31. Park S, Lee H, Kang M. Factors affecting health literacy among immigrants—systematic review. *Euro J Public Health. (*2018).doi: 10.1093/Eurpub/Cky214.283
32. World Health Organization. *Migration and Health: Key Issues. Public Health Aspects of Migration in Europe.* Regional Office for Europe. Geneva. (2019). Available online at: https://www.euro.who.int/en/health-topics/health-determinants/migration-and-health/migration-and-health-in-the-european-region/migration-and-health-key-issues (accessed April 6, 2019).
33. World Health Organization. *Promoting Health in the SDGs. Report on the 9th Global Conference for Health Promotion, Shanghai, China, 21–24 November 2016: all for Health, Health for all. Geneva*. (2017). Available Online At: https://www.who.int/Publications/I/Item/Promoting-Health-in-the-Sdgs (accessed December 2, 2019).
34. Fernandez-Gutierrez M, Bas-Sarmiento P, Albar-Marin MJ, Paloma-Castro O, Romero-Sanchez JM. Health literacy interventions for immigrant populations: a systematic review. *Int Nursing Rev.* (2018) 65:54–64.doi: 10.1111/inr.12373

Conflict of Interest: 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.

The reviewer KS declared a shared affiliation with one of the authors AC to the handling editor at time of review.

Publisher's Note: All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

Copyright © 2022 Medina, Maia and Costa. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

frontiers
in Public Health

ORIGINAL RESEARCH
published: 29 March 2022
doi: 10.3389/fpubh.2022.834172

Connections and Biases in Health Equity and Culture Research: A Semantic Network Analysis

Mireya Martínez-García[1], *José Manuel Villegas Camacho*[2,3] and *Enrique Hernández-Lemus*[4,5]*

[1] Department of Immunology, National Institute of Cardiology Ignacio Chávez, Mexico City, Mexico, [2] Clinical Research Division, National Institute of Cardiology Ignacio Chávez, Mexico City, Mexico, [3] Social Relations Department, Universidad Autónoma Metropolitana, Mexico City, Mexico, [4] Computational Genomics Division, National Institute of Genomic Medicine, Mexico City, Mexico, [5] Center for Complexity Sciences, Universidad Nacional Autónoma de México, Mexico City, Mexico

Health equity is a rather complex issue. Social context and economical disparities, are known to be determining factors. Cultural and educational constrains however, are also important contributors to the establishment and development of health inequities. As an important starting point for a comprehensive discussion, a detailed analysis of the literature corpus is thus desirable: we need to recognize what has been done, under what circumstances, even what possible sources of bias exist in our current discussion on this relevant issue. By finding these trends and biases we will be better equipped to modulate them and find avenues that may lead us to a more integrated view of health inequity, potentially enhancing our capabilities to intervene to ameliorate it. In this study, we characterized at a large scale, the social and cultural determinants most frequently reported in current global research of health inequity and the interrelationships among them in different populations under diverse contexts. We used a data/literature mining approach to the current literature followed by a semantic network analysis of the interrelationships discovered. The analyzed structured corpus consisted in circa 950 articles categorized by means of the *Medical Subheadings* (MeSH) content-descriptor from 2014 to 2021. Further analyses involved systematic searches in the LILACS and DOAJ databases, as additional sources. The use of data analytics techniques allowed us to find a number of non-trivial connections, pointed out to existing biases and under-represented issues and let us discuss what are the most relevant concepts that are (and are not) being discussed in the context of Health Equity and Culture.

Keywords: health equity, culture, education, semantic networks, ontology

OPEN ACCESS

Edited by:
Violeta Alarcão,
University Institute of Lisbon, Portugal

Reviewed by:
Alessandro Stievano,
University of Rome Tor Vergata, Italy
Miodraga Stefanovska-Petkovska,
Universidade de Lisboa, Portugal

***Correspondence:**
Enrique Hernández-Lemus
ehernandez@inmegen.gob.mx

Specialty section:
This article was submitted to
Life-Course Epidemiology and Social
Inequalities in Health,
a section of the journal
Frontiers in Public Health

Received: *13 December 2022*
Accepted: *07 March 2022*
Published: *29 March 2022*

Citation:
Martínez-García M, Villegas Camacho JM and Hernández-Lemus E (2022) Connections and Biases in Health Equity and Culture Research: A Semantic Network Analysis. Front. Public Health 10:834172. doi: 10.3389/fpubh.2022.834172

1. INTRODUCTION

A first step toward an integrated understanding of social determinants and cultural issues contributing to determine the health inequity status and related issues, consists, not only in *enlisting* them, but also in sketching the interplay that these features may have among themselves to give rise to the observed impact of social constraints upon population-level health conditions (1). Social and cultural factors that are related to the inequities in health should be identified through comprehensive research and analysis (2). However, the future of health equity assessment also depends on our continued innovation in developing methods to monitor them and intervene from an integral, inclusive perspective (3–6).

Identifying social and cultural issues, aiming to determine the health inequity status across population subgroups, it has been widely discussed in recent years (7–10). Since the 1980s, there has been a growing call for nations across the globe to address health inequities (3). The past several years have been characterized by an increasing focus on solutions (11–13). Many initiatives scopes include identifying, monitoring, promoting and implementing frameworks to approach health inequities and social determinants of health (SDH) (14–19). In 2013 the WHO started a project named *Equity-oriented analysis of linkages between health and other sectors* (EQuAL) in order to identify possible approaches to the monitoring of equitable progress toward universal health coverage, centered on intersectoral barriers and identifying specific social determinants affecting health (14).

The study of health inequities itself is, however, not devoid of challenges and constraints (20). A number of factors, ranging from the social and economical conditions, to the cultural and educational background of the populations contribute to shape the panorama of health inequities, every one of these, actually a complex issue; hence there is the need for a research framework that allow to study these issues together (21). Such framework must also aim to be free from biases and allow an assessment of the matters in the most objective way possible. Alas, this is easier said than done. Along these lines, the present study intends to help us to characterize, at a large scale, the social and cultural determinants most frequently reported in current global research of health inequity and the interrelationships among them in different population and diverse contexts. To address these goals, we aim to take advantage of the vast corpus of literature already published in the PubMed/MEDLINE and other databases and investigate the research trend by applying network analysis to explore the relationships among their keywords so-called Medical Subject Headings (MeSH) remaining as unbiased as possible while doing this (22).

PubMed is the largest database of life sciences and biomedical literature in the world and is provided by the National Center for Biotechnology Information (NCBI) of the United States of America (23). PubMed's search interface implements at least five recommended search elements (reproducibility of search results, search results can be exported in full, search history, search string builder and forward citation search). PubMed/MEDLINE is indeed one of the top recommended primary sources for literature searches of peer-reviewed research in the biomedical sciences, as it possesses an extensively curated catalog (24). MEDLINE database is indexed by using MeSH terms, which are a collection of selected words or phrases that are able to represent specific concepts and form a fundamental part of the representation of knowledge (25). The MeSH dictionary is actually an *ontology*, its structure formalizes the name and definition of entities and their properties in a taxonomy-like manner able to capture conceptual interrelationships (26, 27).

Occasionally the MeSH classification includes the same term twice. One of these instances is preceded by an asterisk (*). Those entries allude to a MeSH term consider a major main topic of an article or a class. In the context of this study, such terms were treated as separate entities and analyzed accordingly.

The MeSH ontology has gained further relevance since recently, a number of researchers are using automated mining of scientific literature databases and network analysis as a novel methodology to know how the MeSH terms are related to each other and how their connectivity patterns helps better understand them—in terms of finding research ideas and raising or restating some hypotheses, and in summarizing a large amount of information (28). This method has also been useful for finding emergent keywords to further investigate in research areas such as immunotherapy and cancer (22, 29, 30), metabolomics (31), individual cognitive map or semantic networks (32), predictive, preventive and personalized medicine (33), biomedical sciences (34, 35), genetic (36), and other areas of health research (37–41). This will be also the approach we will follow here.

It is worth stressing that the present work is mostly of a descriptive nature. Our aim is to present the state of affairs regarding the scholarly discussion on these quite relevant subjects, to serve as a starting point for deeper analyses. In this regard, documents with the systematic searches, tables with the relationships forming the semantic networks and tables describing the topological data analytics of such networks (all of these included in the **Supplementary Materials**: **Supplementary Tables 1–6** contain network statistics for all 6 networks discussed. **Supplementary Documents 7–12** contain edge-list representations for all 6 networks discussed) will provide the readers interested in further analysis with exploration tools to navigate through the relatively extensive literature corpora on these matters.

2. METHODS

2.1. Study Design

Our methodological framework is founded on a semantic network perspective (42). Meaningful relationships among social and cultural determinants are quite difficult to unveil or highlight by resorting to traditional systematic reviews and meta-analyses that usually present the information fragmented, or at most, integrated according with the subjective appreciation of the reviewer (43, 44). Semantic network approaches to analyzing the literature have been used recently (23, 45, 46), some of them resort to computational mining of the publication databases and archives, ontology-based add context to theme-driven, systematic surveys of the literature (47–49). It is relevant to highlight that the curatorial procedures followed in this work are based on systematic and (whenever possible) objective criteria, even if it was not a completely computational curation but a hybrid approach. The methodological approach is summarized as follows (see **Figure 1**):

2.2. Stage I: Establishing the Research Questions

In recent decades there has been a growing body of evidence on equity and culture in health. Due to the complexity of

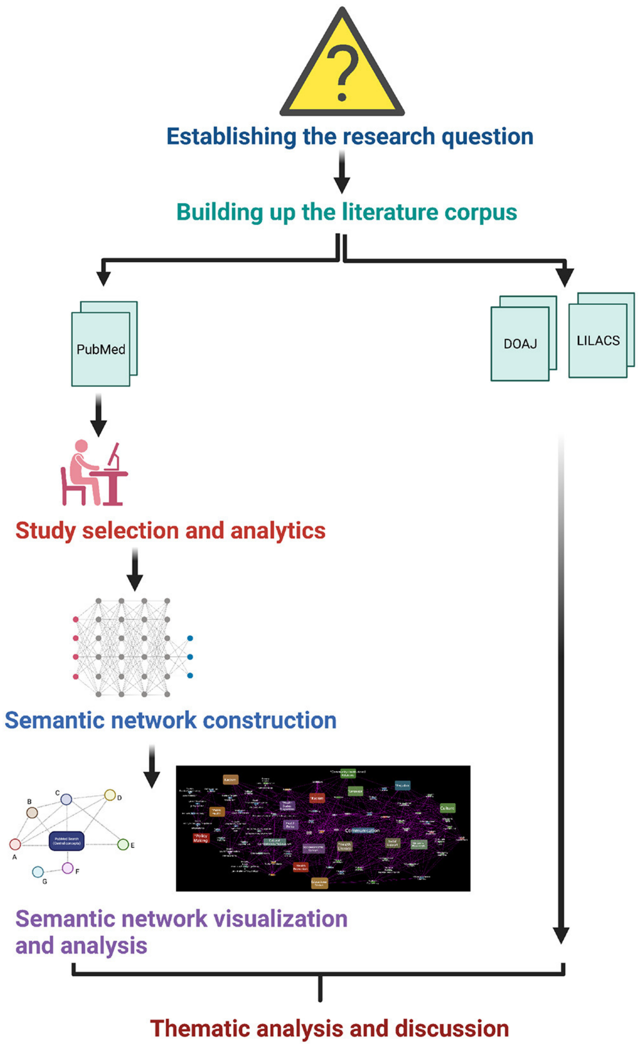


FIGURE 1 | Methodological approach followed in this study. Figure created with BioRender.com.

each of these entities, knowledge has been accumulated in a set of seemingly disparate concepts. Under these conditions, translating knowledge into practice and improving healthcare will require a much greater effort.

In this regard, delimiting a research question in complex, multi-faceted issues such as the intricate relationships between health equity, culture and trust in the context of providing proper healthcare, that is healthcare that takes into account the situation, SDH, education and probable vulnerabilities of the populations is not an easy task. We have decided to face this issue by surveying and analyzing six different frameworks contextualized as automated literature searches used to infer semantic networks. These concepts are expected to be related and even overlapping. We believe, however, that such elections reflect somehow general aspects of this complex phenomenon in a relatively simple form.

We analyzed what are the social and cultural determinants most frequently reported in current global research of health inequity as well as the interrelationships among them. In order to delimit the scope of our work, we have chosen to focus on different aspects, aiming to present a broader (yet admittedly blurry) vision of these complex phenomena. Health equity is particularly challenged in the case of *vulnerable populations* and specially influenced by *social determinants of health*. Hence, we decided to include these two concepts within our research scope. *Culture* and *Trust* are key elements to establish human relationships that may help abridging the gap between health practitioners and healthcare users (patients, families, etc.), hence these two concepts were also considered. Since we think that a good starting point to enhance trust and establishing a proper culture is education, *health literacy* and *education* complemented the concepts we decided to analyze in this work.

In brief, in this project we have decided to investigate on **two broad research questions**:

1. What are the most frequently reported *concepts* on current global research of health equity and inequity, as represented by their associated MeSH identifiers?
2. What are the *relationships* of these concepts with issues such as the social determinants of health, vulnerable populations, culture, trust, literacy, and education in different populations and diverse contexts as captured by the published literature?

Further details on these research questions will be provided in Sections 2.6 and 3.1.

2.3. Stage II: Building Up a Literature Corpus

We assembled a preliminary corpus by mining the articles related to social and cultural determinants and health equity as denoted by corresponding MeSH classifiers. An automated search of the PubMed/MEDLINE database was conducted on November 16, 2021. For the purposes of this study, we used English words, Boolean AND—operator, exact phrase, and parentheses in order to group individual concepts and link them logically. Although with an individual search for a single MeSH term the results are more numerous, it is also true that a large number of documents are generated that may be related to less specific topics. After searching the databases, the documents found were imported into a data frame and duplicates were removed.

The selection criteria applied to the recovered PubMed's documents were the following:

2.4. Inclusion Criteria

- That each bibliographic record contain at least one MeSH term to establish the network connection between each document.
- The bibliographic record could be from any year of publication.
- The bibliographic record could be from any country of affiliation.

2.5. Exclusion Criteria

- The bibliographic record does not contain a title.
- The content of the bibliographic record does not coincide with the relevance of the problem under study.

We also conducted searches in the Virtual Health Library/LILACS database (https:lilacs.bvsalud.orgen) and in the Directory of Open Access Journals, DOAJ (https:doaj.org), using the same linguistic cues strategy. LILACS is a database maintained by the Latin American and Caribbean Center on Health Sciences Information (50). It includes bibliographic information from articles that have been published in a set of scientific and medical journals of the region, and that are often not covered by MEDLINE. Similar to MEDLINE, LILACS uses controlled vocabulary in indexing to ensure accurate retrieval of bibliographic references (51). DOAJ is a website that hosts a community-curated list of open access journals. DOAJ is useful as a direct search for scholarly journals across all academic disciplines. Unfortunately, there is no option to export metadata from a search (52).

2.6. Stage III: Study Selection and Analytics

We performed a curation of the extracted text corpus using both manual and bibliometric automated techniques (26). MEDLINE search results were saved into a plain-text `Mongodb` database document, then a computational literature mining procedure was performed using `Python` pickles to extract the information into either a corpus document or (as we will see in the upcoming Stage IV) to a network-structured file with the `NetworkX` Python library. The computational details of the mining strategy are sketched at the associated `GitHub` repository (https://github.com/CSB-IG/bibliometrics).

2.7. Stage IV: Data Visualization

Once we had a curated corpus, we built semantic networks (using co-occurrence of MeSH terms as links) and performed topological analyses of such networks to find associations between the different concepts (see **Figure 2**). The connectivity maps were built, so that sources and target nodes are the terms that identified the articles in corpus and a link between these nodes was drawn if two articles shared additional terms, the more terms shared, the stronger the link and hence the closer the connection of these articles were assumed. The IDs in the network construction were the PMID's of each publication. Once we have a structured corpus, -network extraction was performed with the Python code included in https://github.com/CSB-IG/bibliometrics/blob/master/mesh_network_from_medline.py, and network analysis with Python's `NetworkX` library(53) and `Cytoscape` version 2.8 (54) with the `NetworkAnalyzer` plugin (55). Visualization was performed using `Cytoscape` (56).

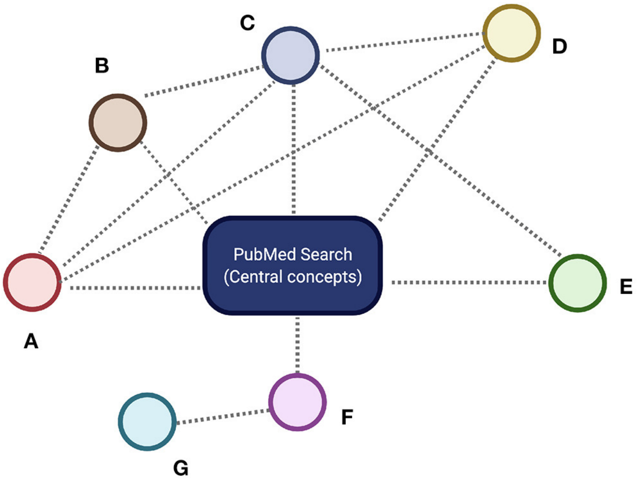


FIGURE 2 | Simplified conceptual representation of a Semantic Network. The central concept or concepts (blue rectangle) are given by the main PubMed search criteria. All the articles fulfilling these search criteria are supplemented with MeSH identifiers corresponding to the different concepts. These concepts are represented by colored circles labeled **(A–G)**. Whenever two different concepts appear in the same publication in the corpus, a semantic relationship is established between them. These semantic relationships are represented by dashed lines. We can see that there are some nodes-concepts with a relatively large number of semantic connections (here nodes **A–D**) whereas others are less connected (nodes **E–G**). Well-connected nodes are deemed to be central to the concepts under discussion and are named *core* nodes in the network science terminology, whereas scarcely connected nodes are called the *periphery*. Here **(C)** is a core concept, whereas **(G)** is a peripheral concept. The hierarchy of connections of the nodes in the semantic network determines the relevance of the related concepts. Figure created with BioRender.com

A first step toward the understanding of the web of interrelationships among items connected on a network is the determination of the network's local and global connectivity patterns (57). Such *topological features* as the individual and the global number of connections (called the *degree*), how are these connections assigned to the different nodes (the *degree distribution*), how important are certain nodes in the networks (called the *centrality measures*), etc. are the ones that will be used to discuss the relative importance and interplay of the different features related to our research (42). Plot visualization was also implemented to depict the main countries and dates from publications. All related source code for general text-processing may be found at https://github.com/CSB-IG/literature/tree/master/text_processing. The specific code for this work is found at https://github.com/CSB-IG/bibliometrics. Country and year mining were performed with custom-made Python scripts (articles_by_country.py and articles_by_year.py, respectively) available at https://github.com/CSB-IG/bibliometrics.

2.8. Stage V: Selecting, Summarizing, and Reporting the Results

As previously presented (see Section 2.2), the review questions deal with (1) **what are the *concepts* (as represented by their associated MeSH identifiers) most frequently reported in current global research of health equity and inequity** and (2) **what are their *interrelationships* with issues such as social determinants of health, vulnerable populations, culture, trust, literacy and education in different populations and diverse contexts, as presented in the published literature** as indexed in Pubmed and other health and biomedicine databases.

It is worth noticing that by building a semantic network based on a manually curated and annotated ontology (as given by the MeSH terms) on top of a comprehensive but not exhaustive database (PubMed), we are indeed introducing important assumptions. Such assumptions need to be considered as a part of our theoretical reference framework so that, all conclusions derived from this study are contingent on the validity of these assumptions (for further details on the constraints and limitations of this study see Section 4.3).

The most relevant assumptions must be summarized as follows:

1. Since only published documents indexed in the PubMed database are being retrieved for the main semantic network analysis, any contribution not indexed there (for whatever reasons) is considered **outside of our semantic universe**. This is a relevant issue since some health, biomedical or social sources, in particular in developing countries, are not indexed in PubMed. To leverage this assumption, further systematic searches were conducted in the LILACS and DOAJ databases.
2. The *concepts* here are considered based on the ontology given by MeSH classifiers. Concepts not defined as MeSH terms cannot be interpreted directly. We are aware that the MeSH ontology introduces representational biases and that other ontology used may give rise to different semantic networks.
3. All PubMed articles are manually curated and annotated with *representative* MeSH identifiers. Our computational approach relies on these annotations. Hence, if a given article was not *labeled* with a certain MeSH term, we will not count the related concept as discussed in such article, even if it is indeed touched upon.

We believe that these assumptions, still provide a general-enough framework to establish the basis for useful research, though more detailed analysis must be done to pursue a deeper understanding in more specific issues. Some of these limitations are further explored in the Section 4, in connection with other sources considered (LILACS and DOAJ).

2.8.1. Thematic Analysis

We concentrated the literature corpus from the three databases reviewed. A thematic analysis of them was carried out using Atlas-ti software version 8.4.5 to identify information patterns, thus delimiting or tagging portions of a certain pre-defined category. It has been suggested that thematic analysis, is a flexible and useful research tool for identifying, analyzing and reporting patterns within data (58, 59). The theoretical approach for the analysis based on three main themes was reinforced when the literature corpus was characterized by specific codes. For the purpose of this research, the methodological strategy suggested by Terry et al. (60).

The first step, along these lines, was to organize and generate categories in the data, then analytical units were selected to assign codes and relationships to them. Subsequently, the emerging codes were contrasted with the thematic categories previously established in each of the semantic networks. Finally, three subnetworks were extracted from each of the main networks, to construct a theoretical discussion and visualize outstanding patterns of connection between various key terms. The main findings are presented in narrative form, including figures and tables in the following section.

3. RESULTS

3.1. Stage I: Establishing the Research Question

As stated in Sections 2.2 and 2.6, the research questions guided us to investigate upon the conceptual relations between the following issues: (i) Health equity and Vulnerable Populations, (ii) Health equity and Social determinants of health, (iii) Health equity and Culture, (iv) Health equity and Trust, (v) Health equity and Health literacy, (vi) Health equity and Education.

3.2. Stage II: Building Up a Literature Corpus

Our automated PubMed/MEDLINE search located 950 documents. As expected, most of the articles were published in English (934/950). As previously noticed associated searches on the LILACS and DOAJ databases are also presented an used later in complementary analyses. These articles were distributed as shown in **Table 1**.

Interestingly for such related frameworks only 235 out of 950 documents (24.7 %) were overlapped between the different searches.

3.3. Stage III: Study Selection and Analytics

Semantic networks were constructed from the data mining of the different sub-corpora (corresponding to the different literature surveys). Some descriptive results for each of these surveys will be presented next.

We used Atlas-ti to perform thematic analysis to identify patterns of meaning across searches. Three themes emerged from the analysis: (1) identify social and cultural determinants of health inequity, (2) targeted populations and (3) Modalities of social and cultural response in various contexts (scientific, academic, political, governmental, among others).

Additional keywords to further investigate emerged from the analysis: SARS-CoV-2, Professional-patient relations, Patient acceptance of Health Care, Health Promotion, Cultural competency, Social support, Health knowledge, Attitudes and practice, Health communication and Communication barriers. These issues are considered in detail in the Section 4 (see Section 4.2).

3.3.1. Health Equity and Vulnerable Populations Network

This network (see **Figure 3**) included 551 nodes (MeSH terms or *concepts*, the basic semantic units) and 6,010 edges, corresponding to the semantic relationships (k) between the nodes-concepts (see Methods). The more connected term, as expected, was *Human* with 550 semantic relationships in this network. This will be the case for all of the studies considered since all human health research in PubMed is labeled with this term. *Vulnerable populations* and *Health equity* (on their two forms), being the basis of our search were also among the most central concepts with 365 (*Vulnerable Populations*), 294 (**Health Equity*), 180 (**Vulnerable Populations*) and 168 (*Health Equity*) semantic relationships, respectively. Aside from demographic classifiers –*Female* ($k = 163$), *Male* ($k = 133$), *United States*

TABLE 1 | Search results for the different databases analyzed in this work.

Database	Results	Period
PubMed (Medline)		
("Health equity" [MeSH Terms]) AND ("Vulnerable Populations" [MeSH Terms])	108	2014–2021
("Health equity" [MeSH Terms]) AND ("Social determinants of health" [MeSH Terms])	254	2015–2021
("Health equity" [MeSH Terms]) AND ("Culture" [MeSH Terms])	127	2015–2021
("Health equity" [MeSH Terms]) AND ("Trust" [MeSH Terms])	14	2015–2021
("Health equity" [MeSH Terms]) AND ("Health literacy" [MeSH Terms])	27	2015–2021
("Health equity" [MeSH Terms]) AND ("Education" [MeSH Terms])	420	2014–2021
Virtual Health Library (LILACS)		
"Health equity" AND "Vulnerable Populations"	19	2008–2021
"Health equity" AND "Social determinants of health"	57	2005–2021
"Health equity" AND "Culture"	27	2003–2021
"Health equity" AND "Trust"	4	2008–2019
"Health equity" AND "Health literacy"	2	2011–2021
"Health equity" AND "Education"	135	1992–2021
DOAJ		
"Health equity" AND "Vulnerable Populations"	93	2004–2021
"Health equity" AND "Social determinants of health"	304	2007–2021
"Health equity" AND "Culture"	249	2000–2021
"Health equity" AND "Trust"	289	2006–2021
"Health equity" AND "Health literacy"	196	2004–2021
"Health equity" AND "Education"	1,817	1995–2021

($k = 123$) and others that, as in the case of *Human* are standard or de facto MeSH classifiers in human health research– *SARS-CoV-2* emerged as an important concept in the discussion with 120 semantic relationships in the network. This is, of course, consistent with the ongoing pandemic, but also reflects the fact that this pandemic has evidenced a number of health disparity issues in vulnerable populations (61, 62).

The following relevant concepts in the scholarly literature discussion on Health Equity and Vulnerable populations point out to known issues. Concepts such as *Middle Aged* ($k = 118$), *Poverty* ($k = 117$), *Healthcare disparities* ($k = 104$), *Socioeconomic factors* ($k = 96$), *Adolescent* ($k = 94$), *Aged* ($k = 91$), **Healthcare disparities* ($k = 89$), *Adult* ($k = 82$), *Child* ($k = 75$), *Health Services Accesibility* ($k = 72$) and *Health Equity/*organization & administration* ($k = 69$). We have presented here the Top20 more connected concepts in this semantic network. For the full list, please refer to **Supplementary Document 7**, network topology statistics for this network can be found in **Supplementary Table 1**.

Close examination of concepts such as *Poverty* within this network reveals important relationships with issues such as *Attitude to health*, *Health behavior*, *Professional-patient relations* and *Patient acceptance of Health Care*. For *Healthcare disparities*, in turn, related topics associated with the role of culture and education included *Attitude to health* and *Health behavior* but in this case there was no published literature linking *Professional-patient relations*, nor *Patient acceptance of Health Care*.

Furthermore, in the discussion of health equity and vulnerable populations in the health professional literature (as captured by this network), known vulnerable populations are not actually *central* to the discussion. Semantic relevance is often captured, not only by the degree centrality, but also by the importance *rank* (R) in the degree distribution. The most connected concept of a given network has rank 1, the second most connected concept has rank 2, and so on. Henceforth, we will often refer to the relevance of a given concept by stating its connectivity degree and rank (k,R). For instance, in this network, we can find less central concepts such as *Social Justice* ($k = 47, R = 37$) and *Racism* ($k = 47, R = 38$). Followed by *Minority groups* ($k = 42, R = 42$), *Hispanic Americans* ($k = 34, R = 59$), *Homeless persons* ($k = 25, R = 88$), *Disabled persons* ($k = 24, R = 104$), and *Refugees* ($k = 24, R = 106$). *Sexual and Gender Minorities* ($k = 13, R = 313$), *Transients and Migrants* ($k = 12, R = 337$) and *Prisoners* ($k = 8, R = 474$) are indeed significantly relegated in this discussion. Other vulnerable populations such as *African Americans*, *American Indians or Alaska Natives*, *Persons with Mental Disabilities* are not even represented in this *comprehensive* survey of the literature.

Let us now take a look at the role that *Culture*, *Education* and related concepts are playing in this discussion (as represented by the semantic network). The first (somewhat) related concepts that appear are *Health Promotion* ($k = 57, R = 26$) and *Attitude to health* ($k = 52, R = 29$), followed by *Patient*

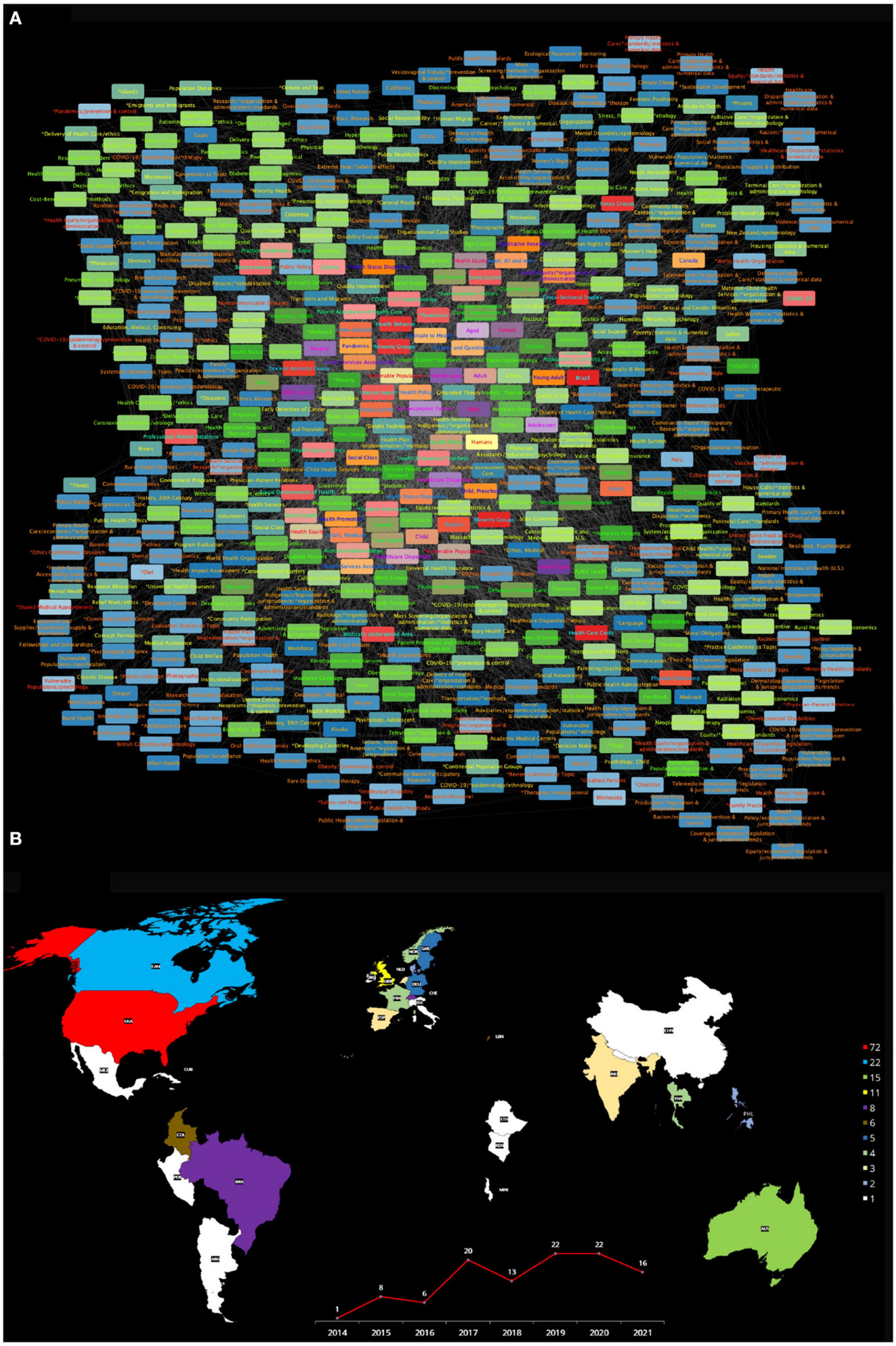


FIGURE 3 | (A) Semantic network built from the search Health equity [MeSH Terms] AND Vulnerable Populations [MeSH Terms]: from 2014 to 2021. **(B)** The publishing countries and number of entries. USA, United States of America; AN, Canada; AUS, Australia; GBR, United Kingdom; BRA, Brazil; CHE, Switzerland; COL, Colombia; LBN, Lebanon; DEU, Germany; SWE, Sweden; FRA, France; NOR, Norway; THA, Thailand; IND, India; NLD, Netherlands; ESP, Spain; DNK, Denmark; PHL, Philippines; ARG, Argentina; CHI, Chile; CHN, China; CUB, Cuba; ETH, Ethiopia; IRL, Ireland; ITA, Italy; JAM, Jamaica; KEN, Kenya; MWI, Malawi; MEX, Mexico; NPL, Nepal; PER, Peru.

acceptance of healthcare ($k = 33$, $R = 62$), *Culture* ($k = 33$, $R = 63$), *Health education, dental* ($k = 21$, $R = 144$), *Cultural diversity* ($k = 21$, $R = 151$), and *Health literacy* ($k = 21$, $R = 165$). Also concepts such as *Cultural competency* ($k = 18$, $R = 215$) and even *Language* ($k = 11$, $R = 356$) that may potentially contribute to both health inequities and population vulnerabilities are notably misrepresented.

In **Figure 3B**, further details about the countries of origin of the publications that formed the corpus for this semantic network (for *Health Equity* and *Vulnerable populations*) (color-coded according with the number of publications generated by each *country*), as well plot presenting the number of articles produced each year from 2014 to 2021 are presented. It can be noticed that a relatively small number of countries contribute to the discussion on these matters, and that many of them are either developed countries or emerging economies. We can also highlight the fact that there is a relatively low number of works discussing *Health Equity* and *Vulnerable populations*, with no more than 22 articles published within a given year.

In brief, the **Health equity and Vulnerable populations** semantic network, as comprehensively curated from the PubMed database, presents some important general tendencies, but also evidences some remarkable biases and misrepresentations (particularly, underrepresenting a number of relevant concepts). On the one hand, we have seen that the network reflects the importance of sociodemographics for the healthcare of vulnerable populations and highlights some health disparities that have become evident with the onset of the COVID-19 pandemic. It also shows that organizational and administrative issues have been at the core of the scholarly discussion on these matters. On the other hand, however, there is a noteworthy underrepresentation of concepts that can be considered relevant for the discussion on health equity and vulnerable populations, such as *Social justice*, *Racism*, *Minorities*, *Migrants*, *Homeless persons*, *Sex and gender minorities*, *Cultural diversity* and *Language* to name but a handful. Other important issues are not only relegated but absolutely absent from the discourse (as captured by this network), among these we can mention *African Americans*, *American Indians or Alaska Natives*, *Persons with Mental Disabilities*. By recognizing the worth of the discussed concepts, as well as the shortcomings and biases in other relevant issues, it will be possible to work toward a more equitable scholarly dialogue on the many dimensions of the health equity and vulnerable populations problem.

It is worth noticing that further exploration of the intricate web of relationships, perhaps with particular questions in mind, may be performed by *navigating* the interactive networks. The use of visual tools such as `Cytoscape` or `iGraph` is recommended, but the **Supplementary Network Documents** are also stand-alone searchable.

3.3.2. Health Equity and Social Determinants of Health Network

This network (see **Figure 4**) is composed of 921 nodes and 10,156 semantic relationships. As in the case of the previous network, the higher ranked concepts in this semantic network referred to *Humans* ($k = 912$), followed by *Health Equity* ($k = 519$), *Social Determinants of Health* ($k = 459$), **Social Determinants of Health* ($k = 378$) as well as demographic items: *United States* ($k = 294$), *Female* ($k = 280$) and *Male* ($k = 244$). Subsequent relevant concepts in the semantic network connectivity structure are *Socioeconomic factors* ($k = 241$, $R = 8$), followed by **Health Status Disparities* ($k = 230$, $R = 9$), *Health equity* ($k = 206$, $R = 10$), *Health Status Disparities* ($k = 177$, $R = 11$), *Adult* ($k = 171$, $R = 12$), *Health policy* ($k = 154$, $R = 13$), *Health Equity/*organization & administration* ($k = 137$, $R = 14$), *United States/epidemiology* ($k = 137$, $R = 15$), **Health Policy* ($k = 133$, $R = 16$), *SARS-CoV-2* ($k = 128$, $R = 17$), *Middle Aged* ($k = 118$, $R = 18$); as well as *Adolescent* ($k = 110$, $R = 19$) and *COVID-19* ($k = 110$, $R = 20$) to complete the Top20 concepts of this semantic network. For the full list, please refer to **Supplementary Document 8**, network topology statistics for this network can be found in **Supplementary Table 2**.

Among well-known SDH, *Poverty* ($k = 103$, $R = 21$), later on come *Educational status* ($k = 65$, $R = 40$), *Social conditions* ($k = 45$, $R = 63$), *Health knowledge, attitudes and Practice* ($k = 42$, $R = 70$), *Residence characteristics* ($k = 41$, $R = 71$), and *Housing* ($k = 40$, $R = 72$), then *Income* with ($k = 38$, $R = 86$) and further down the list come *Social class* ($k = 35$, $R = 91$) and *Social support* ($k = 34$, $R = 98$), that are relatively low ranked (91 and 98 out of 921) in spite of being considered among the more relevant SDH.

In relation to concepts related to *Culture* and *Education*, aside from *Educational status*, again we see that these topics are not central to the current discussion (as proxied by this semantic network). *Culture* ($k = 24$, $R = 157$) for instance is somehow relegated in this the network. With related topics such as **Culturally competent care* ($k = 19$, $R = 247$), *Organizational culture* ($k = 19$, $R = 248$), *Cultural Diversity* ($k = 18$, $R = 266$) and *Cultural competency/*education* ($k = 17$, $R = 282$) even less central to the discussion. Regarding education, the network includes concepts such as *Early intervention, educational* ($k = 31$, $R = 113$), *Patient advocacy/*education* ($k = 19$, $R = 239$), *Public Health/education* ($k = 13$, $R = 453$), *Minority Health/*education* ($k = 12$, $R = 507$). These low relevance ranks confirm the fact that the discussion about these matters is lagging with respect to other issues, more central in the current literature on Health Equity and the SDH.

This network reveals that known SDH are being discussed in relation to *Culture* and *Education* issues. We already commented on the published literature discussion in connection to *Poverty*. Other social determinants observed in this semantic network such as *Health knowledge, attitudes and practice* have been discussed in relation to *Social determinants of health/*ethnology* and *Educational status* however *Culture* as such is somehow absent in the scholarly discussion, that apparently is centered in ethnic features of the populations rather than on cultural issues. A more detailed and careful examination of the literature is however needed before making any conclusion in this regard.

As in the case of the health equity and vulnerable populations, the semantic network representing the interrelationships of **Health equity and Social determinants of health** presents a picture of the scholarly discourse with some nuances. It is a larger network with more than 900 concepts and

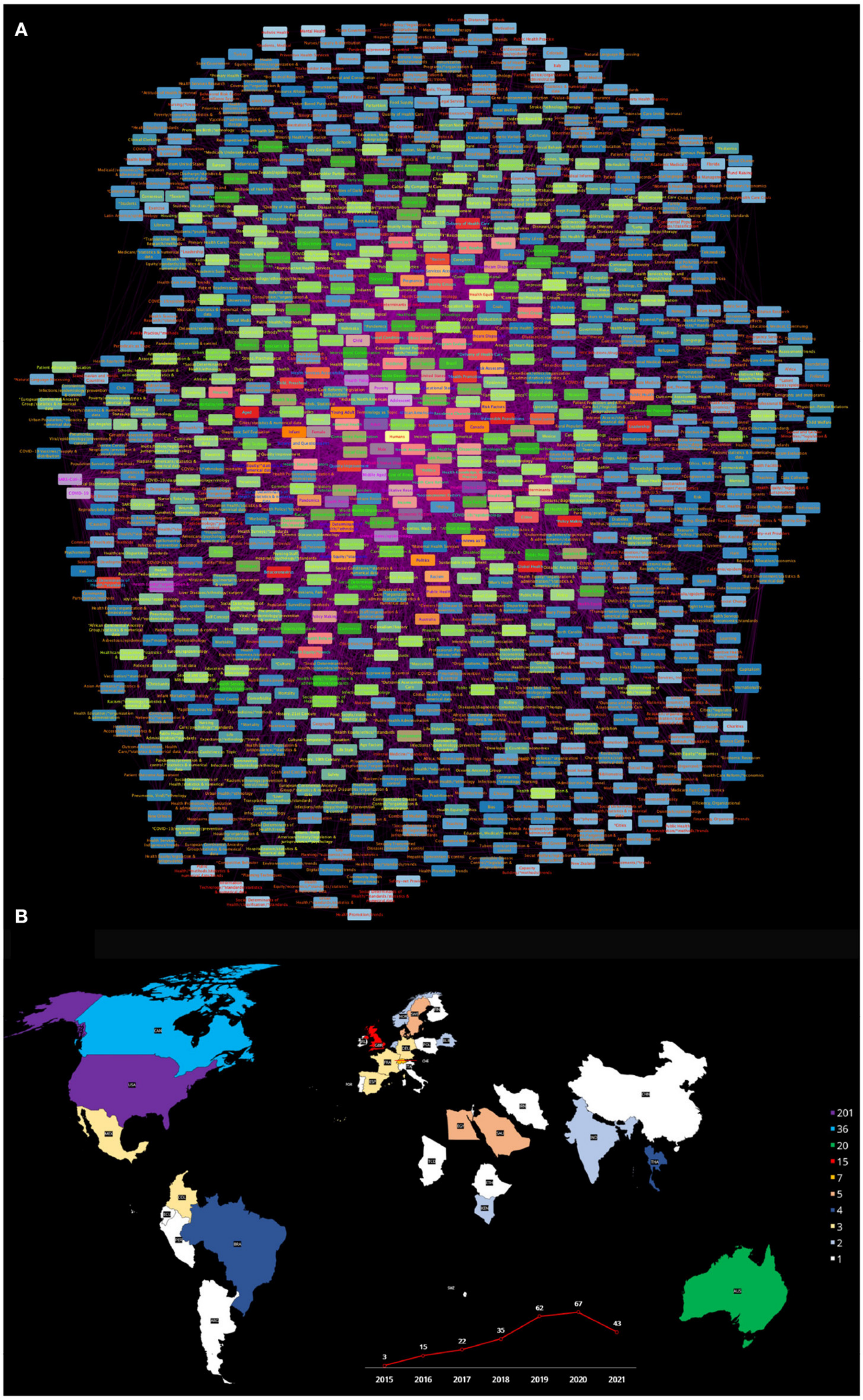


FIGURE 4 | (A) Semantic network built from the search Health equity [MeSH Terms] AND Social determinants of health [MeSH Terms]: from 2015 to 2021. **(B)** The publishing countries and number of entries. USA, United States of America; CAN, Canada; AUS, Australia; GBR, United Kingdom; CHE, Switzerland; DNK, Denmark; EGY, Egypt; SAU, Saudi Arabia; SWE, Sweden; BRA, Brazil; THA, Thailand; CHI, Chile; COL, Colombia; FRA, France; DEU, Germany; MEX, Mexico; ESP, Spain; BLR, Belgium; IND, India; KEN, Kenya; NLD, Netherlands; NOR, Norway; ARG, Argentina; TCD, Chad; CHN, China; ECU, Ecuador; ETH, Ethiopia; FIN, Finland; IRN, Iran; IRL, Ireland; ISR, Israel; ITA, Italy; MLT, Malta; PER, Peru; POL, Poland; PRT, Portugal; SWZ, Swaziland.

10,000 semantic relationships. Demographics and search terms are again dominant in the discourse, as expected. Concepts related to administrative and organizative aspects (*Health policy*, *Health equity/organization and administration*, *United States Epidemiology*) are, one more time, central to the discourse. A bit downgraded are concepts such as *Poverty*, *Educational status*, *Housing*, and *Residence characteristics*, in spite of being relevant components of the SDH problem. However, flagrant underrepresentation can be noticed in the case of item related to culture, being ranked between the 247th and the 507th concepts out of 921, with *Culture* not even incorporated into the discourse on Health equity and Social determinants of health.

As in the case of the previous network, **Figure 4B** presents a map with the different countries contributing to the published literature on *Health equity* and *Social determinants of health*, as well as a plot of the number of works published every year since 2015 on these issues. The main countries contributing to the scholarly discussion on these issues are quite similar to the one in the previous network. This is relevant for a global view of these matters, since it is likely that the academic view on the matters may be biased due to peculiarities of the health systems of these countries.

3.3.3. Health Equity and Culture Network

This network consists of 691 nodes-concepts and 7,836 edges-relationships. Relevant concepts include *Humans* ($k = 684$), **Health equity* ($k = 340$), *Female* ($k = 310$), *Health Equity* ($k = 253$), *Male* ($k = 246$) ranked 1st to 5th. *Cultural diversity* comes 6th with ($k = 192$), followed by *United States* ($k = 191$), **Cultural diversity* ($k = 169$), *Cultural competency* ($k = 163$) and *Culture* ranked in 10th ($k = 161$). The rest of the Top20 concepts are *Adult* ($k = 138$), *Socioeconomic factors* ($k = 130$), *Middle aged* ($k = 122$), *Qualitative research* ($k = 106$), **Cultural competency* ($k = 97$), *Young adult* ($k = 94$), *Surveys and questionnaires* ($k = 93$), **Healthcare disparities* ($k = 90$), *SARS-CoV-2* ($k = 80$), and *Health equity/*standards* ($k = 76$), respectively.

Other concepts related to culture that appear in this network are comparatively lagged behind, such is the case of *Cultural characteristics* ($k = 53, R = 41$), *Organizational culture* ($k = 53, R = 42$), *Cultural competency/*education* ($k = 49, R = 47$), *Paternalism* ($k = 39, R = 63$), **Culture* ($k = 32, R = 88$), **Culturally competent care* ($k = 31, R = 93$). Furthermore, *Language* ($k = 29, R = 103$) a central aspect of culture and **Organizational culture* ($k = 25, R = 121$) which are relevant for healthcare policy, design and practice are also less central to the discourse than expected. We can also mention –in connection to culture– *Religion* ($k = 24, R = 131$), *Culturally competent care/*organization & administration* ($k = 20, R = 170$) and **Cross-cultural comparison* ($k = 19, R = 214$). The rest of the concepts are presented in **Supplementary Document 9**, network topology statistics for this network can be found in **Supplementary Table 3**.

Although this network seems to be less biased that other semantic graphs analyzed, it remains worrying that concepts such as *Empathy* ($k = 13, R = 443$), *Self-concept* ($k = 11, R = 530$) and *Attitude to health/ethnology* ($k = 10, R = 564$), that we consider to be central to understand how culture contributes to shaping health equity (or inequities), remain somehow low ranked in the literature's discourse on the matters.

3.3.4. Health Equity and Trust Network

This is a relatively smaller semantic network consisting in 140 nodes-concepts and 1433 edges. Main concepts regarding network centrality degree are as follows (Top20 ordered according to their ranking) *Humans* ($k = 139$), *Trust* ($k = 93$), *Male* ($k = 63$), *Health equity* ($k = 63$), *Aged* ($k = 57$), *Middle Aged* ($k = 57$), **Health equity* ($k = 54$), *Communication* ($k = 51$), *Qualitative research* ($k = 46$), **Trust* ($k = 46$), *Female* ($k = 40$), *Adult* ($k = 40$), *Social support* ($k = 28$), *Pulmonary disease, Chronic obstructive/*rehabilitation* ($k = 28$), *Saskatchewan* ($k = 28$), **Patient Acceptance of health care* ($k = 28$), *Shame* ($k = 28$), *Self-Management* ($k = 28$), *Case management* ($k = 28$) and *Disease management* ($k = 28$).

Other related items are **Attitude to health* ($k = 28$), *Focus groups* ($k = 28$) and *Patient participation* ($k = 28$). Followed further down in relevance by concepts like *Stakeholder participation* ($k = 22, R = 37$), *Trust/psychology* ($k = 18, R = 52$) and *Culture* ($k = 14, R = 83$). Issues such as **Health communication* ($k = 9, R = 125$) and **Communication barriers* ($k = 7, R = 134$) that are instrumental for a balanced discussion of trust are also somehow disregarded. It calls into attention that a number of terms related to organizational and administrative aspects are included in (and central to) the discussion (as proxied by the semantic network), but fewer aspects of a personal or emotional side of the issue are included.

For the full list, please refer to **Supplementary Document 10**, network topology statistics for this network can be found in **Supplementary Table 4**.

3.3.5. Health Equity and Health Literacy Network

The Health equity and Health literacy network is conformed by 166 concepts (nodes) and 1,670 semantic relationships or edges. The more connected components are *Humans* ($k = 165$), **Health equity* ($k = 110$), **Health literacy* ($k = 97$), *Health literacy* ($k = 86$), *Health equity* ($k = 78$), *Adult* ($k = 69$), *Male* ($k = 66$), *Female* ($k = 58$), *United States* ($k = 47$), and *Middle aged* ($k = 46$) that appears in the first 10 positions respectively. Concepts ranked from the 10th to the 20th are as follows: *Qualitative research* ($k = 39$), *Aged* ($k = 37$), *Social support* ($k = 37$), **Health services accessibility* ($k = 36$), *Social determinants of health* ($k = 36$), *Australia* ($k = 30$), *Adolescent* ($k = 30$), *Public health* ($k = 30$), *Pulmonary disease, Chronic obstructive/*rehabilitation* ($k = 28$) and *Saskatchewan* ($k = 28$).

Other relevant concepts that appear on this network are **Attitude to health* ($k = 28$), *Patient participation* ($k = 28$), *Patient education as topic* ($k = 28$), *Culture* ($k = 24$) and *Health promotion* ($k = 22$). Interestingly *Literacy* ($k = 16, R = 75$) and *Reading* ($k = 16, R = 80$) appear in unexpectedly low key positions. *Communication barriers* ($k = 14, R = 93$) comes still later on, aside with *Educational status* ($k = 14, R = 94$) and *Culture* ($k = 14, R = 95$) and *Health knowledge, attitudes, practice* ($k = 12, R = 112$). It seems that the emphasis on health literacy in the current discourse is in relation to literacy in the healthcare professionals and not so much about health literacy in the general population. The recent *infodemic* around COVID-19 has further highlighted the strong urgency for the general population to be more literate on healthcare and public health issues.

For the full list, please refer to **Supplementary Document 11**, network topology statistics for this network can be found in **Supplementary Table 5**.

3.3.6. Health Equity and Education Network

This semantic graph (see **Figure 5**) consists of 1,673 nodes (MeSH terms) and 21,952 edges-relationships among them. Most connected concepts were: *Humans* ($k = 1659$), **Health equity* ($k = 853$), *Female* ($k = 737$), *Health equity* ($k = 602$), *Male* ($k = 570$), *United States* ($k = 505$), *Adult* ($k = 475$), *Middel aged* ($k = 321$), *Socioeconomic factors* ($k = 475$) and *Health promotion* ($k = 271$), ranked in the first 10 places, respectively. The next 10 more connected concepts were: *Surveys and questionnaires* ($k = 2641$), *Aged* ($k = 254$), *Needs assessment* ($k = 241$), *Health equity/*organization & administration* ($k = 235$), **Health promotion* ($k = 231$), *Child* ($k = 227$), *Adolescent* ($k = 218$), *Health status disparities* ($k = 210$), *Qualitative research* ($k = 208$) and *Young adult* ($k = 206$).

Other MeSH terms related to education within this semantic network, aside from *Educational status* ($k = 200$, $R = 21$), calls to attention that other concepts, essential to understand the role of education in health equity are placed in less relevant positions in the semantics of the scholarly discussion, such is the case of **Health literacy* ($k = 97$, $R = 53$), *Health literacy* ($k = 86$, $R = 63$), *Cultural competency* ($k = 79$, $R = 70$), *Universities* ($k = 73$, $R = 78$), as well as *Health knowledge, attitudes, practice* ($k = 66$, $R = 91$), *Patient education as topic* ($k = 65$, $R = 94$), **Education, medical* ($k = 62$, $R = 101$), as well as its counterpart *Education, medical* ($k = 57$, $R = 112$). Additional concepts have even lower connectivity degrees, such as **Healt education* ($k = 41$, $R = 169$), **Culture* ($k = 32$, $R = 258$), *Learning* ($k = 32$, $R = 259$), *Knowledge* ($k = 29$, $R = 293$), as well as *Health education/*ethics* ($k = 24$, $R = 382$). Further down the list come **Access to information* ($k = 19$, $R = 573$) and **Mothers/education* ($k = 19$, $R = 594$). These latter concepts are strikingly underrepresented: *how can one envision health equity through education when access to information is ranked 573 in the list of relevant concepts and mothers' education comes in place 594?*

The fact that this semantic network is relatively large and somehow rich in terms, may be connected with a nascent interest in the role of education in the context of health equity, which is, in itself, remarkable. As in other networks discussed, however, it seems that technical and administrative issues are dominant, whereas issues more closely related to the individuals and populations are somehow relegated or even absent. Again, as it can be seen in **Figure 5B**, there may be a representation bias in the discussion toward the situation in the countries that are contributing to this discourse.

For the full list, please refer to **Supplementary Document 12**, network topology statistics for this network can be found in **Supplementary Table 6**.

3.4. Stage IV: Data Visualization

Data visualization is indeed a relevant component of network analytics. Visual display provides a helpful overview of the structure of complex networks. Since network depictions are indeed aimed at being *representational*, looking at their full structure allow us to generate conceptual maps. This is all the more relevant when the network themselves encode conceptual information such as the case of semantic networks (23, 45, 46).

In this section, we will provide a general schematic view of the semantic networks analyzed in this work as well as three representative examples. As previously mentioned the information to build all of the networks (not just the three shown here) is given in the **Supplementary Materials**.

3.4.1. Feature Specific Subnetworks

To continue extracting semantic context from the analyzed networks, three subnetworks were extracted from each of the main networks (**Figures 3–5**) to construct a theoretical discussion and visualize outstanding patterns based on the following MeSH terms and their first neighbors (see **Figures 6–8**):

- *Health equity*, *Vulnerable Populations* and *Culture* subnetwork (HVC subnetwork).
- *Health equity*, *Social determinants of health* and *Culture* subnetwork (HSDHC subnetwork).
- *Health equity*, *Education* and *Culture* subnetwork (HEC subnetwork).

These subnetworks and the underlying concepts will be further discussed in the context of the thematic analysis. This will considered in the corresponding heading in the Section 4 (Section 4.2).

4. DISCUSSION

4.1. Semantic Relations

Semantic networks have been used to represent conceptual or referential relationships between concepts to generate knowledge *via* representation (63–65). In the present study, we have built several semantic networks over an ontology, that serves as a referential framework and is given by the Medical Subheading (MeSH) controlled and hierarchically organized vocabulary (23, 45, 46). These semantic networks were aimed to analyze the structural relationships behind concepts relevant to our understanding of how health equity and inequity phenomena arise in the different contexts of culture, education and other SDH. We believe that this approach is useful to analyze large literature corpora (such as the ones comprising the current health literature) and characterize the conceptual relationships of what is being discussed in a systematic and unbiased manner. Aside from setting the foundations for deeper explorations and critique, this approach allows us to discern biases and limitations, even fields of opportunity in the scholarly discussion of such relevant issues.

This is, in our view, a timely discussion. In recent times, the fact that health equity theory and practice are indeed subject to implicit and structural biases, has been highlighted

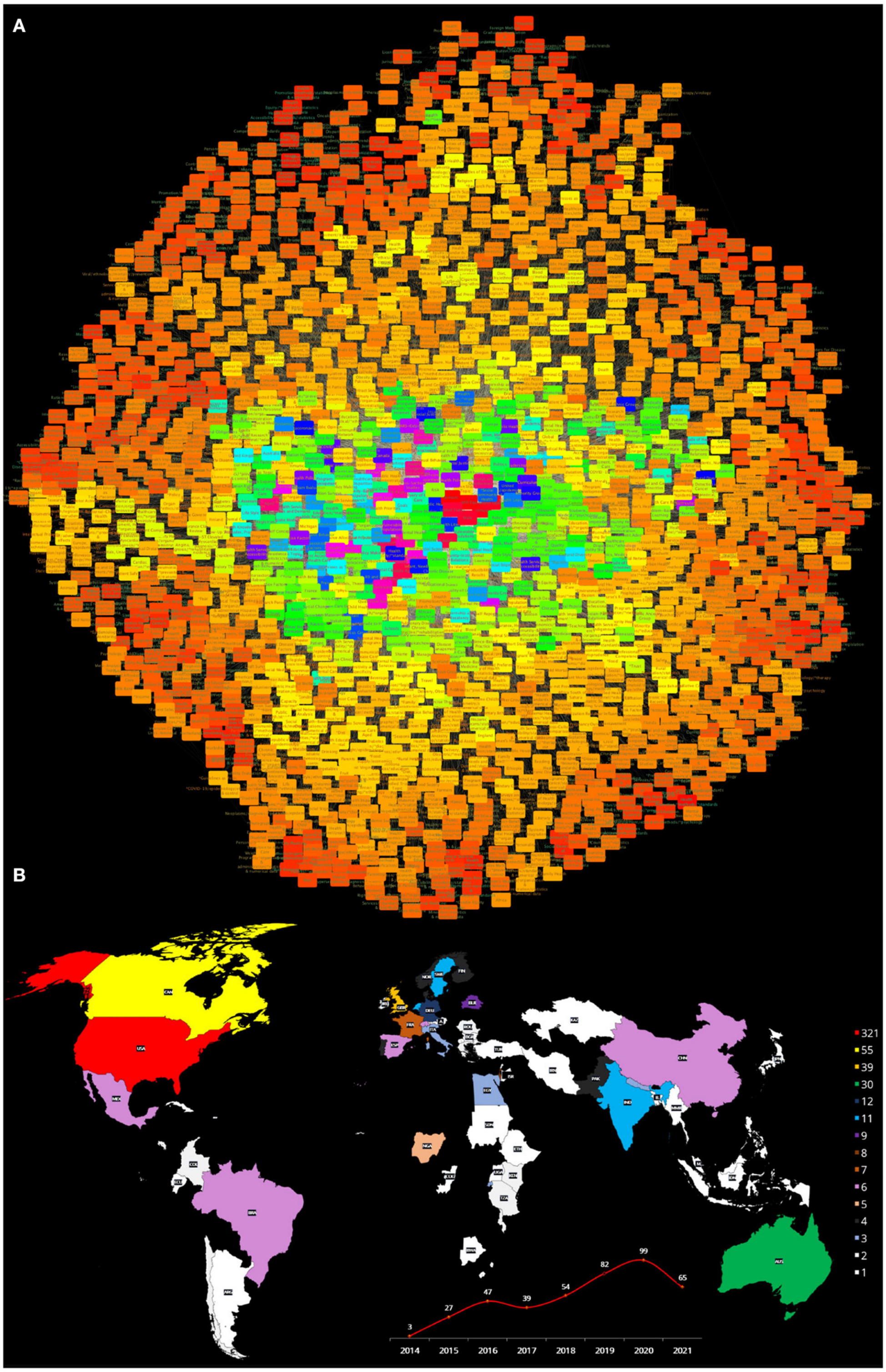


FIGURE 5 | (A) Semantic network built from the search Health equity [MeSH Terms] AND Education [MeSH Terms]: from 2014 to 2021. **(B)** The publishing countries and number of entries. USA, United States of America; CAN, Canada; GBR, United Kingdom; AUS, Australia; DEU, Germany; IND, India; NLD, Netherlands; SWE, Sweden; BLR, Belgium; ISR, Israel; FRA, France; BRA, Brazil; CHN, China; MEX, Mexico; ESP, Spain; CHE, Switzerland; NGA, Nigeria; DNK, Denmark; FIN, Finland; NOR, Norway; PAK, Pakistan; PRT, Portugal; EGY, Egypt; ITA, Italy; NPL, Nepal; RWA, Rwanda; AUT, Austria; CHL, Chile; COL, Colombia; CRI, Costa Rica; GRC, Greece; IRL, Ireland; JOR, Jordan; KEN, Kenya; LBN, Lebanon; MWI, Malawi; ROU, Romania; TZA, Tanzania; ARG, Argentina; BGD, Bangladesh; BWA, Botswana; BGR, Bulgaria; COG, Congo; CUB, Cuba; ECU, Ecuador; ETH, Ethiopia; GMB, Gambia; IDN, Indonesia; IRN, Iran; JPN, Japan; KAZ, Kazakhstan; LSO, Lesotho; MKD, Macedonia; MYS, Malaysia; MLT, Malta; MMR, Myanmar; PHL, Philippines; PRI, Puerto Rico; SGP, Singapore; SDN, Sudan; SWZ, Swaziland; TUR, Turkey; UGA, Uganda.

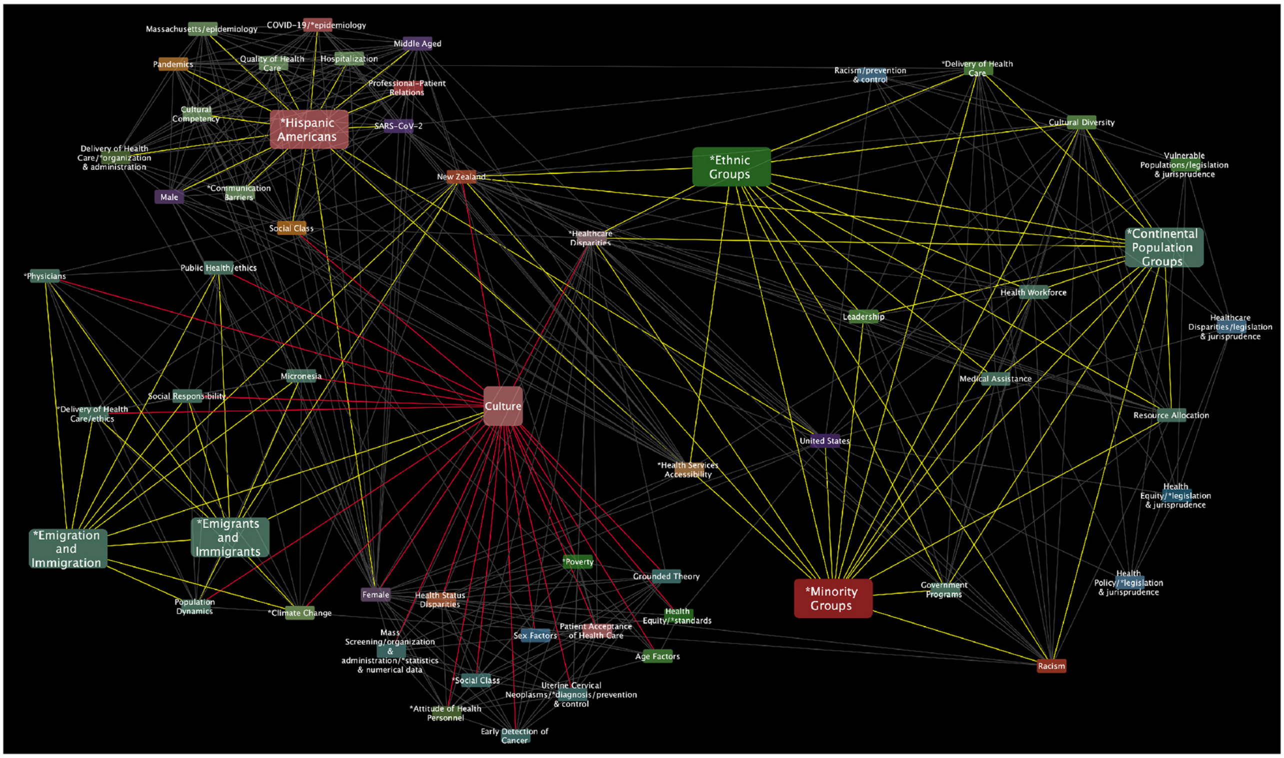


FIGURE 6 | Health equity, Vulnerable Populations, and Culture MeSH terms subnetwork. This *HVC subnetwork* has 57 nodes and 381 edges.

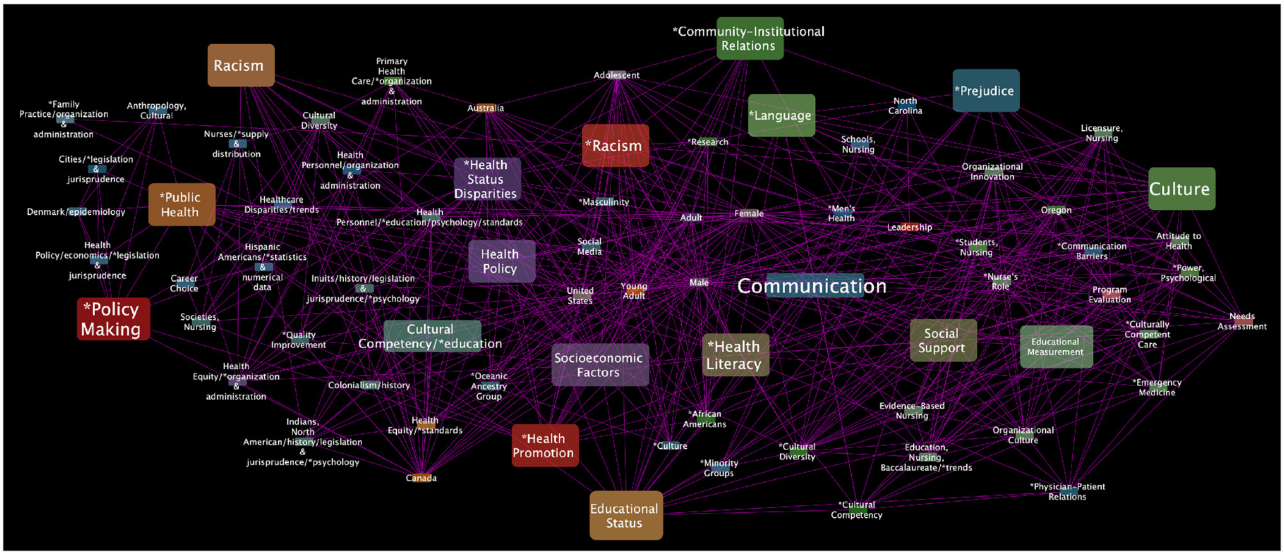


FIGURE 7 | Health equity, Social determinants of health, and Culture MeSH terms subnetwork. This *HSDHC subnetwork* has 75 nodes and 519 edges.

repeatedly (66–69). This has been further noticed in the context of the ongoing COVID-19 pandemic (61, 62, 70–73). In what follows, we will briefly discuss some issues we find revealing and interesting regarding the current health professional literature trends on the relationships between concepts like *Health equity*, *Vulnerable populations*, *Social determinants of health*, *Culture*, *Trust*, *Health literacy* and *Education* as observed from analyzing the six semantic networks derived from the systematic literature

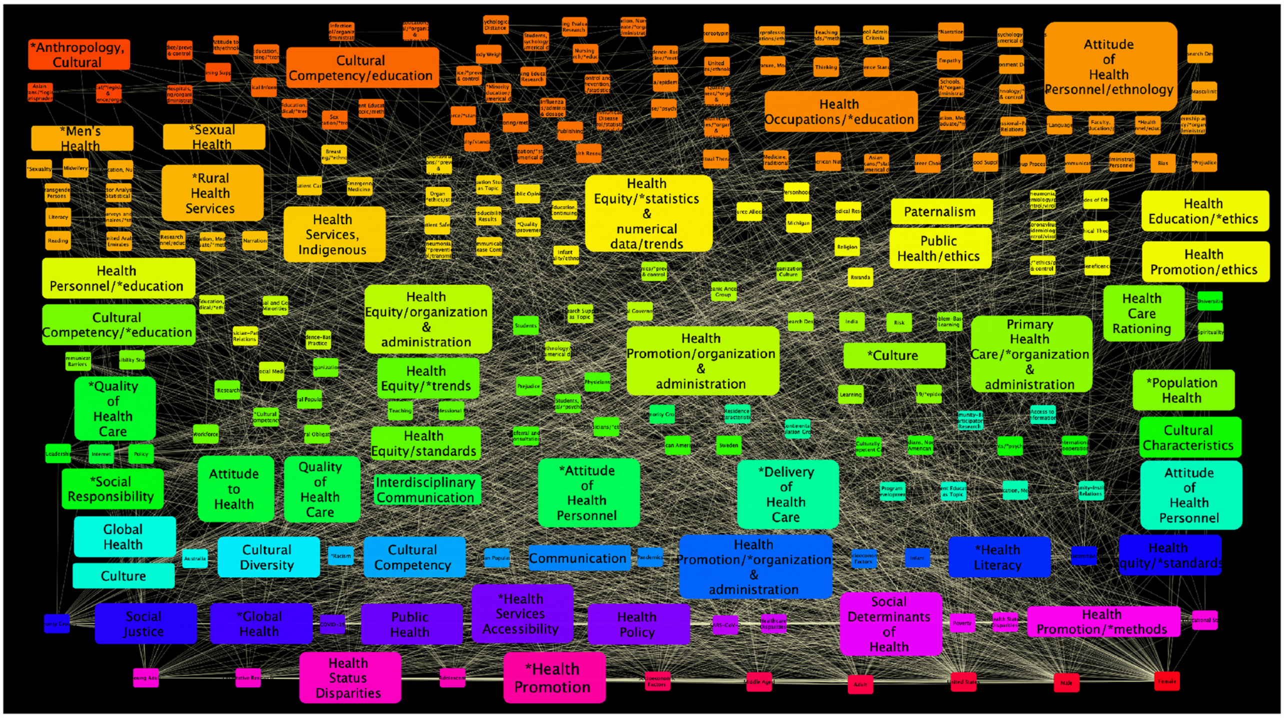


FIGURE 8 | Health equity, Education, and Culture MeSH terms subnetwork. This *HEC subnetwork* has 232 nodes and 2,359 edges.

search instances introduced in Section 2.6, and described in Section 3 (Sections 3.3.1 to 3.3.6), and in the related subnetworks as presented in Section 3.4.

We will first discuss, what we have learned by analyzing the semantic network obtained from the literature corpus of the joint search of *Health equity* [MeSH] and *Vulnerable populations* [MeSH] (Section 3.3.1). Even before actual analysis of the concepts and relationships associated. We were able to notice some particularities of the scholarly discussion on these matters.

First of all, the PubMed database grows in hundreds of thousands to millions of articles every year, in all fields of life sciences and biomedical topics (74). With this in mind, it seems astonishing that only 108 articles were found, with no more than 22 articles written on any given year regarding *Health equity* and *Vulnerable populations*. The second issue is that (as is often the case in the health research literature), the articles forming this corpus were written in a handful of countries, mostly corresponding to developed nations or emerging economies. These include the United States of America, Canada, Australia, United Kingdom, Brazil, Switzerland, Colombia, Lebanon, Germany, Sweden, France, Norway, Thailand, India, Netherlands, Spain, Denmark, Philippines, Argentina, Chile, China, Cuba, Ethiopia, Ireland, Italy, Jamaica, Kenya, Malawi, Mexico, Nepal, and Peru. The main contributors (as it can be seen in **Figure 3B**) are indeed the United States of America, Canada, and the United Kingdom whose socioeconomic conditions and the specificities of their health systems may drive important biases in the conclusions of their research making difficult to generalize some of the knowledge generated and presented in said articles, an issue that has been already documented in the context of the health professional literature (75–77), but also has been noticed in the context of health equity differences (5, 78–80).

Moving onto the results of the semantic network itself, one can notice that the distribution of *degree centralities* reveals interesting clues. Centrality degree—i.e., the number of relationships a given node-concept has in a semantic network—, has been recognized as a key indicator of the relevance of the concept to the overall conceptual picture of an issue as represented by the semantic network (81, 82). In this context, we have observed that some central concepts related to the situation of vulnerable populations with regards to health equity, such as *poverty*, *healthcare disparities*, and *age* are being recognized as such in the published health professional literature as indicated by their high degrees and priority rankings in the network. These terms are all in the Top 20 more *central* concepts, discussed in a larger number of publications in relation to different aspects of the health equity/vulnerable population studies. However, as previously commented in the Section 3, other terms that are intuitively relevant to this discussion are being somehow disregarded. For instance, the fact that *Social justice* and *Racism* are ranked in the 37th and 38th place for being connected with just 47 other concepts (out of 550 possible), a fact that may reveal important gaps in the literature, as has been known for some time (83–86).

Even more intriguing is the fact that *Sexual and gender minorities*, *Transients and migrants* and *Prisoners* admittedly some of the most vulnerable groups in relation to social equity are in the *periphery* of the semantic network, ranked in the 313, 337, and 474 out of 551 concepts, hence stressing even more their vulnerable role, not only in connection with healthcare, but even with respect to the health research literature (87–90). Perhaps, the most striking finding of our semantic network analysis of the relationship between *Health equity* and *Vulnerable populations* is the fact that *African Americans*, *American Indians or Alaska natives* and *Persons with mental disabilities* are not even explicitly mentioned as relevant concepts (i.e., these issues may be touched-upon in some of these articles, but no MeSH identifier has been recorded for these issues in any of the 108 articles analyzed to build this network.

In connection with the role that *Culture* and *Education* may play in the context of *Health equity* and *Vulnerable populations*, these are still rather peripheric concepts on this network. Hence, in the corpus of published health literature on health equity and vulnerable populations, topics such as *Attitude to health*, *Patient acceptance of healthcare*, *Culture* and *Cultural diversity* are not connected to the main concepts in this semantic network. This points out to the need, to bring the discussion on these important concepts into the mainstream health professional literature on health equity, integrating them appropriately.

Let us now analyze what we found in the semantic network built upon the search on *Health equity* and *Social determinants of health* encompassing 921 concepts as discussed in 254 published works. Following a similar pattern in the distribution of countries contributing to the scholarly discussion on these issues as it can be seen in **Figure 4B**, most of the literature comes from countries such as the United States of America, Canada, Australia and the United Kingdom, with some contributions by authors in Switzerland, Denmark, Egypt, Saudi Arabia, Sweden, Brazil, Thailand, Chile, Colombia, France, Germany, Mexico, Spain, Belgium, India, Kenya, Netherlands, Norway, Argentina, Chad, China, Ecuador, Ethiopia, Finland, Iran, Ireland, Israel, Italy, Malta, Peru, Poland, Portugal, and Swaziland. So we can notice the addition of countries with a broader scope of socioeconomic and cultural conditions. The number of yearly publications is a bit higher than in the previous network, but still rather small, with a maximum of 67 articles per year.

We observed that *some* terms associated with SDH, such as *Socioeconomic factors* and *Health status disparities* occupy relevant places in the semantic network (ranked 9 and 11 out of 921, respectively). However, other SDH are less connected in this network; *Social conditions* is ranked in the 63rd place, furthermore *Social support* which is a key structural determinant of health ranks in the 98th place with only 34 connections out of 920. In spite of its relevance as a relief factor to modulate SDH, *Social support* has been documented to be underrepresented in the specialized literature (91, 92). It has been discussed that publication biases regarding SDH may indeed obey cultural reasons, an issue that is central to the discussion of the role of culture and education in health equity (93–96).

The relationship between *Culture* itself and *Health equity* has been studied here, as it is presented in Section 3.3.3. Important concepts such as *Cultural diversity* and *Cultural competency* are well-represented concepts in this semantic network. These two concepts are indeed closely connected: embracing *Cultural diversity* helps healthcare providers to offer their services, recognizing the unique social, cultural and even linguistic features of their patients in the context of their populations (97, 98), this in turn leads to *Cultural competency* of the health systems, that is, the ability of such systems to provide care consistent with the values beliefs and behaviors of the patients (5, 99). However, other concepts such as *Empathy*, *Self-concept* and *Attitude to health/ethnology* are scarcely connected to the main discourse as reflected by their degree rankings (places 443, 530 and 564 out of 691 respectively). Calls to attention that, apparently, healthcare systems features are in a more common and centralized discussed in the current literature on health equity and culture than *individual* or *personal* concepts.

Such *personal* characteristics are indeed a central part in the establishment of *Trust* in the healthcare setting (100–104). The conceptual relationships around *Health equity* and *Trust* were also studied in detail here. In Section 3.3.4, we have presented some results of the analysis of this semantic network. As we already mentioned, such individual, even personal, *Human* features are scarcely discussed in the literature on *Healt equity*. We can notice, for instance, that the *Health equity - Trust* network is based on a smaller literature corpus of just 14 articles leading to a reduced network of only 140 concepts. Terms such as *Communication* and *Social support* are relatively well-connected in this network (ranks 8 and 12 out of 140, respectively), but related issues such as *Health communication* and *Communication barriers* are still peripheral concepts connected to just 9 and 7 out of 139 terms, ranking 125th and 134th. Since good communication is key to build proper trust relationships between patients and healthcare providers (105, 106), improving the discussion on these issues seems desirable.

Building up trust in healthcare systems needs improving communication channels (105, 107). To do this, often is needed to improve *Health literacy* (108, 109). The web of concepts related to the role played by *Health literacy* in *Health equity* was also explored, main results were outlined in Section 3.3.5. The network described therein is also a somewhat small network comprising 166 concepts connected by 1,670 semantic relationships taken from 27 articles. Aside from the search terms, few concepts pertinent to an actual discussion of the role that culture, education and trust play in the construction of health equity are at the core of this network. For instance, the MeSH term *Health knowledge, attitudes, practice* that in our view would be quite relevant is indeed placed 112 out of 166 concepts in terms of conceptual connectivity. Furthermore, most of the discussion along these lines refer to the knowledge, attitudes and literacy *of healthcare workers* to attain health equity. While enormously important, healthcare workers and providers are just one side of the story. The discussion on health literacy, attitudes to knowledge and culture from the standpoint of the patients and their families has been largely disregarded (110–112).

These issues are indeed closely related to our findings in the context of the *Health equity - education* axis, as presented in Section 3.3.6. In contrast with the two previously discussed conceptual networks, this one is much larger (1,673 concepts) and *denser* (21,952 semantic relationships) with information coming from 420 published research works. *Health promotion* appears in the core of the network (ranked 10 out of 1,673 concepts), something we consider to be positive. Also relatively central to the discussion are concepts like *Educational status*, *Health literacy* and *Cultural competency* (ranked 21, 53, and 70, respectively). Though somewhat less connected, *Patient education as topic* is still within the top100 (rank 94 out of 1,673) more relevant terms. We believe that some improvement can be made in this regard, in particular since patient education has been described as instrumental to achieve health equity (113–115).

4.2. Thematic Analysis

To deepen on the discussion about focal issues, we have performed *thematic analysis* of the literature corpus using Atlas-ti over the associated domains in the semantic network. In this regard, we can deliberate upon the following matters:

Regarding vulnerable populations, we examined the specific connections of the *Culture* MeSH terms in a subnetwork of the semantic network in **Figure 3**. As you can see in the resulting (**Figure 6**), some main terms are directly or indirectly connected with *Culture* MeSH term, some of them are highlighted (larger node size) such as *Hispanic Americans*, *Emigrants and Immigrants*, *Ethnic groups*, *Minority groups* and *Continental populations groups*. However, there are other critical terms related to vulnerable population that are not connected within the Culture MeSH term subnetwork (composed of 490 MeSH terms), such as, *Homeless persons*, *Prisoners*, *Disabled persons*, *Refugees*, *Rural population*, *Intellectual disability*, *Disabled persons*, *Sexual and gender minorities*, *Terminally ill people*, *People suffering violence*. In light of the thematic analysis carried out with Atlas.ti, they seem to be important social and cultural determinants that can determine some inequities in health. For example, people experiencing homelessness or vulnerable housing are often marginalized and are known to face barriers to accessing appropriate healthcare services (9). Although changes have been recommended in the complex health systems, so that it should be more equitable, more sensitive and empathic, and more informed about the traumatic situations experienced by homeless people, barriers related to cultural aspects are barely mentioned in those key documents (116–118). Similar patterns can be seen in the studies of other conditions of vulnerability and adverse circumstances that apparently seem disconnected from their own cultural aspects or they seem irrelevant for health equity (119).

Regarding the Health equity, Education and Culture subnetwork (see **Figure 8**) the terms *Health promotion*, *Health services accessibility*, and *Social justice* are highly interconnected and related to other key terms such as *Cultural diversity*, *Cultural competence*, *Attitude to Health*, *Interdisciplinary communication*, *Health policy* and *Health literacy*. However, in this triad of terms, some no less important but not directly connected to culture have been excluded (the subnetwork is composed of 1440 MeSH terms), such as: *Health behavior*, *Cooperative behavior*, *Social support*, *Social stigma*, *Health knowledge, attitudes and practice*, *Social Class*, *Education Medical*, *Community participation*, *Life style*, *Consumer health information*, *Decision making*, *Self-management*, *Quality of life*, *Social change*, *Personal satisfaction*, *Social welfare*, *Motivation*, *Interpersonal relations*, *Professional competence*, *Social environment*, *Social conditions*, *Health services needs and demand*, *Treatment outcome*, *Social skills*, *Resilience*, *Social values*, *Social norms*, *Life expectancy*.

The thematic analysis also shows that are important social and cultural determinants that can influence some inequities in health. Health literacy has been a particularly prominent issue on the political, academic and scientific discourse on equity in health. The World Health Organization has established an urgent mandate for public policy action on health literacy as a key pillar for achieving health equity worldwide (120). In the reviewed documents, health literacy is presented as a universal challenge associated with wellbeing, access to healthcaere and improved health outcomes (121–123). However, this concept is interrelated with others that apparently are not related to culture and health equity. For example, the health literacy community movement driving for social change toward empowerment and health equity is related to some concepts like *Public health*, *Social change*, or *Social support*. But, the literature mentions that social movements are developed to impact health by generating changes related to cultural and social norms (124). Also, other documents mention that health literacy is necessary to make appropriate decisions regarding health. And again points out that some cultural factors contribute to reducing health inequities in this regard. The scholarly discussion on these issues is hence still far from being conclusive.

Other prominent social determinants of health derived from the thematic analysis of literature were connected to social and cultural responses for health equity. Not only the health care sector, but also, the education sector, administrators, financial systems, reimbursement mechanisms, industry, community centers, civil society groups, social networks, political organizations, even artists or cultural workers among others play a critical role in creating conditions for intersectoral collaboration and distributing resources that promote health equity (125–134). Theses roles appear, indeed, disconnected from the main discourse on the matters.

Finally, we explored the subnetwork of Health equity, Social Determinants of health and Culture subnetwork in the light of the thematic analysis. Some relevant terms emerged such as: *Socioeconomic factors*, *Social support*, *Cultural competency*, *Health promotion*, *Educational status*, *Health literacy*, *Comunication*, *Health policy*, *Language*, *Racism* and *Prejudice* (see **Figure 7**). However, some MeSH terms were less common in the studies reviewed and are not directly connected to the culture term (around 846 MeSH terms in the subnetwork). For example, *Poverty*, *Housing*, *Residence characteristics*, *Income*, *Environment*, *Food security*, *Race factors*, *Adverse childhood experiences*, *Social discrimination*, *Public assistance*, *Employment*, *Healthcare financing*, *Language*, *Health services*, *Social capital*, *Social welfare*, *Healthy lifestyle*, *Social networking*, *Social segregation*, *Urbanization*, *Social Isolation*, among others. Many of these overlap in the same individuals or communities, exacerbating their vulnerability and the health inequities (135). The characterization of SDH is critical to implementing actions that are more inclusive of and more sensitive to the different needs of the population as the WHO has instructed in many regions of the world, especially for disadvantaged sectors of society (14). However, we again identify some research gaps in terms of social determinants and cultures that can be explored in the future to help understand their interaction with health equity. We have presented and discussed only a few instances of the many connections and biases that can be found in the healthcare literature about health equity and culture. By resorting to the generated searches and resources available here and in other studies, the interested researchers may indeed discover many more instances and relationships relevant to these important yet somehow understudied issues.

4.3. Study Assumptions, Scope, and Limitations

As previously mentioned, the current study is founded on several basic assumptions that will shape the scope and present some limitations. The main analysis is somehow constrained by the use of the MeSH term classifiers. While this is an excellent method for identifying major research topics, emerging or potentially interesting topics may not be easy to spot, still presents an incomplete picture (33).

Although we carried out a complementary search in other multidisciplinary databases (LILACS and DOAJ) and both offer some kind of controlled vocabulary, the results differ significantly, though the main conclusions still hold. This happens because in some archiving schemes data curation is often limited to a simple thesaurus of keywords or concepts; which can also affect the correct interpretation of the search, and the reproducibility or comparability of the results.

Another issue that may be considered a limitation is that we used open access databases, which can also contribute to not achieving a complete search. However, the use of restricted access databases (some of which are behind expensive "paywalls") would bring another set of limitations, mostly regarding accession bias. Furthermore, restricted access to health information contributes to deepening the gaps and increasing health inequities.

Also, we are aware that different search systems may give rise to different results even when the same query has been employed, as these systems have different indexing methods, data presentation, and curation methods (136). Retrospective coverage of the controlled vocabulary may be limited, for instance. Thus, it may be difficult to quantify the quality of such controlled vocabulary as their features are diverse.

Additionally, as already mentioned, the choice of MeSH classifiers as the basis for the semantic network analysis introduced a number of assumptions (see Section 2.6).

Other methodological constraints arise from the use of automated or semi-automated analysis tools. The use of software such as Cytoscape or Atlas.ti to conduct data analysis also has a limitation related to the decontextualization of the findings, which can result in data interpretation weaknesses. However, the statements of researchers from different fields of knowledge concerning the advantages and disadvantages of the software used have diminished over the time with the evidence of its usefulness for analysis (137).

5. FINAL REMARKS

This work aimed to characterize, at a large scale, how social and cultural determinants may interact with health inequity and the interrelationships among them in different populations and diverse contexts. To this end, we have introduced semantic networks as a theoretical framework and methodological tool to carry out this analysis in a comprehensive, minimally-biased manner. We have built semantic models based on an ontology representation given by the Medical Subheading (MeSH) identifiers as developed by the National Library of Medicine of the United States of America and implemented our network construction based on a set of preselected searches in the PubMed database. Since MeSH terms were developed to be general purpose identifiers and being PubMed the most comprehensive database of academic publications in medicine and related topics; we believe that using these resources, though not ideal, is the least-biased and more comprehensive automated approximation to analyze the scholarly literature on these issues.

Our semantic network approach confirms the central role of some concepts in the academic discussion on health equity and culture, in the context of vulnerable populations, taking into account their SDH and how trust may arise in the different circumstances of health literacy and education. However, we have also found some biases and under-representation of several relevant concepts, likely influenced by the fact that the academic literature is both relatively scarce and produced in a few countries. Most of these countries are actually developed or emerging economies characterized by firmly established trends in their health systems. By pointing out such biases and sub-represented concepts in the discussion, it is possible to identify areas of opportunity for further academic development. Our view as presented here is of course, of a rather general and broad scope. However, the curated literature corpora, the semantic networks built and their statistical and topological structure analysis provided as **Supplementary Materials** may constitute a useful resource to navigate the full body of literature on these issues. Further insight was derived by considering additional data sources and by performing thematic analysis of discourse. However, perhaps the main *conclusion* is that there is still a long way to go toward a full scholarly representation of health equity and its relation to culture, with their many facets and complexities.

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.

AUTHOR CONTRIBUTIONS

EH-L and MM-G: conceptualization, methodology, software, supervision, and validation. EH-L, MM-G, and JMVC: formal analysis and writing—original draft preparation, review, and editing. All authors read and approved the final manuscript.

ACKNOWLEDGMENTS

The authors want to thank Mrs. Gabriela Graham for her support with language editing and proofreading of this manuscript.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2022.834172/full#supplementary-material

REFERENCES

1. Lathrop B. Moving toward health equity by addressing social determinants of health. *Nurs Women's Health*. (2020) 24:36–44. doi: 10.1016/j.nwh.2019.11.003
2. Browne AJ, Varcoe C, Ford-Gilboe M, Nadine Wathen C, Smye V, Jackson BE, et al. Disruption as opportunity: Impacts of an organizational health equity intervention in primary care clinics. *Int J Equity Health*. (2018) 17:1–16. doi: 10.1186/s12939-018-0820-2
3. Penman-Aguilar A, Talih M, Huang D, Moonesinghe R, Bouye K, Beckles G. Measurement of health disparities, health inequities, and social determinants of health to support the advancement of health equity. *J Public Health Manage Pract*. (2016) 22(Suppl 1):S33. doi: 10.1097/PHH.0000000000000373
4. Pratt B, de Vries J. Community engagement in global health research that advances health equity. *Bioethics*. (2018) 32:454–63. doi: 10.1111/bioe.12465
5. Curtis E, Jones R, Tipene-Leach D, Walker C, Loring B, Paine SJ, et al. Why cultural safety rather than cultural competency is required to achieve health equity: a literature review and recommended definition. *Int J Equity Health*. (2019) 18:1–17. doi: 10.1186/s12939-019-1082-3
6. Pratt B. Research for Health Justice: an ethical framework linking global health research to health equity. *BMJ Global Health*. (2021) 6:e002921. doi: 10.1136/bmjgh-2020-002921
7. Dubowitz T, Orleans T, Nelson C, May LW, Sloan JC, Chandra A. Creating healthier, more equitable communities by improving governance and policy. *Health Affairs*. (2016) 35:1970–5. doi: 10.1377/hlthaff.2016.0608
8. de Groot N, Bonsel GJ, Birnie E, Valentine NB. Towards a universal concept of vulnerability: broadening the evidence from the elderly to perinatal health using a Delphi approach. *PLoS ONE*. (2019) 14:e0212633. doi: 10.1371/journal.pone.0212633
9. Purkey E, MacKenzie M. Experience of healthcare among the homeless and vulnerably housed a qualitative study: opportunities for equity-oriented health care. *Int J Equity Health*. (2019) 18:1–7. doi: 10.1186/s12939-019-1004-4
10. Kostareva U, Albright CL, Berens EM, Levin-Zamir D, Aringazina A, Lopatina M, et al. International perspective on health literacy and health equity: factors that influence the former Soviet Union immigrants. *Int J Environ Res Public Health*. (2020) 17:2155. doi: 10.3390/ijerph17062155
11. Kneipp SM, Schwartz TA, Drevdahl DJ, Canales MK, Santacroce S, Santos Jr HP, et al. Trends in health disparities, health inequity, and social determinants of health research: a 17-year analysis of NINR, NCI, NHLBI, and NIMHD funding. *Nurs Res*. (2018) 67:231–41. doi: 10.1097/NNR.0000000000000278
12. Liburd LC, Hall JE, Mpofu JJ, Williams SM, Bouye K, Penman-Aguilar A. Addressing health equity in public health practice: frameworks, promising strategies, and measurement considerations. *Annu Rev Public Health*. (2020) 41:417–32. doi: 10.1146/annurev-publhealth-040119-094119
13. Townsend B, Friel S, Freeman T, Schram A, Strazdins L, Labonte R, et al. Advancing a health equity agenda across multiple policy domains: a qualitative policy analysis of social, trade and welfare policy. *BMJ Open*. (2020) 10:e040180. doi: 10.1136/bmjopen-2020-040180
14. Pedrana L, Pamponet M, Walker R, Costa F, Rasella D. Scoping review: national monitoring frameworks for social determinants of health and health equity. *Global Health Action*. (2016) 9:28831. doi: 10.3402/gha.v9.28831
15. Alexandra K, Charlotte L, Jeannine C, Marjorie M, Bernie P. Xpey-relational environments: an analytic framework for conceptualizing indigenous health equity. *Health Promot Chron Dis Prev Can Res Policy Pract*. (2017) 37:395. doi: 10.24095/hpcdp.37.12.01
16. Ward M, Schulz AJ, Israel BA, Rice K, Martenies SE, Markarian E. A conceptual framework for evaluating health equity promotion within community-based participatory research partnerships. *Eval Prog Plann*. (2018) 70:25–34. doi: 10.1016/j.evalprogplan.2018.04.014
17. Eslava-Schmalbach J, Garzón-Orjuela N, Elias V, Reveiz L, Tran N, Langlois E. Conceptual framework of equity-focused implementation research for health programs (EquIR). *Int J Equity Health*. (2019) 18:1–11. doi: 10.1186/s12939-019-0984-4
18. Chandanabhumma PP, Narasimhan S. Towards health equity and social justice: an applied framework of decolonization in health promotion. *Health Promot Int*. (2020) 35:831–40. doi: 10.1093/heapro/daz053
19. Blagev DP, Barton N, Grissom CK, McKee KE, Harrison AM. On the journey toward health equity: data, culture change, and the first step. *NEJM Catal Innovat Care Delivery*. (2021) 2:1–17. doi: 10.1056/CAT.21.0118
20. Davison CM, Ndumbe-Eyoh S, Clement C. Critical examination of knowledge to action models and implications for promoting health equity. *Int J Equity Health*. (2015) 14:1–11. doi: 10.1186/s12939-015-0178-7
21. Mason KA, Willen SS, Holmes SM, Herd DA, Nichter M, Castaneda H, et al. How do you build a "culture of health"? A critical analysis of challenges and opportunities from medical anthropology. *Popul Health Manage*. (2020) 23:476–81. doi: 10.1089/pop.2019.0179
22. Yang H, Lee HJ. Research trend visualization by MeSH terms from PubMed. *Int J Environ Res Public Health*. (2018) 15:1113. doi: 10.3390/ijerph15061113
23. Martinez-Garcia M, Salinas-Ortega M, Estrada-Arriaga I, Hernández-Lemus E, Garcia-Herrera R, Vallejo M. A systematic approach to analyze the social determinants of cardiovascular disease. *PLoS ONE*. (2018) 13:e0190960. doi: 10.1371/journal.pone.0190960
24. Klopfenstein D, Dampier W. Commentary to Gusenbauer and Haddaway 2020: evaluating retrieval qualities of Google scholar and PubMed. *Res Synth Methods*. (2021) 12:126–35. doi: 10.1002/jrsm.1456
25. Kastrin A, Rindflesch TC, Hristovski D. Large-scale structure of a network of co-occurring MeSH terms: statistical analysis of macroscopic properties. *PLoS ONE*. (2014) 9:e102188. doi: 10.1371/journal.pone.0102188
26. Motschall E, Falck-Ytter Y. Searching the MEDLINE literature database through PubMed: a short guide. *Oncol Res Treat*. (2005) 28:517–22. doi: 10.1159/000087186
27. Rivera-Romano LS, Juárez-Cano G, Hernández-Lemus E, Vallejo M, Martinez-Garcia M. Structure of communities in semantic networks of biomedical research on disparities in health and sexism. *Biomédica*. (2020) 40:702–21. doi: 10.7705/biomedica.5182
28. Yan YH, Chien TW. The use of forest plot to identify article similarity and differences in characteristics between journals using medical subject headings terms: a protocol for bibliometric study. *Medicine*. (2021) 100:e24610. doi: 10.1097/MD.0000000000024610
29. Kim J, Bang C, Hwang H, Kim D, Park C, Park S. IMA: Identifying disease-related genes using MeSH terms and association rules. *J Biomed Inform*. (2017) 76:110–23. doi: 10.1016/j.jbi.2017.11.009
30. Wang K, Feng C, Li M, Pei Q, Li Y, Zhu H, et al. A bibliometric analysis of 23,492 publications on rectal cancer by machine learning: basic medical research is needed. *Therapeut Adv Gastroenterol*. (2020) 13:1756284820934594. doi: 10.1177/1756284820934594
31. Kanazawa S, Shimizu S, Kajihara S, Mukai N, Iida J, Matsuda F. Automated recommendation of research keywords from pubmed that suggest the molecular mechanism associated with biomarker metabolites. *Metabolites*. (2022) 12:133. doi: 10.3390/metabo12020133
32. Ilgisonis E, Lisitsa A, Kudryavtseva V, Ponomarenko E. Creation of individual scientific concept-centered semantic maps based on automated

text-mining analysis of PubMed. *Adv Bioinform*. (2018) 2018:4625394. doi: 10.1155/2018/4625394

33. Zolotarev O, Berberova M, Charnine M. A scientometric analysis of publications related to predictive medicine. In: *CEUR Workshop Proceedings* (Moscow) (2020).
34. Shu F, Qiu J, Lariviére V. Mapping the biomedical sciences using Medical Subject Headings: a comparison between MeSH co-assignments and MeSH citation pairs. *J Med Library Assoc*. (2021) 109:441. doi: 10.5195/jmla.2021.1173
35. Wang L, Feng W, Duan J, Liang J. Pharmacovigilance bibliometrics: visualizing thematic development in the category of pharmacology and pharmacy in web of science. *Front Pharmacol*. (2021) 12:731757. doi: 10.3389/fphar.2021.731757
36. Gan J, Cai Q, Galer P, Ma D, Chen X, Huang J, et al. Mapping the knowledge structure and trends of epilepsy genetics over the past decade: a co-word analysis based on medical subject headings terms. *Medicine*. (2019) 98:e16782. doi: 10.1097/MD.0000000000016782
37. Son YJ, Lee SK, Nam S, Shim JL. Exploring research topics and trends in nursing-related communication in intensive care units using social network analysis. *Comput Inform Nurs*. (2018) 36:383–92. doi: 10.1097/CIN.0000000000000444
38. Zhao F, Shi B, Liu R, Zhou W, Shi D, Zhang J. Theme trends and knowledge structure on choroidal neovascularization: a quantitative and co-word analysis. *BMC Ophthalmol*. (2018) 18:86. doi: 10.1186/s12886-018-0752-z
39. Yang A, Lv Q, Chen F, Wang D, Liu Y, Shi W. Identification of recent trends in research on vitamin D: a quantitative and co-word analysis. *Med Sci Monit*. (2019) 25:643. doi: 10.12659/MSM.913026
40. Liu Y, Li X, Ma L, Wang Y. Mapping theme trends and knowledge structures of dignity in nursing: a quantitative and co-word biclustering analysis. *J Adv Nurs*. (2021) 1–10. doi: 10.1111/jan.15097
41. Yang N, Sun L, Tan J. Theme trends and knowledge structure of assisted reproductive technology and birth defects: a quantitative and co-word analysis. *J Obstetr Gynaecol Res*. (2021) 47:1780–8. doi: 10.1111/jog.14702
42. Drieger P. Semantic network analysis as a method for visual text analytics. *Proc Soc Behav Sci*. (2013) 79:4–17. doi: 10.1016/j.sbspro.2013.05.053
43. Mallett R, Hagen-Zanker J, Slater R, Duvendack M. The benefits and challenges of using systematic reviews in international development research. *J Dev Effect*. (2012) 4:445–55. doi: 10.1080/19439342.2012.711342
44. Bartolucci AA, Hillegass WB. Overview, strengths, and limitations of systematic reviews and meta-analyses. In: Chiappelli F, editor. *Evidence-Based Practice: Toward Optimizing Clinical Outcomes*. Berlin; Heidelberg: Springer (2010). p. 17–33. doi: 10.1007/978-3-642-05025-1_2
45. Siqueiros-Garcia JM, Hernández-Lemus E, Garcia-Herrera R, Robina-Galatas A. Mapping the structure and dynamics of genomics-related MeSH terms complex networks. *PLoS ONE*. (2014) 9:e92639. doi: 10.1371/journal.pone.0092639
46. Martinez-Garcia M, Vallejo M, Hernández-Lemus E, Álvarez-Diaz JA. Novel methods of qualitative analysis for health policy research. *Health Res Policy Syst*. (2019) 17:1–13. doi: 10.1186/s12961-018-0404-z
47. Lee K, Jung H. Dynamic semantic network analysis for identifying the concept and scope of social sustainability. *J Clean Product*. (2019) 233:1510–24. doi: 10.1016/j.jclepro.2019.05.390
48. Danowski JA, Yan B, Riopelle K. A semantic network approach to measuring sentiment. *Qual Quan*. (2021) 55:221–55. doi: 10.1007/s11135-020-01000-x
49. Castelblanco G, Guevara J, Mesa H, Sanchez A. Semantic network analysis of literature on public-private partnerships. *J Construct Eng Manage*. (2021) 147:04021033. doi: 10.1061/(ASCE)CO.1943-7862.0002041
50. Téllez ALN. LILACS: solidarity is to share 35 years watching over the visibility of scientifific production in health in the region. *Revista Repertorio de Medicina y Cirugia*. (2021). 30:4-6. doi: 10.31260/RepertMedCir.01217372.1164
51. Jiménez Miranda J. Acceso a MEDLINE y LILACS mediante el MeSH y el DeCS. *Acimed*. (1998) 6:153–62.
52. Morrison H. Directory of open access journals (DOAJ). *Charleston Advisor*. (2017) 18:25–8. doi: 10.5260/chara.18.3.25
53. Hagberg A, Swart P, Chult SD. *Exploring Network Structure, Dynamics, and Function Using NetworkX*. Los Alamos National Lab, Los Alamos, NM (2008).
54. Smoot ME, Ono K, Ruscheinski J, Wang PL, Ideker T. Cytoscape 2.8: new features for data integration and network visualization. *Bioinformatics*. (2011) 27:431–2. doi: 10.1093/bioinformatics/btq675
55. Doncheva NT, Assenov Y, Domingues FS, Albrecht M. Topological analysis and interactive visualization of biological networks and protein structures. *Nat Protoc*. (2012) 7:670–85. doi: 10.1038/nprot.2012.004
56. Shannon P, Markiel A, Ozier O, Baliga NS, Wang JT, Ramage D, et al. Cytoscape: a software environment for integrated models of biomolecular interaction networks. *Genome Res*. (2003) 13:2498–504. doi: 10.1101/gr.1239303
57. Cruz-Ávila HA, Vallejo M, Martinez-Garcia M, Hernández-Lemus E. Comorbidity networks in cardiovascular diseases. *Front Physiol*. (2020) 11:1009. doi: 10.3389/fphys.2020.01009
58. Braun V, Clarke V. Using thematic analysis in psychology. *Qual Res Psychol*. (2006) 3:77–101. doi: 10.1191/1478088706qp063oa
59. Vaismoradi M, Turunen H, Bondas T. Content analysis and thematic analysis: Implications for conducting a qualitative descriptive study. *Nurs Health Sci*. (2013) 15:398–405. doi: 10.1111/nhs.12048
60. Terry G, Hayfield N, Clarke V, Braun V. Thematic analysis. *SAGE Handb Qual Res Psychol*. (2017) 2:17–37. doi: 10.4135/9781526405555.n2
61. Williams DR, Cooper LA. COVID-19 and health equity–a new kind of "herd immunity". *JAMA*. (2020) 323:2478–80. doi: 10.1001/jama.2020.8051
62. Farley JH, Hines J, Lee NK, Brooks SE, Nair N, Brown CL, et al. Promoting health equity in the era of COVID-19. *Gynecol Oncol*. (2020) 158:25–31. doi: 10.1016/j.ygyno.2020.05.023
63. Shapiro SC. *Encyclopedia of Artificial Intelligence*. 2nd ed. New York, NY: John (1992).
64. Borge-Holthoefer J, Arenas A. Semantic networks: structure and dynamics. *Entropy*. (2010) 12:1264–302. doi: 10.3390/e12051264
65. Brachman RJ. What's in a concept: structural foundations for semantic networks. *Int J Man Mach Stud*. (1977) 9:127–52. doi: 10.1016/S0020-7373(77)80017-5
66. Avant ND, Gillespie GL. Pushing for health equity through structural competency and implicit bias education: a qualitative evaluation of a racial/ethnic health disparities elective course for pharmacy learners. *Curr Pharm Teach Learn*. (2019) 11:382–93. doi: 10.1016/j.cptl.2019.01.013
67. Perdomo J, Tolliver D, Hsu H, He Y, Nash KA, Donatelli S, et al. Health equity rounds: an interdisciplinary case conference to address implicit bias and structural racism for faculty and trainees. *MedEdPORTAL*. (2019) 15:10858. doi: 10.15766/mep_2374-8265.10858
68. Raphael JL, Oyeku SO. Implicit bias in pediatrics: an emerging focus in health equity research. *Pediatrics*. (2020) 145:e20200512. doi: 10.1542/peds.2020-0512
69. Leslie KF, Sawning S, Shaw MA, Martin LJ, Simpson RC, Stephens JE, et al. Changes in medical student implicit attitudes following a health equity curricular intervention. *Med Teacher*. (2018) 40:372–8. doi: 10.1080/0142159X.2017.1403014
70. Johnson R, Scott J, Randolph SD. COVID-19 and Black America: the intersection of health equity and the NP workforce. *Nurse Pract*. (2020) 45:11–4. doi: 10.1097/01.NPR.0000696932.97210.37
71. Milam AJ, Furr-Holden D, Edwards-Johnson J, Webb B, Patton III JW, Ezekwemba NC, et al. Are clinicians contributing to excess African American COVID-19 deaths? Unbeknownst to them, they may be. *Health Equity*. (2020) 4:139–41. doi: 10.1089/heq.2020.0015
72. Golden SH, Galiatsatos P, Wilson C, Page KR, Jones V, Tolson T, et al. Approaching the COVID-19 pandemic response with a health equity lens: a framework for academic health systems. *Acad Med*. (2021) 96:1546–52. doi: 10.1097/ACM.0000000000003999
73. Arkfeld CK. Focus: health equity: the sick bias. *Yale J Biol Med*. (2021) 94:517.
74. Fiorini N, Lipman DJ, Lu Z. Cutting edge: towards PubMed 2.0. *eLife*. (2017) 6:e28801. doi: 10.7554/eLife.28801
75. Yousefi-Nooraie R, Shakiba B, Mortaz-Hejri S. Country development and manuscript selection bias: a review of published studies. *BMC Med Res Methodol*. (2006) 6:37. doi: 10.1186/1471-2288-6-37
76. Harris M, Macinko J, Jimenez G, Mullachery P. Measuring the bias against low-income country research: an Implicit Association Test. *Global Health*. (2017) 13:1–9. doi: 10.1186/s12992-017-0304-y

77. Campbell FM. National bias: a comparison of citation practices by health professionals. *Bull Med Library Assoc.* (1990) 78:376.
78. Braveman P. Measuring health equity within countries: the challenge of limited information. *JAMA*. (2002) 288:1650–0. doi: 10.1001/jama.288.13.1650-JMS1002-4-1
79. Drobac P, Morse M. Medical education and global health equity. *AMA J Ethics*. (2016) 18:702–9. doi: 10.1001/journalofethics.2016.18.7.medu1-1607
80. White AA, Logghe HJ, Goodenough DA, Barnes LL, Hallward A, Allen IM, et al. Self-awareness and cultural identity as an effort to reduce bias in medicine. *J Racial Ethnic Health Disparities*. (2018) 5:34–49. doi: 10.1007/s40615-017-0340-6
81. Park HW, Leydesdorff L. Decomposing social and semantic networks in emerging "big data" research. *J Informetr*. (2013) 7:756–65. doi: 10.1016/j.joi.2013.05.004
82. Griffiths TL, Steyvers M. A probabilistic approach to semantic representation. In: *Proceedings of the Annual Meeting of the Cognitive Science Society, Vol. 174*. New York, NY. (2002).
83. Rathore SS, Krumholz HM. Differences, disparities, and biases: clarifying racial variations in health care use. *Am Coll Phys*. (2004) 141:635–8. doi: 10.7326/0003-4819-141-8-200410190-00011
84. Duggan CP, Kurpad A, Stanford FC, Sunguya B, Wells JC. Race, ethnicity, and racism in the nutrition literature: an update for 2020. *Am J Clin Nutr*. (2020) 112:1409–14. doi: 10.1093/ajcn/nqaa341
85. Sexton SM, Richardson CR, Schrager SB, Bowman MA, Hickner J, Morley CP, et al. Systemic racism and health disparities: statement from editors of family medicine journals. *Can Fam Phys*. (2021) 67:13–4. doi: 10.22454/PRiMER.2020.370906
86. Boutain D. Social justice in nursing: a review of literature. In: de Chesnay M, Anderson B, editors. *Caring for the Vulnerable: Perspectives in Nursing Theory, Practice, and Research*. Burlington, MA: Jones & Bartlett Learning (2005). p. 21–9.
87. McDowell MJ, Goldhammer H, Potter JE, Keuroghlian AS. Strategies to mitigate clinician implicit bias against sexual and gender minority patients. *Psychosomatics*. (2020) 61:655–61. doi: 10.1016/j.psym.2020.04.021
88. Fallin-Bennett K. Implicit bias against sexual minorities in medicine: cycles of professional influence and the role of the hidden curriculum. *Acad Med*. (2015) 90:549–52. doi: 10.1097/ACM.0000000000000662
89. Wandschneider L, Batram-Zantvoort S, Razum O, Miani C. Representation of gender in migrant health studies-a systematic review of the social epidemiological literature. *Int J Equity Health*. (2020) 19:1–15. doi: 10.1186/s12939-020-01289-y
90. Hall R, de Antueno C, Webber A. Publication bias in the medical literature: a review by a Canadian Research Ethics Board. *Can J Anesth*. (2007) 54:380–8. doi: 10.1007/BF03022661
91. Graf S, Paolini S. Investigating positive and negative intergroup contact: Rectifying a long-standing positivity bias in the literature. In: Vezzali L, Stathi S, editors. *Intergroup Contact Theory*. New York, NY: Routledge (2016). p. 100–21.
92. van de Vijver FJ, Tanaka-Matsumi J. Multicultural research methods. In: McKay D, editor. *Handbook of Research Methods in Abnormal and Clinical Psychology*. New York, NY: Sage (2008). p. 463–81.
93. Baah FO, Teitelman AM, Riegel B. Marginalization: Conceptualizing patient vulnerabilities in the framework of social determinants of health-An integrative review. *Nurs Inquiry*. (2019) 26:e12268. doi: 10.1111/nin.12268
94. Garg A, Byhoff E, Wexler MG. Implementation considerations for social determinants of health screening and referral interventions. *JAMA Netw Open*. (2020) 3:e200693. doi: 10.1001/jamanetworkopen.2020.0693
95. Hand M, Wren B, Jennifer C, Creel D. Ethical issues surrounding weight bias and stigma in healthcare. *Online J Health Ethics*. (2013) 8:4. doi: 10.18785/ojhe.0802.04
96. Ferryman K, Pitcan M. Fairness in precision medicine. *Data Soc*. (2018) 1:9–14.
97. O'Connor R. Integrating diversity, equity, and inclusion topics into an undergraduate nursing research course. *J Nurs Educ*. (2019) 58:494. doi: 10.3928/01484834-20190719-13
98. Thompson AC. Health equity, diversity, and inclusion. *Hispanic Health Care Int*. (2016) 14:199. doi: 10.1177/1540415316681883
99. Dickerson D, Baldwin JA, Belcourt A, Belone L, Gittelsohn J, Kaholokula JK, et al. Encompassing cultural contexts within scientific research methodologies in the development of health promotion interventions. *Prev Sci*. (2020) 21:33–42. doi: 10.1007/s11121-018-0926-1
100. Baron RJ, Khullar D. *Building Trust to Promote a More Equitable Health Care System*. American College of Physicians (2021). p. 548–9. doi: 10.7326/M20-6984
101. Berwick DM. Improvement, trust, and the healthcare workforce. *BMJ Qual Saf*. (2003) 12:448–52. doi: 10.1136/qhc.12.6.448
102. Lien Ch, Wu JJ, Chen YH, Wang CJ. Trust transfer and the effect of service quality on trust in the healthcare industry. *Manag Serv Qual*. (2014) doi: 10.1108/MSQ-11-2013-0255
103. Erasmus E, Gilson L, Govender V, Nkosi M. Organisational culture and trust as influences over the implementation of equity-oriented policy in two South African case study hospitals. *Int J Equity Health*. (2017) 16:1–14. doi: 10.1186/s12939-017-0659-y
104. Griffith DM, Bergner EM, Fair AS, Wilkins CH. Using mistrust, distrust, and low trust precisely in medical care and medical research advances health equity. *Am J Prev Med*. (2021) 60:442–5. doi: 10.1016/j.amepre.2020.08.019
105. Ward P. Trust and communication in a doctor-patient relationship: a literature review. *Arch Med*. (2018) 3:36. doi: 10.4172/2472-1654.100146
106. DeLemos D, Chen M, Romer A, Brydon K, Kastner K, Anthony B, et al. Building trust through communication in the intensive care unit: HICCC. *Pediatr Crit Care Med*. (2010) 11:378–84. doi: 10.1097/PCC.0b013e3181b8088b
107. Hong H, Oh HJ. The effects of patient-centered communication: exploring the mediating role of trust in healthcare providers. *Health Commun*. (2020) 35:502–11. doi: 10.1080/10410236.2019.1570427
108. Chen X, Hay JL, Waters EA, Kiviniemi MT, Biddle C, Schofield E, et al. Health literacy and use and trust in health information. *J Health Commun*. (2018) 23:724–34. doi: 10.1080/10810730.2018.1511658
109. Paige SR, Krieger JL, Stellefson ML. The influence of eHealth literacy on perceived trust in online health communication channels and sources. *J Health Commun*. (2017) 22:53–65. doi: 10.1080/10810730.2016.1250846
110. Thorne SE, Nyhlin KT, Paterson BL. Attitudes toward patient expertise in chronic illness. *Int J Nurs Stud*. (2000) 37:303–11. doi: 10.1016/S0020-7489(00)00007-9
111. Mannava P, Durrant K, Fisher J, Chersich M, Luchters S. Attitudes and behaviours of maternal health care providers in interactions with clients: a systematic review. *Global Health*. (2015) 11:1–17. doi: 10.1186/s12992-015-0117-9
112. Carel H, Kidd IJ. Epistemic injustice in healthcare: a philosophial analysis. *Med Health Care Philos*. (2014) 17:529–40. doi: 10.1007/s11019-014-9560-2
113. Wong WF, LaVeist TA, Sharfstein JM. Achieving health equity by design. *JAMA*. (2015) 313:1417–8. doi: 10.1001/jama.2015.2434
114. Nelson HD, Cantor A, Wagner J, Jungbauer R, Quinones A, Stillman L, et al. Achieving health equity in preventive services: a systematic review for a national institutes of health pathways to prevention workshop. *Ann Internal Med*. (2020) 172:258–71. doi: 10.7326/M19-3199
115. Purnell TS, Calhoun EA, Golden SH, Halladay JR, Krok-Schoen JL, Appelhans BM, et al. Achieving health equity: closing the gaps in health care disparities, interventions, and research. *Health Affairs*. (2016) 35:1410–5. doi: 10.1377/hlthaff.2016.0158
116. Magwood O, Hanemaayer A, Saad A, Salvalaggio G, Bloch G, Moledina A, et al. Determinants of implementation of a clinical practice guideline for homeless health. *Int J Environ Res Public Health*. (2020) 17:7938. doi: 10.3390/ijerph17217938
117. Feldman CT, Stevens GD, Lowe E, Lie DA. Inclusion of the homeless in health equity curricula: a needs assessment study. *Med Educ Online*. (2020) 25:1777061. doi: 10.1080/10872981.2020.1777061
118. Purkey E, MacKenzie M. Experiences of palliative health care for homeless and vulnerably housed individuals. *J Am Board Fam Med*. (2019) 32:858–867. doi: 10.3122/jabfm.2019.06.190093
119. Binagwaho A. University of Global Health Equity's contribution to the reduction of education and health services rationing. *Int J Health Policy Management*. (2017) 6:427. doi: 10.15171/ijhpm.2017.56
120. Trezona A, Rowlands G, Nutbeam D. Progress in implementing national policies and strategies for health literacy–what have we learned so far?

Int J Environ Res Public Health. (2018) 15:1554. doi: 10.3390/ijerph15071554

121. Nair SC, Satish KP, Sreedharan J, Ibrahim H. Assessing health literacy in the eastern and middle-eastern cultures. *BMC Public Health*. (2016) 16:831. doi: 10.1186/s12889-016-3488-9
122. Guo S, Yu X, Okan O. Moving health literacy research and practice towards a vision of equity, precision and transparency. *Int J Environ Res Public health*. (2020) 17:7650. doi: 10.3390/ijerph17207650
123. Sentell T, Vamos S, Okan O. *Interdisciplinary Perspectives on Health Literacy Research Around the World: More Important Than Ever in a Time of COVID-19, Vol. 17*. Multidisciplinary Digital Publishing Institute (2020). p. 3010. doi: 10.3390/ijerph17093010
124. Sørensen K, Karuranga S, Denysiuk E, McLernon L. Health literacy and social change: exploring networks and interests groups shaping the rising global health literacy movement. *Global Health Promot*. (2018) 25:89–92. doi: 10.1177/1757975918798366
125. Jackson B, Huston P. Advancing health equity to improve health: the time is now. *Health Promot Chronic Dis Prev Can Res Policy Pract*. (2016) 36:17. doi: 10.24095/hpcdp.36.2.01
126. Welch V, Petkovic J, Pardo JP, Rader T, Tugwell P. Interactive social media interventions to promote health equity: an overview of reviews. *Health Promot Chronic Dis Prev Can Res Policy Pract*. (2016) 36:63. doi: 10.24095/hpcdp.36.4.01
127. Kum SS, Northridge ME, Metcalf SS. Using focus groups to design systems science models that promote oral health equity. *BMC Oral Health*. (2018) 18:99. doi: 10.1186/s12903-018-0560-0
128. Javanparast S, Windle A, Freeman T, Baum F. Community health worker programs to improve healthcare access and equity: are they only relevant to low-and middle-income countries? *Int J Health Policy Manage*. (2018) 7:943. doi: 10.15171/ijhpm.2018.53
129. van Hees SG, O'Fallon T, Hofker M, Dekker M, Polack S, Banks LM, et al. Leaving no one behind? Social inclusion of health insurance in low-and middle-income countries: a systematic review. *Int J Equity Health*. (2019) 18:1–19. doi: 10.1186/s12939-019-1040-0
130. Latif A, Waring J, Pollock K, Solomon J, Gulzar N, Choudhary S, et al. Towards equity: a qualitative exploration of the implementation and impact of a digital educational intervention for pharmacy professionals in England. *Int J Equity Health*. (2019) 18:1–11. doi: 10.1186/s12939-019-1069-0
131. Karpf M. The role of academic health centers in addressing health equity and social determinants of health. *Acad Med*. (2019) 94:1273–5. doi: 10.1097/ACM.0000000000002834
132. Heo HH, Jeong W, Che XH, Chung H. A stakeholder analysis of community-led collaboration to reduce health inequity in a deprived neighbourhood in South Korea. *Global Health Promot*. (2020) 27:35–44. doi: 10.1177/1757975918791517
133. Friel S, Townsend B, Fisher M, Harris P, Freeman T, Baum F. Power and the people's health. *Soc Sci Med*. (2021) 2021:114173. doi: 10.1016/j.socscimed.2021.114173
134. Backholer K, Baum F, Finlay SM, Friel S, Giles-Corti B, Jones A, et al. Australia in 2030: what is our path to health for all? *Med J Australia*. (2021) 214:S5–40. doi: 10.5694/mja2.51020
135. Haggerty J, Chin MH, Katz A, Young K, Foley J, Groulx A, et al. Proactive strategies to address health equity and disparities: recommendations from a bi-national symposium. *J Am Board Family Med*. (2018) 31:479–83. doi: 10.3122/jabfm.2018.03.170299
136. Gusenbauer M, Haddaway NR. Which academic search systems are suitable for systematic reviews or meta-analyses? Evaluating retrieval qualities of Google Scholar, PubMed, and 26 other resources. *Res Synth Methods*. (2020) 11:181–217. doi: 10.1002/jrsm.1378
137. Soratto J, Pires DEPd, Friese S. Thematic content analysis using ATLAS.ti software: Potentialities for researches in health. *Revista brasileira de enfermagem*. (2020) 73:e20190250. doi: 10.1590/0034-7167-2019-0250

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: All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

Copyright © 2022 Martínez-García, Villegas Camacho and Hernández-Lemus. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

frontiers | Frontiers in Public Health

ORIGINAL RESEARCH
published: 04 April 2022
doi: 10.3389/fpubh.2022.813608

Complicated Appendicitis Among Adults With and Without Disabilities: A Cross-Sectional Nationwide Study in South Korea

Kyoung Eun Yeob[1†], So Young Kim[1,2,3†], Jong Eun Park[1] and Jong Hyock Park[1,3]*

[1] Institute of Health and Science Convergence, Chungbuk National University, Cheongju, South Korea, [2] Department of Public Health and Preventive Medicine, Chungbuk National University Hospital, Cheongju, South Korea, [3] Harvard T.H. Chan School of Public Health, Harvard University, Boston, MA, United States

OPEN ACCESS

Edited by:
Violeta Alarcão,
University Institute of Lisbon, Portugal

Reviewed by:
Enver Envi Roshi,
University of Medicine, Albania
Athina Tatsioni,
University of Ioannina, Greece

Correspondence:
Jong Hyock Park
jonghyock@gmail.com

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

Specialty section:
This article was submitted to Life-Course Epidemiology and Social Inequalities in Health, a section of the journal Frontiers in Public Health

Received: *12 November 2021*
Accepted: *31 January 2022*
Published: *04 April 2022*

Citation:
Yeob KE, Kim SY, Park JE and Park JH (2022) Complicated Appendicitis Among Adults With and Without Disabilities: A Cross-Sectional Nationwide Study in South Korea. Front. Public Health 10:813608. doi: 10.3389/fpubh.2022.813608

Objective: Appendicitis is usually diagnosed based on a reliable set of signs and symptoms, and can be effectively treated with surgery, with low morbidity and mortality rates. However, appendicitis is often overlooked in vulnerable populations, including people with disabilities. This study compared 10-year trends of complicated appendicitis between South Koreans with a disability, according to disability severity and type, and those without disabilities

Methods: To identify cases of appendicitis, we used the DRG codes in the National Health Information Database of South Korea. Patients with appendicitis were classified in terms of severity based on the DRG codes. Age-standardized incidence rates were calculated for each year during 2008–2017 according to the presence, type, and severity of the disability. Factors associated with complicated appendicitis were examined by multivariate logistic regression using the most recent data (i.e., 2016–2017).

Results: The incidence of complicated appendicitis was higher in people with disabilities, especially those with severe disabilities (26.9 vs. 11.6%). This difference was particularly marked when considering those with a severe disability (aOR = 1.868, 95% CI:1.511–2.309), internal organ problems (aOR = 10.000, 95% CI:5.365–18.638) or a mental disability (aOR = 2.779, 95% CI:1.563–4.939).

Conclusions: The incidence of complicated appendicitis was higher in people with disability than in those without disability in all years. There was a substantial difference in the incidence of complicated appendicitis between the severe disability and non-disabled groups. Among the various disability types, the incidence of complicated appendicitis was highest for major internal organ problems, followed by intellectual or psychological disabilities. Our findings may be explained by barriers to healthcare access among people with disabilities, particularly those with a severe disability, internal organ problem, or mental disability.

Keywords: disability, complicated appendicitis, disparity, healthcare access, Korea

INTRODUCTION

Appendectomy is one of the most common operations performed worldwide, including in South Korea (1). Appendicitis is typically diagnosed according to a reliable set of signs and symptoms, and can be effectively treated with surgery, with low morbidity and mortality rates. However, appendicitis is often overlooked in vulnerable populations, including people with disabilities (2).

A delay in the diagnosis and treatment of some conditions can result in serious adverse outcomes; delayed diagnosis and treatment of appendicitis can result in appendiceal rupture, peritonitis, and death (3). The risk of rupture is negligible within the first 24 h; however, the rupture rate reaches 6% 36 h after the onset of symptoms (4, 5).

It has been suggested that rupture rate of appendicitis could serve as an indicator of access to healthcare (6). In the US, racial/ethnic differences in the rate of poor outcomes of acute appendicitis, such as perforation of the appendix or complicated disease, have been reported (6). Appendectomy is the most common intra-abdominal surgical procedure (6), and has no known behavioral or social risk factors and only one treatment option (appendectomy). Appendectomy should be performed urgently, regardless of the time of day.

Timely and appropriate healthcare plays a key role in wellness, illness prevention, and optimal recovery when illness occurs (7). Several studies have reported healthcare disparities between people with and without disabilities; the former group are more likely to experience a delay in healthcare. These disparities contribute to differences in outcomes, such as mortality (8–11). Such disparities have been reported in studies of women with disabilities being screened for cervical cancer or undergoing dental examinations, as well in studies on the diagnosis and treatment of cancer (12–14).

Although numerous epidemiological studies on complicated appendicitis have been conducted, most focused on children or older adults; relatively few focused on people with disabilities, especially with a long observation period. Previous studies were mainly concerned with the incidence of complicated appendicitis (15–17), differences in the incidence of complicated appendicitis between patients with and without disabilities, and factors affecting complicated appendicitis in people with disabilities. Our target population was people with disabilities diagnosed with complicated appendicitis; the comparison group was patients with complicated appendicitis without disabilities and the outcome variable was the incidence of complicated appendicitis. To explore potential differences in the incidence of complicated appendicitis between patients with and without disabilities, we conducted a cross-sectional study.

MATERIALS AND METHODS

Data Source and Study Subjects

This study used information from the National Health Insurance Service (NHIS) database of the National Health Insurance Sharing Service. The Korean NHIS covers 97% of the Korean population; only Medical Aid beneficiaries in the lowest income bracket are not covered. The NHIS contains information about age, sex, residential area, monthly insurance contributions (a proxy for income status), disability type and severity, and vital statistics. The NHIS claims database enables easy retrieval and analysis of population-based epidemiological data. For this study, population-based medical data for patients of all ages with appendicitis were retrospectively extracted from the NHI claims database from January 2008 to December 2017. We collected information on disability severity and type from Using a disability registry. The database covered 93.8% of the total disabled population as of 2011 (18). Using Korean personal identification numbers, disability severity and type were linked with variables selected from the NHIS claims database. We excluded patients aged <19 years at the time of diagnosis ($n = 84{,}981$), as well as those who had missing data ($n = 1{,}561$) or an appendectomy of unknown severity (i.e., missing codes; $n = 11{,}834$) **(Figure 1)**. During the study period, 6,47,068 patients were screened for eligibility. Finally, the study sample included 9,687 patients with disabilities upon their complicated appendicitis diagnosis and 70,797 patients without disabilities upon the complicated appendicitis diagnosis during a 10-year period from 2008 to 2017.

Definition of Complicated Appendicitis, and Other Variables

The primary study outcome of the rate of complicated appendicitis. Patients with complicated appendicitis were classified based on KDRG codes according to severity. Cases of appendicitis treated with appendectomy were identified using diagnosis-related group (DRG) codes G081–G084. The DRG-based payment system in South Korea is a case-based prospective payment system under which payments are made according to physician-determined diagnoses (19). The Korean Diagnosis Related Group (KDRG) comprises eight surgical diseases and procedures (appendectomy, tonsillectomy, hernia, cesarean section, hysterectomy, eutocia, cataract, and hemorrhoids). Each code was subdivided according to the severity of the complications/comorbidities [Patient Clinical Complexity Level (PCCL): 0 = No Clinical Complexity (CC); 1 = Minor CC; 2 = Moderate CC; 3 = Severe CC]. Severity codes 1–3 were defined as complicated appendicitis. Other variables collected from the NHIS included age, sex, insurance premium, residential area, and comorbidities. Insurance premiums for each household were calculated based on income, property, and automobile taxes (20). Residential area was classified as metropolitan, urban, or rural based on the ZIP code. The subjects were classified into four categories based on the Charlson Comorbidity Index (CCI): 0, 1–2, 3–4, and ≥ 5 (the most severe) (21).

Statistical Analysis

Descriptive statistics were generated on disability status (present or absent) and the type and severity of disability. Age-standardized incidence rates were calculated using 2005 Korean census data as the reference. To examine the association between disability and the incidence of complicated appendicitis, we conducted a series of multivariate logistic regression analyses adjusted for age, income level, place of residence, smoking status, the CCI score, diabetes, hypertension, chronic obstructive

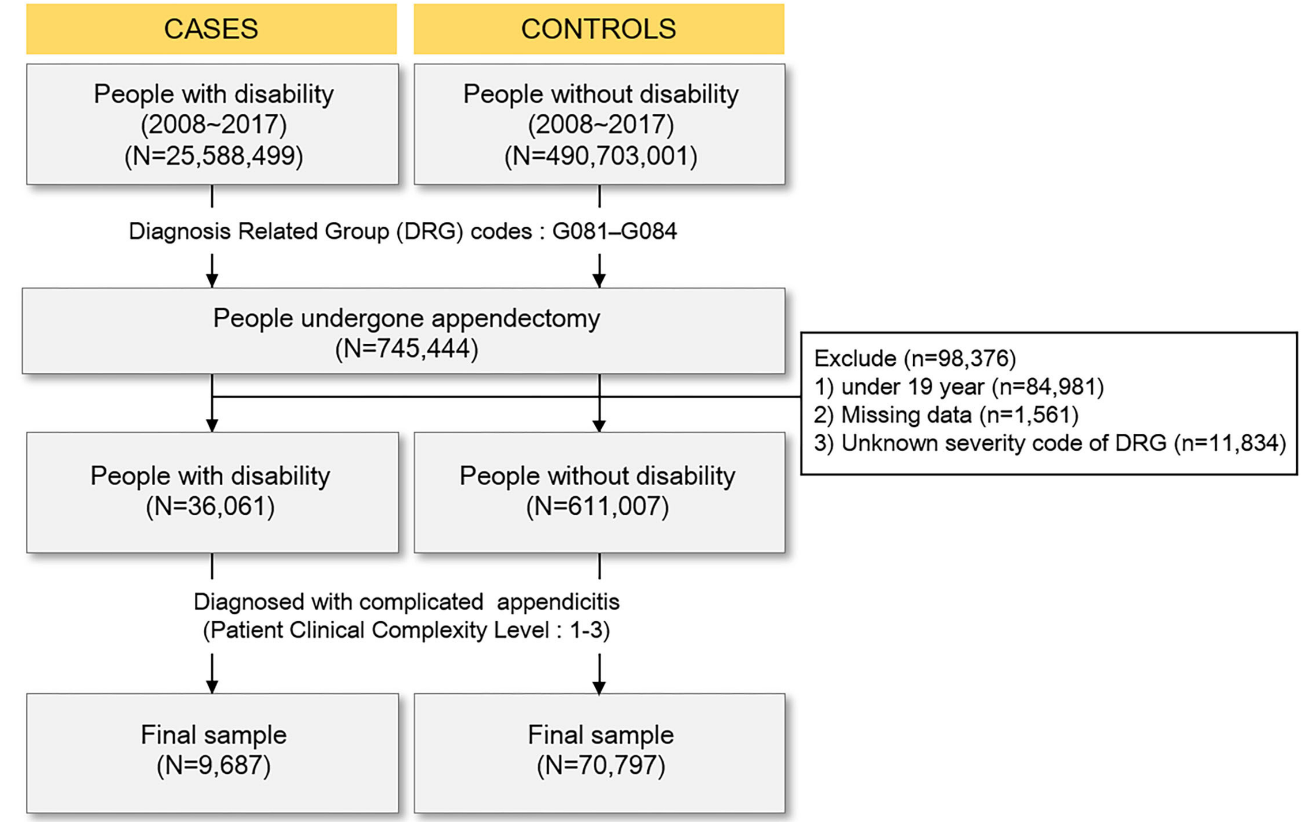


FIGURE 1 | Flow chart.

pulmonary disease, coronary artery disease, obesity, and malignancies (16, 22, 23) using the most recent dataset available (2016–17). These variables were all treated as categorical in the analyses (e.g., "presence" or "absence" of diabetes, hypertension, chronic obstructive pulmonary disease, etc.,). For missing data, we applied the listwise deletion method; although this can lead to the omission of many cases, which affects the statistical power of the tests conducted (24, 25), if the percentage of missing data is very small or the sample is sufficiently large, the power should still be sufficient to detect meaningful effects. This study analyzed the national NHIS claims database, which includes valid and accurate information, especially on socioeconomic status and healthcare utilization, and has very little missing data (26). Given the large size of our sample, which included most of the general population with and without disabilities, listwise deletion did not adversely affect the statistical power. All analyses were performed using SAS software (version 9.3; SAS Institute, Cary, NC, USA), and a p-value < 0.05 was considered significant. This study was approved by the Institutional Review Board of Chungbuk National University (CBNU-202010-HRHR-0717).

RESULTS

Study Participants

Of the non-disabled patients, 50.9% (n = 3,10,833) were male and 49.1% (n = 3,00,174) were female, compared to 61.0% (n = 21,990) and 39.0% (n = 14,071) among the patients with a disability, respectively (<0.0001). Patients with appendicitis and a disability were slightly older than the control subjects (males: 56.8 vs. 42.5 years, females: 62.0 vs. 43.8 years, <0.0001). The economic status of the cohort with disabilities was lower than that of those without a disability. The proportion of patients with medical aid was higher in the disabled group, but the number in the fifth (highest) quartile was larger in the non-disabled group (<0.0001). People with versus without disabilities had more comorbidities and a higher mean CCI score (males: 2.3 vs. 1.0, females: 2.4 vs. 1.1, <0.0001), and were more likely to live in a rural area (<0.0001). In total, 12.2 and 11.0% of patients without a disability (<0.0001), and 27.3 and 26.2% of those with a disability (p = 0.0209), had been diagnosed with complicated appendicitis (**Table 1**).

Incidence of Complicated Appendicitis According to the Presence and Absence of a Disability for 2008–2017

Trends in the crude and age-adjusted incidence rates of complicated appendicitis per 1,00,000 population according to the presence or absence of a disability are shown in **Figure 2**. The age-adjusted incidence of complicated appendicitis according to the presence or absence of a disability decreased gradually from 2008 to 2017, but the incidence was higher in disabled

TABLE 1 | Baseline characteristics of study population in South Korea during 2008–2017 according to disability status and sex.

			People without disability				***P*-value**	**People with disability**				***P*-value**
	Total		**Male**		**Female**			**Male**		**Female**		
	N	**%**	***n***	**%**	***N***	**%**		***n***	**%**	***n***	**%**	
Total	647,068		310,833 (50.9)		300,174 (49.1)			21,990 (61.0)		14,071 (39.0)		
Age												
Mean ± SD	44.0 ± 16.3		42.5 ± 15.4		43.8 ± 16.5			56.8 ± 15.5		62.0 ± 15.6		
95% confidence intervals	43.960–44.040		42.446–42.554		43.741–43.859			56.595–57.005		61.742–62.258		
20–29	144,369	22.3	72,937	23.5	69,805	23.3	<0.0001	1,121	5.1	506	3.6	<0.0001
30–39	151,022	23.3	79,320	25.5	68,608	22.9		2,215	10.1	879	6.2	
40–49	126,246	19.5	63,621	20.5	57,308	19.1		3,676	16.7	1,641	11.7	
50+	225,431	34.8	94,955	30.5	104,453	34.8		14,978	68.1	11,045	78.5	
Income level												
Medical aid and First quartile (lowest)	109,065	16.9	41,723	13.4	55,712	18.6	<0.0001	6,845	31.1	4,785	34.0	<0.0001
Second quartile	104,214	16.1	46,491	15.0	53,398	17.8		2,796	12.7	1,529	10.9	
Third quartile	123,176	19.0	62,511	20.1	55,537	18.5		3,344	15.2	1,784	12.7	
Fourth quartile	139,804	21.6	71,502	23.0	61,700	20.6		4,171	19.0	2,431	17.3	
Fifth quartile (highest)	157,163	24.3	81,356	26.2	67,921	22.6		4,532	20.6	3,354	23.8	
Unknown	13,646	2.1	7,250	2.3	5,906	2.0		302	1.4	188	1.3	
Residence												
Metropolitan	396,448	61.3	190,916	61.4	186,941	62.3	<0.0001	11,412	51.9	7,179	51.0	<0.0001
City	182,953	28.3	88,408	28.4	83,107	27.7		7,069	32.1	4,369	31.0	
Rural	65,386	10.1	30,141	9.7	29,277	9.8		3,466	15.8	2,502	17.8	
Unknown	2,281	0.4	1,368	0.4	849	0.3		43	0.2	21	0.1	
Charlson comorbidity index												
Mean ± SD	1.1 ± 1.7		1.0 ± 1.6		1.1 ± 1.5			2.3 ± 2.5		2.4 ± 2.3		
95% confidence intervals	1.096–1.104		1.094–1.106		1.095–1.105			2.267–2.333		2.362–2.438		
0	314,808	48.7	165,127	53.1	140,737	46.9	<0.0001	5,991	27.2	2,953	21.0	<0.0001
1∼2	242,931	37.5	107,656	34.6	121,709	40.5		8,024	36.5	5,542	39.4	
3∼4	60,023	9.3	25,489	8.2	26,981	9.0		4,359	19.8	3,194	22.7	
≥5	29,306	4.5	12,561	4.0	10,747	3.6		3,616	16.4	2,382	16.9	
Complicated appendicitis												
No	566,584	87.6	272,957	87.8	267,253	89.0	<0.0001	15,988	72.7	10,386	73.8	0.0209
Yes	80,484	12.4	37,876	12.2	32,921	11.0		6,002	27.3	3,685	26.2	

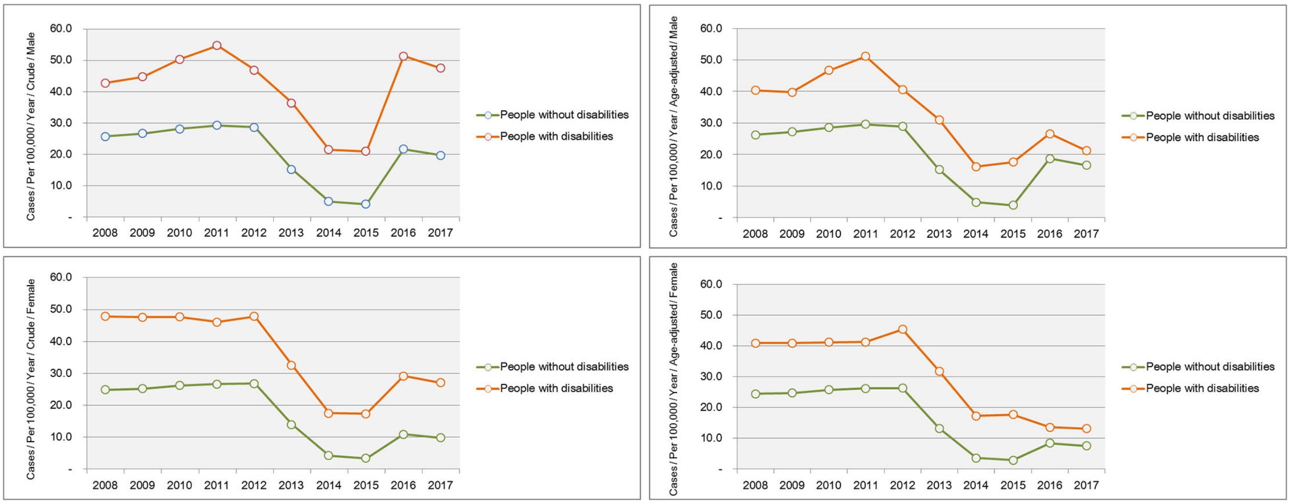


FIGURE 2 | Trends in the incidence of complicated appendicitis according to the presence and absence of a disability for 2008–2017.

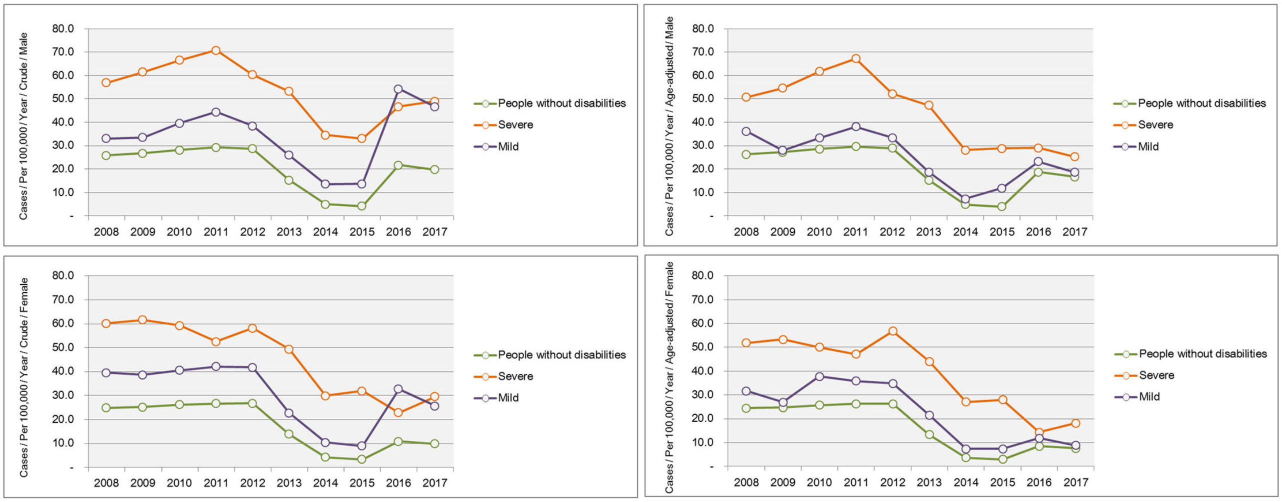


FIGURE 3 | Trends in the incidence of complicated appendicitis according to the severity of a disability for 2008–2017.

than non-disabled males and females in all years. The age-adjusted incidence rates of complicated appendicitis in males and females were 21.2 and 13.2 per 1,00,000 population (2017) in people with disabilities, and 16.6 and 7.5 (2017) per 1,00,000 population in people without disabilities, respectively (see **Supplementary Table 1**).

Incidence of Complicated Appendicitis According to the Severity of Disability for 2008–2017

Trends in the crude and age-adjusted incidence rates of complicated appendicitis per 1,00,000 population according to the severity of disability are shown in **Figure 3**. The greatest group difference in the incidence of complicated appendicitis was seen between the severe disability and non-disabled groups. In particular, in 2017 the age-adjusted incidence for severely disabled males was 1.5 times higher than that of non-disabled people (age-adjusted incidence per 1,00,000 population: 25.2 vs. 16.6) and severely disabled females was 2.4 times higher than that of non-disabled people (age-adjusted incidence per 1,00,000 population: 18.2 vs. 7.35) (see **Supplementary Table 2**).

Incidence of Complicated Appendicitis According to Type of Disability for 2008–2017

Trends in the crude and age-adjusted incidence rates of complicated appendicitis per 1,00,000 population according to the type of disability are shown in **Figure 4**. Among the various disability types, the highest incidence of age-adjusted complicated appendicitis was observed for major internal organ problems (e.g., 94.8 and 50.4 males and females per 1,00,000 population in 2017, respectively), followed by intellectual or

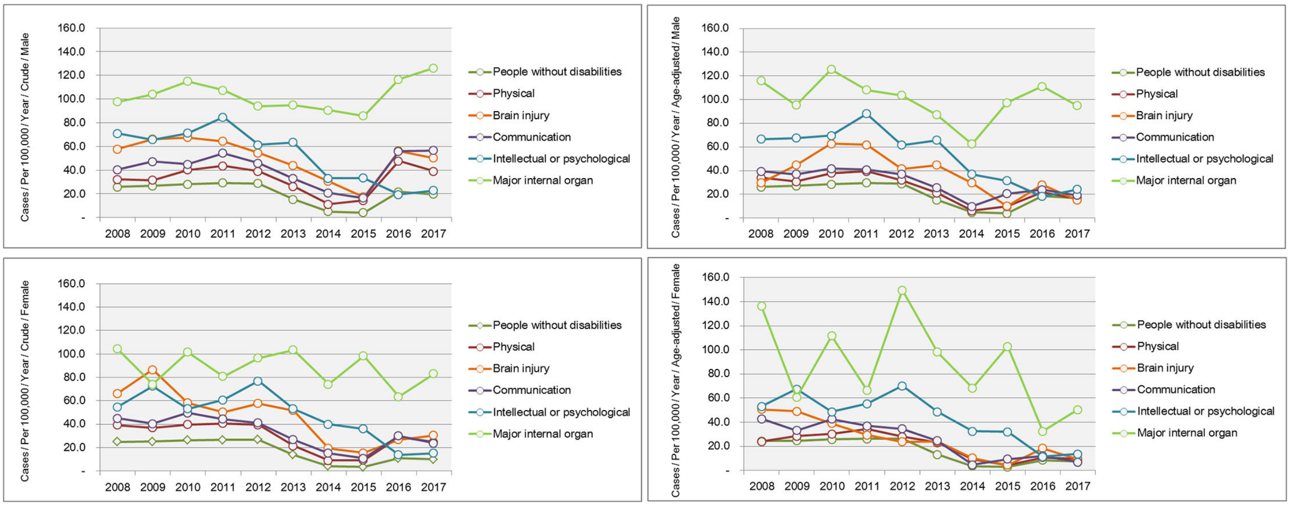


FIGURE 4 | Trends in the incidence of complicated appendicitis according to the type of a disability for 2008–2017.

psychological disabilities (e.g., 24.1 and 13.7 males and females per 1,00,000 population in 2017, respectively) in every year (see **Supplementary Table 3**).

Factors Associated With Complicated Appendicitis During 2016–2017

Males and females with disabilities had a higher incidence of complicated appendicitis than those without a disability [adjusted odds ratio (aORs) = 1.204 and 1.389 for males and females, respectively]. This difference was greater in those with a severe disability (aORs = 1.792 and 1.894 for males and females, respectively). The risk was high for males in the internal organ problems [aOR = 5.581, 95% confidence interval (CI): 3.047–10.222] and intellectual/psychological disabilities (aOR = 2.790, 95% CI: 1.281–6.073) groups, particularly in those with severe internal organ problems (aOR = 10.857, 95% CI: 5.247–22.465) and intellectual/psychological disorders (aOR = 2.818, 95% CI: 1.295–6.136). Similarly, the risk of complicated appendicitis was particularly high in females with severe internal organ problems (aOR = 7.660, 95% CI: 2.297–25.545) (**Table 2**).

DISCUSSION

This is the first study to comprehensively analyze potential disparities in the incidence of complicated appendicitis according to disability status. The strengths of this study included the large number of participants, who were representative of the entire population of South Korea, and the accurate disability diagnoses.

The incidence of complicated appendicitis has been continuously decreasing in both disabled and non-disabled patient groups since 2011. The incidence fell particularly sharply from 2014 to 2015, as also reported in previous studies based on KDRG codes. According to Shin, the ratio of the 1–3 to 0 severity classes decreased 0.49-fold during 2014–2015 among health insurance and medical aid patients. This obviously suggests a decrease in the proportion of high-severity patients, but may also reflect more accurate claims and KDRG code data in association with the introduction of a new payment system in 2013 (27). Further study on this topic is necessary.

In this study, the incidence rates of complicated appendicitis were higher in disabled people compared to those without disabilities. The main drivers of complicated appendicitis are considered to be delayed diagnosis and treatment. Although we could not establish the reasons for late diagnosis and treatment in our disabled group, barriers to healthcare have been suggested in previous studies, including access to facilities, equipment, and transportation (7, 28, 29). Accessing medical facilities is more difficult for disabled than non-disabled people due to physical and transportation problems; this leads to late diagnosis or treatment of appendicitis, which in turn increases the likelihood of complicated appendicitis developing. Efforts are needed to resolve this problem, such as increasing the number of accessible facilities and availability of medical equipment, and providing support for transportation. People with disabilities may also face financial barriers due to the cost of diagnosis and treatment. According to a previous study of people with disabilities in Australia, one in four (24%) delayed going, or did not go, to a hospital; one in five (19%) delayed or did not see a general practitioner, and one in four (27%) did not see a medical specialist because of the cost (30). Thus, financial barriers may lead to delayed diagnosis or treatment of complicated appendicitis. In addition, limitations of healthcare providers themselves (e.g., poor knowledge, negative attitudes, lack of time, and failure to prioritize disabled people in the face of multiple demands), and patient factors (e.g., lack of knowledge and access to the usual source of care), are associated with complicated appendicitis (28, 31, 32). Thus, policies aimed at improving attitudes and access to the usual source of care are needed to decrease the incidence of complicated appendicitis among the disabled.

In our study, the incidence of complicated appendicitis was highest among patients with internal organ problems; these patients have more comorbidities than those with other types

TABLE 2 | Factors associated with complicated appendicitis during 2016–2017.

	Male: OR (95% CI)		Female: OR (95% CI)	
	Model 1*	**Model 2****	**Model 1***	**Model 2****
Disability				
Yes (vs. no)	3.149 (2.951–3.360)	1.204 (1.059–1.369)	3.972 (3.609–4.370)	1.389 (1.135–1.701)
By disability severity				
Severe (vs. no)	4.377 (3.928–4.879)	1.792 (1.394–2.304)	4.905 (4.166–5.775)	1.894 (1.251–2.868)
Mild (vs. no)	2.713 (2.509–2.933)	1.068 (0.924–1.234)	3.625 (3.234–4.064)	1.278 (1.020–1.602)
By disability type				
Physical (vs. no)	2.451 (2.241–2.681)	1.018 (0.863–1.200)	3.414 (2.997–3.889)	1.306 (1.019–1.675)
Brain injury (vs. no)	4.891 (3.985–6.003)	2.102 (1.297–3.406)	4.467 (3.339–5.976)	1.109 (0.522–2.359)
Communication (vs. no)	2.945 (2.605–3.329)	1.133 (0.904–1.419)	3.541 (2.913–4.303)	1.184 (0.774–1.812)
Intellectual or psychological (vs. no)	2.579 (1.937–3.435)	2.790 (1.281–6.073)	3.037 (2.072–4.450)	2.413 (0.984–5.920)
Major internal organ (vs. no)	19.581 (15.230–25.174)	5.581 (3.047–10.222)	25.247 (17.544–36.332)	6.901 (2.731–17.433)
By disability type and severe				
Physical				
Severe (vs. no)	2.867 (2.340–3.514)	1.300 (0.873–1.936)	3.404 (2.340–4.952)	1.116 (0.514–2.421)
Mild (vs. no)	2.370 (2.147–2.616)	0.973 (0.814–1.164)	3.415 (2.976–3.920)	1.329 (1.024–1.725)
Brain injury				
Severe (vs. no)	4.728 (3.555–6.288)	1.777 (0.85–3.715)	4.330 (2.949–6.359)	1.914 (0.696–5.266)
Mild (vs. no)	5.070 (3.783–6.795)	2.390 (1.269–4.501)	4.660 (2.993–7.257)	0.570 (0.173–1.884)
Communication				
Severe (vs. no)	3.014 (2.361–3.847)	1.132 (0.713–1.797)	3.461 (2.403–4.986)	1.598 (0.692–3.691)
Mild (vs. no)	2.922 (2.539–3.363)	1.135 (0.879–1.467)	3.572 (2.841–4.491)	1.074 (0.658–1.755)
Intellectual or psychological				
Severe (vs. no)	2.593 (1.947–3.453)	2.818 (1.295–6.136)	3.037 (2.072–4.450)	2.407 (0.981–5.908)
Mild (vs. no)				
Major internal organ				
Severe (vs. no)	27.109 (19.863–36.999)	10.857 (5.247–22.465)	32.041 (20.639–49.743)	7.660 (2.297–25.545)
Mild (vs. no)	8.430 (5.291–13.432)	1.303 (0.474–3.582)	13.665 (6.899–27.064)	5.947 (1.444–24.495)

**Crude.*

***Adjusted for age, Income level, area of residence, smoking, obesity, CCI, diabetes, hypertension, COPD, coronary artery disease, and any malignancy.*

of disabilities (14, 33, 34). The CCI score predicts mortality due to acute appendicitis. A CCI score > 5 (OR $= 52.45$, $p < 0.05$) was shown to be an independent predictor of mortality due to acute appendicitis (22). Another study of perforated appendicitis in Asians reported that the risk of perforation was higher in patients with one or more comorbidities (15). Therefore, the presence of serious comorbidities is associated with a worse prognosis even for a relatively benign disease, and even in the absence of complications. Patients with a disability due to renal failure have a high incidence of complicated appendicitis. Patients on long-term dialysis undergoing non-emergent procedures are at high risk for complications; an operative mortality rate of 13% has been reported (34). Patients with a disability due to renal failure lack adequate kidney function, so must rely on dialysis to regulate fluid and electrolyte balance, as well as the metabolism of drugs and toxins (35). These problems make postoperative and intraoperative monitoring of a disability due to renal failure challenging, and limit the pharmacological options for surgeons and anesthesiologists (35). As a result, patients with a disability due to renal failure are at increased risk of morbidity and mortality in association with operative procedures.

Our patients with intellectual or psychological disabilities had a high incidence of complicated appendicitis. These results are consistent with previous studies showing that patients with severe intellectual disabilities have more comorbidities than the general population (36), and are susceptible to delayed diagnosis, adverse surgical outcomes, impaired communication, pain and adverse drug reactions (37). Lin reported that surgical patients with an intellectual disability are at higher risk of many complications compared to the general population, including acute renal failure (OR = 3.81, 95% CI: 2.28–6.37), pneumonia (OR = 2.01, 95% CI: 1.61–2.49), postoperative bleeding (OR = 1.35, 95% CI: 1.09–1.68), and septicemia (OR = 2.43, 95% CI: 1.85–3.21) (38). These findings show that strategies are needed to reduce postoperative adverse outcomes in this population.

Our study had several limitations. First, it was retrospective, so we were unable to collect data on all factors that may have affected the outcomes, such as clinical data (e.g., fever, white blood cell count, type of appendicitis, and type of surgery). We believe that adjusting for other covariates in the multivariate model would be sufficient to address the confounding effect of differences in patient characteristics between people with and without disabilities, in terms of risk factor identification. Matching the subjects could prevent such confounding. Second, we could not ascertain why some patients were more at risk of complicated appendicitis, where potential reasons include patient or family refusal to undergo treatment, economic/transportation problems, or clinical decision-making. Further studies using other research methods and statistical analyses (e.g., calculation of absolute and relative risk), including patient surveys and interviews, are required to precisely determine how these factors affect the incidence of complicated appendicitis. Third, although we included subjects with the DRG code for appendectomy, whether they were actually diagnosed with appendicitis was unclear, and patients who did not undergo surgery (e.g., those treated with antibiotics only) were not included (although the number of such patients was small). However, the main treatment for appendicitis is appendectomy; previous studies reported that more than 96% of appendicitis patients underwent this surgery (39). Therefore, most of the patients in this study likely had appendicitis.

CONCLUSION

Our findings indicate disparities in access to healthcare between non-disabled and disabled populations, particularly for those with severe or mental disabilities, or internal organ problems. Although the disparity might in part be due to clinical decision-making, unequal access to healthcare for people with disabilities is unjustifiable. Public health policies should focus on people with disabilities to reduce disparities in health outcomes. Healthcare professionals, as well as people with disabilities and their families, should be educated to improve attitudes, and regarding the need for equal access to diagnosis and treatment.

DATA AVAILABILITY STATEMENT

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

AUTHOR CONTRIBUTIONS

KEY, SYK, JEP, and JHP: conceptualization. JEP and JHP: data curation. JEP: formal analysis. KEY, SYK, and JHP: funding acquisition. JEP and KEY: investigation and methodology. SYK: project administration. JHP: resources. KEY and SYK: software, supervision, validation, visualization, roles/writing—original draft, and writing—review and editing. All authors contributed to the article and approved the submitted version.

FUNDING

This work was supported by National Research Foundation of Korea (NRF) grants funded by the Ministry of Education (Nos. 2020R1I1A1A01054268 and 2019R1D1A3A03103862) and by the Korean Government (MSIT) (No. 2019R1A2C1087507).

ACKNOWLEDGMENTS

We thank the Korean National Health Insurance Services for their provision of data.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2022.813608/full#supplementary-material

REFERENCES

1. Service NHI. *Main Surgery Statistical Yearbook 2017*. Wonju: Service NHI (2017). p. 1–518.
2. Strauther GR, Longo WE, Virgo KS, Johnson FE. Appendicitis in patients with previous spinal cord injury. *Am J Surg.* (1999) 178:403–5. doi: 10.1016/S0002-9610(99)00207-X
3. Bickell N, Siu A. Why do delays in treatment occur? Lessons learned from ruptured appendicitis. *Health Serv Res.* (2001) 36:1–5.
4. Cappendijk V, Hazebroek F. The impact of diagnostic delay on the course of acute appendicitis. *Arch Dis Child.* (2000) 83:64–6. doi: 10.1136/adc.83.1.64
5. Nance ML, Adamson WT, Hedrick HL. Appendicitis in the young child: a continuing diagnostic challenge. *Pediatr Emerg Care.* (2000) 16:160–2. doi: 10.1097/00006565-200006000-00005
6. Jablonski KA, Guagliardo MF. Pediatric appendicitis rupture rate: a national indicator of disparities in healthcare access. *Popul Health Metr.* (2005) 3:1–9. doi: 10.1186/1478-7954-3-4
7. Horner-Johnson W, Dobbertin K, Lee JC, Andresen EM. Disparities in health care access and receipt of preventive services by disability type: analysis of the medical expenditure panel survey. *Health Serv Res.* (2014) 49:1980–99. doi: 10.1111/1475-6773.12195
8. Chevarley FM, Thierry JM, Gill CJ, Ryerson AB, Nosek MA. Health, preventive health care, and health care access among women with disabilities in the 1994–1995 national health interview survey, supplement on disability. *Women's Health Issues.* (2006) 16:297–312. doi: 10.1016/j.whi.2006.10.002
9. Lee JC, Hasnain-Wynia R, Lau DT. Delay in seeing a doctor due to cost: disparity between older adults with and without disabilities in the United States. *Health Serv Res.* (2012) 47:698–720. doi: 10.1111/j.1475-6773.2011.01346.x
10. McGinnis JM, Williams-Russo P, Knickman JR. The case for more active policy attention to health promotion. *Health Aff.* (2002) 21:78–93. doi: 10.1377/hlthaff.21.2.78
11. Drum CE, Krahn G, Culley C, Hammond L. Recognizing and responding to the health disparities of people with disabilities. *Californian J Health Promot.* (2005) 3:29–42. doi: 10.32398/cjhp.v3i3.647
12. Horner-Johnson W, Dobbertin K, Beilstein-Wedel E. Disparities in dental care associated with disability and race and ethnicity. *J Am Dent Assoc.* (2015) 146:366–74. doi: 10.1016/j.adaj.2015.01.024
13. Shin DW, Lee JW, Jung JH, Han K, Kim SY, Choi KS, et al. Disparities in cervical cancer screening among women with disabilities: a national database study in South Korea. *J Clin Oncol.* (2018) 36:2778–86. doi: 10.1200/JCO.2018.77.7912
14. Shin DW, Cho JH, Noh JM, Han H, Han K, Park SH, et al. Disparities in the diagnosis and treatment of lung cancer among people with disabilities. *J Thorac Oncol.* (2019) 14:163–75. doi: 10.1016/j.jtho.2018.10.158
15. Lin K-B, Lai KR, Yang N-P, Chan C-L, Liu Y-H, Pan R-H, et al. Epidemiology and socioeconomic features of appendicitis in Taiwan: a 12-year population-based study. *World J Emerg Surg.* (2015) 10:42. doi: 10.1186/s13017-015-0036-3
16. Paquette IM, Zuckerman R, Finlayson SR. Perforated appendicitis among rural and urban patients: implications of access to care. *Ann Surg.* (2011) 253:534–8. doi: 10.1097/SLA.0b013e3182096d68
17. Akhtar-Danesh G-G, Doumouras AG, Flageole H, Hong D. Geographic and socioeconomic predictors of perforated appendicitis: a national Canadian cohort study. *J Pediatr Surg.* (2019) 54:1804–8. doi: 10.1016/j.jpedsurg.2018.10.065
18. Welfare MoHa. *National Survey of the Disabled Persons.* (2013). Available online at: https://kosis.kr/statHtml/statHtml.do?orgId=117&tblId=DT_11732S0103&conn_path=I2 (accessed February 14, 2021).
19. Kim Y-K. Forecasting the future reimbursement system of Korean national health insurance: a contemplation focusing on global budget and Neo-KDRG-based payment systems. *J Korean Med Sci.* (2012) 27:S25–32. doi: 10.3346/jkms.2012.27.S.S25
20. Yoon TH, Lee S-Y, Kim C-W, Kim SY, Jeong B-G, Park H-K. Inequalities in medical care utilization by South Korean cancer patients according to income: a retrospective cohort study. *Int J Health Serv.* (2011) 41:51–66. doi: 10.2190/HS.41.1.d
21. Birim Ö, Kappetein AP, Bogers AJ. Charlson comorbidity index as a predictor of long-term outcome after surgery for nonsmall cell lung cancer. *Eur J Cardiothorac Surg.* (2005) 28:759–62. doi: 10.1016/j.ejcts.2005.06.046
22. Sartelli M, Baiocchi GL, Di Saverio S, Ferrara F, Labricciosa FM, Ansaloni L, et al. Prospective observational study on acute appendicitis worldwide (POSAW). *World J Emerg Surg.* (2018) 13:19. doi: 10.1186/s13017-018-0179-0
23. Naderan M, Babaki AES, Shoar S, Mahmoodzadeh H, Nasiri S, Khorgami Z. Risk factors for the development of complicated appendicitis in adults. *Ulus Cerrahi Derg.* (2016) 32:37–42. doi: 10.5152/UCD.2015.3031
24. Olinsky A, Chen S, Harlow L. The comparative efficacy of imputation methods for missing data in structural equation modeling. *Eur J Oper Res.* (2003) 151:53–79. doi: 10.1016/S0377-2217(02)00578-7
25. Roth PL. Missing data: a conceptual review for applied psychologists. *Pers Psychol.* (1994) 47:537–60. doi: 10.1111/j.1744-6570.1994.tb01736.x
26. Boo S, Lee J, Oh H. Cost of care and pattern of medical care use in the last year of life among long-term care insurance beneficiaries in South Korea: using national claims data. *Int J Environ Res Public Health.* (2020) 17:9078. doi: 10.3390/ijerph17239078
27. Affairs KKIfHaS. *Evaluation of New Diagnosis-Related Group System (DRG) Pilot Project*. Sejong: Affairs KKIfHaS (2020). p. 300.
28. Wei W, Findley PA, Sambamoorthi U. Disability and receipt of clinical preventive services among women. *Women's Health Issues.* (2006) 16:286–96. doi: 10.1016/j.whi.2006.09.002
29. Iezzoni L, Pendo E. Accessibility of medical diagnostic equipment-implications for people with disability. *New England Journal of Medicine.* (2018) 378:1371–3. doi: 10.1056/NEJMp1800606
30. Health AIo, Welfare. *Access to Health Services by Australians with Disability 2012*. Canberra: AIHW (2015).
31. Ramjan L, Cotton A, Algoso M, Peters K. Barriers to breast and cervical cancer screening for women with physical disability: a review. *Women Health.* (2016) 56:141–56. doi: 10.1080/03630242.2015.1086463
32. World Health Organization. *Promoting Sexual and Reproductive Health for Persons with Disabilities: WHO/UNFPA Guidance Note*. Geneva: World Health Organisation (2009).
33. Sharma A, Zhao X, Hammill BG, Hernandez AF, Fonarow GC, Felker GM, et al. Trends in noncardiovascular comorbidities among patients hospitalized for heart failure: insights from the get with the guidelines–heart failure registry. *Circ Heart Fail.* (2018) 11:e004646. doi: 10.1161/CIRCHEARTFAILURE.117.004646
34. Gajdos C, Hawn MT, Kile D, Robinson TN, Henderson WG. Risk of major nonemergent inpatient general surgical procedures in patients on long-term dialysis. *JAMA Surg.* (2013) 148:137–43. doi: 10.1001/2013.jamasurg.347
35. Smith MC, Boylan MR, Tam SF, Lee R, Alfonso AE, Sugiyama G. End-stage renal disease increases the risk of mortality after appendectomy. *Surgery.* (2015) 158:722–7. doi: 10.1016/j.surg.2015.03.064
36. Bittles AH, Petterson BA, Sullivan SG, Hussain R, Glasson EJ, Montgomery PD. The influence of intellectual disability on life expectancy. *J Gerontol A Biol Sci Med Sci.* (2002) 57:M470–2. doi: 10.1093/gerona/57.7.M470
37. Khalid K, Al-Salamah SM. Surgery for acute abdominal conditions in intellectually-disabled adults. *ANZ J Surg.* (2006) 76:145–8. doi: 10.1111/j.1445-2197.2006.03621.x
38. Lin J-A, Liao C-C, Chang C-C, Chang H, Chen T-L. Postoperative adverse outcomes in intellectually disabled surgical patients: a nationwide population-based study. *PLoS ONE.* (2011) 6:e26977. doi: 10.1371/journal.pone.0026977

39. Becker P, Fichtner-Feigl S, Schilling D. Clinical management of appendicitis. *Visceral medicine.* (2018) 34:453–8. doi: 10.1159/000494883

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: All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

Copyright © 2022 Yeob, Kim, Park and Park. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

frontiers | Frontiers in Public Health

ORIGINAL RESEARCH
published: 15 April 2022
doi: 10.3389/fpubh.2022.812700

Beliefs, Perceptions, and Behaviors Regarding Chronic Respiratory Diseases of Roma in Crete, Greece: A Qualitative FRESH AIR Study

Marilena Anastasaki [1*†], *Egid M. van Bree* [2†], *Evelyn A. Brakema* [2], *Ioanna Tsiligianni* [1], *Dimitra Sifaki-Pistolla* [1], *Vasiliki E. Chatzea* [1], *Matty C. Crone* [2], *Andreas Karelis* [1], *Rianne M. J. J. van der Kleij* [2], *Charlotte C. Poot* [2], *Ria Reis* [2], *Niels H. Chavannes* [2] and *Christos Lionis* [1] *on behalf of the FRESH AIR collaborators*

[1] Clinic of Social and Family Medicine, School of Medicine, University of Crete, Heraklion, Greece, [2] Department of Public Health and Primary Care, Leiden University Medical Centre, Leiden, Netherlands

OPEN ACCESS

Edited by:
Violeta Alarcão,
University Institute of Lisbon, Portugal

Reviewed by:
Ipsita Nita Chaudhuri,
The American University of Paris, France
Olga Magano,
Universidade Aberta, Portugal & CIES_Iscte, Portugal

***Correspondence:**
Marilena Anastasaki
anastasakimarilena@yahoo.gr

† These authors have contributed equally to this work

Specialty section:
This article was submitted to Life-Course Epidemiology and Social Inequalities in Health, a section of the journal Frontiers in Public Health

Received: 10 November 2021
Accepted: 14 March 2022
Published: 15 April 2022

Citation:
Anastasaki M, van Bree EM, Brakema EA, Tsiligianni I, Sifaki-Pistolla D, Chatzea VE, Crone MC, Karelis A, van der Kleij RMJJ, Poot CC, Reis R, Chavannes NH and Lionis C (2022) Beliefs, Perceptions, and Behaviors Regarding Chronic Respiratory Diseases of Roma in Crete, Greece: A Qualitative FRESH AIR Study. Front. Public Health 10:812700. doi: 10.3389/fpubh.2022.812700

Background: The global burden of chronic respiratory diseases (CRDs) disproportionally affects Roma populations. Health interventions addressing CRD among Roma or other vulnerable groups often fail to be effective, as their implementation strategy misaligns with the local context. To design context-driven strategies, we studied CRD-related beliefs, perceptions, and behaviors among a Greek Roma population, focussing on asthma and COPD.

Methods: For this qualitative study in Crete, Greece, we used a Rapid Assessment Process. We conducted interviews and focus groups with purposively selected Roma community members (CMs), key informants (KIs) and healthcare professionals (HPs) serving the population. Data were triangulated using observations of households and clinical consultations. Key themes were identified using Thematic Content Analysis. The Health Belief Model, the Explanatory Model of Illness, and the Theory of Planned Behavior that are complementary is some aspects, guided our methodology with the several variables from them to be integrated to better understand CRD risk preventative behavior.

Results: We conducted six focus groups, seven interviews and 13 observations among 15 CMs, four KIs, and three HPs. Five themes emerged: (1) Poor CRD-awareness (smoking and household air pollution were perceived as harmful, but almost exclusively associated with acute rather than chronic symptoms); (2) Low perceived susceptibility to CRD (and CMs tended to ignore respiratory symptoms); (3) High risk exposure (smoking was common, and air pollution was perceived inevitable due to financial constraints); (4) Healthcare seeking (healthcare was sought only for persistent, severe symptoms, daily needs were a priority); (5) Perceived barriers/facilitators to care (health illiteracy, perceived discrimination and financial constraints were main barriers; established trust the main facilitator).

Conclusion: These five themes highlight that strategies to tackle CRD in the studied Roma setting require a multilevel approach: bridging awareness gaps at the population level, providing resources to enhance the adoption of healthy behaviors, and fighting discrimination at the societal level, whilst establishing trusted relationships at the local level. Similar methodologies to address local context may strengthen the implementation of effective interventions for similarly vulnerable and/or low-resource populations.

Keywords: Roma, chronic respiratory diseases, health literacy, culture, trust, health behavior, barriers to care, low-resource settings

INTRODUCTION

Chronic respiratory diseases (CRDs) are a major burden to global health (1, 2). According to the World Health Organization, the majority of asthma and more than 90% of chronic obstructive pulmonary disease (COPD)-related deaths occur in low-resource settings (3, 4). The major CRD risk factors are tobacco smoking, household air pollution (HAP), and occupational exposure (2, 5). HAP is caused by biomass fuel burning for cooking or heating, and results in about 3 million deaths annually (6, 7).

Particularly vulnerable to CRDs are Roma (8–10), Europe's largest ethnic minority (11). Especially in older age groups, Roma experience more frequent activity-limiting disabilities and health problems compared to non-Roma (10, 12, 13). These poor health outcomes have been linked to poverty, deprivation, marginalization, and discrimination across multiple European countries (14–16). High rates of tobacco smoking are well-documented and an increased risk of HAP exposure has been suggested (10, 14, 15, 17). Similarly, Roma in Greece were previously found to be at risk of a low health-related quality of life strongly associated with chronic diseases and a reduced lung function due to poor living conditions and exposure to tobacco smoke (16, 18). In Greece, 56.5% of Roma were habitual smokers and 22.0% lived in shanty towns (19). Although recent Roma-specific data in Greece are unavailable, the overall burden and prevalence of CRDs have increased in the wake of austerity measures following the financial crisis (20, 21).

Despite national and European efforts to improve access to care for Roma, health improvements remain limited (22). Previous census among Roma in Greece have indicated a low education level, high uninsured rate, and difficulty in accessing healthcare (23, 24). Aggravated poverty and increased discrimination result from the coronavirus pandemic and measures taken to contain it, disproportionately affecting Roma communities (25, 26). According to the recently published "Roma strategic framework" of the European Commission, 80.0% of Roma are still at risk of poverty and 61.0% experience housing deprivation (27). In terms of general health status, a recent study of over 500 adults living in Roma settlements throughout Greece indicated that, although the majority of participants perceived their health as good/very good, about half had been diagnosed with at least one chronic disease. Socio-economic determinants of health including sex, age and poverty indicators were significantly associated with self-perceived health status and with the presence of a chronic disease (28).

Health interventions developed in affluent settings often fail to be successful when implemented in low-resource settings, such as Roma communities. Evidence is scarce regarding facilitators and obstacles of implementation processes (29). Compatibility with the local context, in particular with health beliefs, attitudes, and behaviors, is crucial for successful implementation of health interventions (30–32). To better understand Roma's vulnerability to CRDs and to develop context-appropriate interventions, insight into their health beliefs, attitudes, and behavior is necessary (15, 33). Therefore, the aim of this study was to explore beliefs, perceptions, and behaviors related to CRDs, and the experienced barriers to healthcare among Roma in Crete, Greece.

MATERIALS AND METHODS

Study Design

This was a qualitative study. The methodological approach was based on the "SETTING" tool for context mapping of health interventions in low-resource settings (34), starting with co-setting study priorities with local stakeholders. Our multidisciplinary team was composed of external and local experts and members of the Roma population. We conducted interviews and focus group discussions (FGDs) with Roma community members (CMs), healthcare professionals (HPs) serving the population and key informants (KIs). Observations of households and clinical consultations were additionally conducted for data triangulation. This study was part of the European Horizon 2020 project "FRESH AIR" (35) (trial registration number NTR5759), targeting the prevention, diagnosis, and treatment of CRDs in low-resource settings. As part of FRESH AIR this study was also conducted in other low-resource populations of Greece, namely in rural settings. This paper follows the COREQ guidelines for reporting qualitative research (36) (**Appendix 1**).

Setting

The study took place in the largest Roma camp in Crete with approximately 580 residents. Selection of the study setting was based on convenient preference of a site where the research team or our engaged stakeholders already had established relationships with the communities. Living conditions in the camp are characterized by poor housing and hygiene. Most inhabitants live in improvised constructions (tents and shacks of low-quality materials), while there is no sewage system or electrical network. Although Roma have been tolerated to reside on location for over 10 years, the camp is not recognized as a legal place of residence. According to a census by the local support center (below), half of the adult population has not attended school. Access to the camp by non-Roma is socially challenging, making the population very hard-to-reach. **Appendix 2** includes detailed information about the setting and population of this study.

The Support Center for Roma and Minority Groups (SCRMG) is a municipal service that provides basic primary and social care to the Roma community which is subject of this study. Continuous support by medical personnel is lacking, partially due to fluctuating funding. The SCRMG is located in close vicinity to the camp, but regular healthcare services are distant (7–10 km to hospitals and 35 km to the nearest primary care facility).

In Greece, 96.0% of Roma have been reported to live below the country's at-risk-of-poverty threshold, compared with 22.0% of the general population (24). The risk of poverty has not been found to be substantially different across neighborhoods. In Roma youth, 81.0% of women and 38.0% of men report neither work or education as their main activity, compared to 17.0% of Greek youth. Living in dwellings with e.g., damp walls or rot in window frames was documented for 37.0% of Roma compared to 13.7% of the general population (24). Settlements have previously been described to be located on vacant sites with limited to no access of basic amenities and risk of compulsory removal (37). In addition, a strong sense of community and behaviors of introversion and resistance to influences from the outside were mentioned. Hardly any scientific reports exist comparing ways of life and functioning of local support centers between different Roma settlements in Greece. One previous study reported 76% of Roma in settlements to live in permanent houses and found extensiveness of available services between support centers to vary considerably (38).

On a European scale, it is difficult to make a direct comparison of living conditions and socio-cultural values of the study population with other Roma communities given the strong heterogeneity of the European Roma minority (19). However, a general characterization can be made. Recent reporting by the EU Agency for Fundamental Rights has documented 80.0% of Roma to live below their country's at-risk-of-poverty threshold, 10.0% to live in housing without access to electricity, 53.0% of Roma children to participate in compulsory primary education, and only 25% of Roma reporting to be (self)-employed (24). Compared with the general population, more Roma women report "domestic work" as their main activity.

Participants

Participants were selected by a combination of purposive and convenience sampling. As the study aim was explorative, we aimed for a diverse sample in terms of sex, age and background. We included:

- CMs: Any Adult Camp Resident (≥18 years old).
- HPs: Any health professional working with Roma CMs in the camp.
- KIs: Any relevant stakeholder with either in-depth knowledge or an overview of beliefs, perceptions and behaviors of the camp population.

Apart from the age criterion (≥18 years old), no other inclusion criteria were employed for sample selection. People living outside the camp and not in direct contact with the population, or people unable to participate due to physical or mental disabilities, were excluded.

For participant inclusion, we first engaged with the SCRMG. Over the years, these professionals have built trust with the Roma population. SCRMG professionals were firstly included as HPs or KIs in the study. Afterwards, they accompanied us inside the camp to facilitate trust from the CMs. A mediator, who was a member of the Roma community and SCRMG collaborator, ensured smooth and effective access to residences. Sample size was intended to be guided by data saturation, yet in execution dependent on CM's willingness to participate and time availability of SCRMG-professionals (see discussion).

Theoretical Framework

Our theoretical framework was based on a combination of three health behavior models: the Health Belief Model, the Explanatory Model of Illness, and the Theory of Planned Behavior (39–41). The framework consisted of elements including perceptions of CRD identity, susceptibility, barriers toward behavioral change and risk reduction, help seeking behavior by CMs, and helping behavior by HPs. All study materials (topic guides, observation forms, surveys) were guided by this framework (**Figure 1**, **Appendix 3** detailing the reasoning behind development and use).

Data Collection

We simultaneously conducted semi-structured interviews, FGDs and observations in September 2016, using the Rapid Assessment Process (RAP) (42). Our initial intention was to perform a mixed-methods study including questionnaires to quantify CRD risk factor exposure and frequency of perceived potential causes, yet in execution found willingness to participate, time availability and practical aspects to strongly influence our ability to collect responses. Therefore, during the RAP, we decided to use the obtained questionnaires for data triangulation rather than analyse them separately. Within the RAP, researchers immersed into the community for 2 days in total and collected data in a concise and time-efficient way. CM interviews, FGDs, and household observations took place inside the camp. Interviews with HPs and KIs and observations of clinical consultations were conducted in SCRMG facilities. Two Greek and one Dutch interviewer conducted each activity. No CMs other than participants were present during interviews or FGDs.

Study purposes and interviewers' backgrounds were explained to CMs by SCRMG professionals. During research activities, interviewers also introduced themselves, explained research objectives, and addressed confidentiality. Participants were provided with a study information sheet, while explicit information and opportunity to ask questions were provided verbally. All individuals provided signed informed consent before participation. The study had been approved by the 7th Health Region of Crete (6951;27/05/2016) and the Leiden University Medical Center Medical Ethical Committee (P16.063;04/15/2016) prior to its initiation.

Following the RAP (42), data were preliminarily analyzed at several time points. This allowed for iterative adjustment of the data collection strategy, ensuring that it was driven by local developments and research needs.

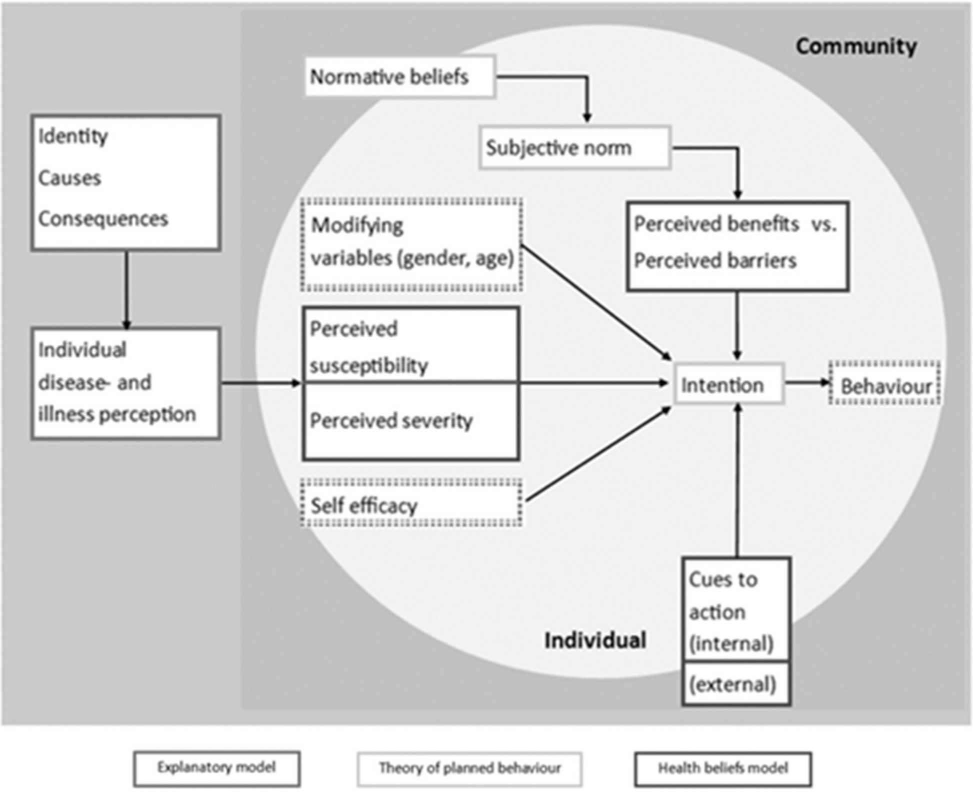


FIGURE 1 | The combined theoretical framework (reprinted with permission), as applied in the SETTINGtool (33).

Based on the theoretical model, topic guides were developed for data collection (**Appendix 4**). Since we aimed at exploring participants' own perceptions, no particular definition for chronic respiratory disease was provided to them during interviews. However, our approach was mainly focused on asthma and COPD, which were introduced to participants through a vignette describing a typical case of an individual with chronic respiratory symptoms (**Appendix 4**). Starting with the presentation of this vignette, topic guides prompted participants' reflections on presented respiratory symptoms, perceived causes and implications, beliefs and behaviors toward risk reduction, helping behaviors, and healthcare-seeking. Activities were audio-recorded if participants gave consent to do so, and field notes were taken. Interviews and FGDs lasted approximately 1 h. Debriefings with the research team occurred after each activity to allow further adjustments.

Research Team

The research team comprised of two Dutch and five Greek researchers, both male and female. The participation of SCRMG professionals and one mediator from the Roma community helped facilitate trust of the Roma toward researchers. Interviews and FGDs were conducted in Greek, with Greek team members interpreting for Dutch researchers. Before starting the RAP, all researchers were extensively trained by the leading expert researcher (E.B.).

Interviewers had no previous relationship with participants. The SCRMG employees were professionally related to CMs.

Data Analysis

Audio-recordings were transcribed verbatim and translated to English before coding. An inductive-deductive approach was used for transcript analysis. Specifically, transcript coding was both open and deductive, following the combined theoretical framework applied in this study, while allowing for new emergent themes and concepts. Coding and analysis were conducted manually and individually by two researchers (E.v.B. and M.A.) using Microsoft Word version 16. Discrepancies were solved through discussion until consensus was reached. Data structuring and reduction was first performed per informant group (CMs, HPs, and KIs), before combining the data of the different groups. Subsequently, emerging themes were identified using Thematic Content Analysis by E.v.B. and M.A., supervised by E.B. (43). Findings from observations, and field notes were then studied for presence of additional or contradicting themes to triangulate data before final themes were concluded.

RESULTS

Twenty-two participants (15 CMs, 3 HPs, 4 KIs) were included. In particular, interviewed HPs were doctors and nurses, while KIs were population mediators and social care professionals. We

TABLE 1 | Basic sample characteristics and field activities performed.

	Community members	Healthcare professionals	Key informants
Number of individuals	15	3	4
Male	1	1	1
Female	14	2	3
Age range	20–60	30–50	30–50
Focus groups	5	–	1
Interviews	2	3	2
Observations*	7	6	-

**Household observations for community members and consultation observations for healthcare professionals.*

All observed consultations were performed by a single healthcare professional.

TABLE 2 | Overview of key themes.

1. **CRD-related awareness and beliefs**
   - Limited awareness of CRDs and long-term exposure to risk factors
   - CM's beliefs and perceptions connected to acute symptoms (e.g. coughing, runny nose)
2. **Perceived severity and susceptibility**
   - Respiratory health is perceived as important, peers are urged to seek care
   - Low perceived disease susceptibility, especially among young Roma
3. **Risk exposure**
   - High prevalence of smoking and limited intention to quit, despite known harmful effects
   - HAP caused by widespread woodstove usage, attributed to financial constraints and poor living conditions
4. **Healthcare seeking behavior**
   - Daily needs and the perception of being strong are prioritized over personal health, especially among men
   - Persistent and activity-hampering symptoms drive help seeking behavior
5. **Perceived barriers/facilitators to care**
   - Health illiteracy, experienced discrimination, and financial constrains as barriers to care
   - Established trust and continuity of support services as facilitators to care

also collected three CM questionnaires which were used for data triangulation. They contained information on risk exposure and perceived causes of CRDs which did not contrast or hold any new information compared to the information obtained during the interviews and FGDs

As shown in **Table 1**, female sex prevailed our sample (14/15 CMs, 2/3 HPs and 3/4 KIs). The age range was 20–60 years for CMs, 30–50 years for HPs and 30–50 years for KIs. Seven interviews and six FGDs were conducted. Additionally,13 observations were performed. For one interview and two FGDs CMs did not consent to audio-recording. In these cases, the findings were documented in field notes and included in preliminary analysis during researcher debriefings.

During our fieldwork, the aforementioned poor living conditions were confirmed. Lack of electricity was reported as a major problem by all encountered camp residents and as a frequent reason of conflict with local authorities (**Appendix 2**). As elaborated on in the Results section below and **Appendix 2**, lack of electricity was also linked to health behavior as it prevented, for example, the proper practice of hygiene and proper medication storage. In terms of exposure to risk factors, tobacco smoking was observed and reported at high frequencies during our camp visits. Additionally, indications related to increased levels of household air pollution were documented, as improvised biomass-burning heating devices were observed during almost all our household visits, while burning materials (wood, melamine pieces, plastics and others) were seen stored outside of many households throughout the camp.

Themes emerging from the analysis are presented in **Table 2** and elaborated on below, based on reflections of the combining elements of our theoretical framework. **Appendix 5** presents additional quotes supporting findings or providing contextual information.

CRD-Related Awareness and Beliefs

In terms of perceived disease identity, beliefs expressed by CMs were predominantly connected to acute respiratory symptoms, such as coughing and a runny nose, rather than chronic symptoms. While medical respiratory terminology (such as dyspnoea) was hardly used, asthma was a familiar term. However, the meanings attached to asthma (e.g., the chronic nature of the disease and its health implications) differed from medical connotations.

- *Interviewer: Asthma... have you heard about it?*
- *All CMs: Yes...*
- *CM9: This disease where you use a mask...*
- *CM8: It's mostly for older people... (CMs, female, age 20–30, in FGD)*

In contrast, COPD was completely unknown (**Appendix 4**).

In terms of perceived causes, CMs believed several factors to influence respiratory health. Heredity, aging, allergies, and exposure to observable risk factors were mentioned as potential causes, such as tobacco smoke and HAP from burning low-quality fuels (**Appendix 3**). Triangulation data showed similar findings.

> *"(. . .) when we light the fireplace, and smoke is coming out... Maybe the cause [of breathlessness and productive coughing] is having the fire inside... (CM1, female, age 31)"*
> *"I believe that it comes with age... Because everything appears when you get old... And from smoking! There are some people that when they have it [i.e. the lung problem described in the vignette] they quit smoking. (CM8, female)"*

However, according to a HP, comprehensive understanding of CMs regarding the medical aspects of CRD and the long-term effects of exposure to risk factors was limited:

> *"A kid may catch a cold more frequently than other children, because his or her respiratory system is no longer in a good condition due to smoke from the woodstove (. . .) even if you explain this [to Roma], they can't conceive it. (HP3, female)"*

According to CMs, the prevalence of CRD in the camp was low. HPs confirmed this, yet cautioned that it could be underdiagnosed. Circulatory, metabolic and mental health disorders were noted among the most frequently encountered conditions in the camp:

> *"We may see problems of diabetes, blood pressure, heart problems. Respiratory problems not so much. (KI1, female)"*
> *"(. . .) the percentage of people that suffer from respiratory diseases is not high. . . there is the possibility of not knowing the exact number [of respiratory patients] because they do not make appointments or undergo medical examinations, especially regarding respiratory issues. (HP2, female)."*
> *"The first thing, that comes to mind, as main [..] basic issue they seek to solve, is their psychiatric problems. (KI3, female)"*

Perceived Severity and Susceptibility

In agreement with the components of our theoretical framework, individual disease and illness perception of the studied population were linked to perceived susceptibility and perceived severity, which were further linked to action cues and, ultimately, behaviors. According to CMs, Roma perceived respiratory health as important. If a CM experienced persisting symptoms, peers would usually consider this important enough to advise them to visit a doctor, reflecting a subjective norm toward healthcare seeking behavior.

> *"(. . .) when I see someone who coughs and doesn't stop, I tell him to visit a doctor, without waiting, because something may happen to his lungs and he (may) suffer more. (CM3, female, age 45)"*
> *"Because everything comes from the lung... All people should take care... maybe your breath will stop... this is what I know... (CM8, female)"*

For personal health issues, however, lower perceived disease susceptibility was mentioned. According to CMs and HPs, especially young Roma tended to ignore respiratory symptoms and rather waited for them to pass, suggesting that age can be a modifying variable toward this direction.

> *"As far as their own health is concerned, younger ones are more relaxed... They believe they will never get something. (HP3, female)"*

Symptoms among children were perceived as more alarming. Children's health was clearly prioritized over adults' health (**Appendix 5**).

> *"I don't leave my child without going to the doctor (. . .) whatever the doctor says, I do everything (. . .) If I have 100 euros, I will prefer to give them for my kid (...) For myself, I will go to the hospital. But (. . .) even if I don't go, I have no problem. (CM15, female)"*

Risk Exposure

Tobacco smoking and exposure to HAP were identified as core risk behavior variables. Namely, in terms of perceptions related to these risk behaviors, all participants (smokers or non-smokers) described smoking to be harmful for health, particularly for smokers (active smoking) and children (passive smoking). Participants consistently reported Roma to be heavy smokers, starting at an early age. Sex seemed to be an important modifying variable for smoking behavior. In particular, although our sample consisted mainly of women, both male and female participants reported that smoking was much more frequent and heavier among men than among women. Passive smoking was recognized to happen frequently. Both CMs and HPs mentioned that the intention to stop smoking was extremely low. Smoking appeared to hold strong emotional value.

> *"(. . .) the doctors refer them [men who smoke] to cessation clinics and we book their appointments, yet they never go. (. . .) They declare: "I don't want to quit! Do whatever you want, I will not quit!". (...) They think it's something that makes them forget their troubles, as they say. (HP3, female)"*
> *"Interviewer: Do most people smoke inside or outside the house?"*
> *"CM14 (female): Inside. Our babies also smoke inside [i.e. refers to passive smoking]."*

HAP, however, appeared to be strongly related to financial constraints, poor living conditions and low health literacy, indicating low socioeconomic status as a significant modifying variable of health behavior. CMs explained that they usually burned cheap, low quality wooden pallets indoors for heating. According to HPs and KIs, CMs' awareness regarding the long-term consequences of HAP for respiratory health may be generally low. HPs and CMs described that "clean" alternatives were too expensive for most CMs and that electricity was unavailable. Especially in winter, ventilation was limited to a minimum to keep the poorly insulated houses warm, leading to high HAP exposure. The presence of indoor woodstoves and wooden pallets throughout the camp was confirmed in our household observations.

> *"There are important fears for health during winter with the woodstoves. We [CMs] are naive to be using melamine woods [synthetic woods] (...) We don't have to pay for them, and unfortunately, we can't conceive there will be a problem. (KI2, male, about CMs)"*
> *"Interviewer: Do you try to ventilate the house? (. . .)"*
> *"CM3 (female): We don't take much care of it. I close the windows and the door sometimes (. . .) We all sit close to the woodstove. And I don't open something to ventilate the area. Only when the weather is good. . . ."*

Healthcare Seeking Behavior

Internal and external cues to action were reflected in the healthcare seeking behavior of the studied population. In particular, all participants mentioned that CMs usually prioritized their daily needs over personal long-term health. Additionally, a generalized negative attitude toward visiting doctors and healthcare services was expressed, attributed mainly to fear of the unknown consequences of a potential diagnosis. As such, healthcare seeking was driven by perceived disease severity, wherein only persistent and activity-hampering symptoms were a direct cue to action.

> *"If he has something that needs rest he will not go [to work], but if it is not something serious he will go. Most people go [to work] in order to feed their families. (CM8, female, age 38)"*

> *"(. . .) health is not important for them [i.e. CMs]. It is not a priority. What's important for them is to work, make some money, and have food today. (HP2, female)"*

Interviewed KIs and CMs and fieldnotes, including one male perspective, also suggested that men tend to be more reluctant to talk about disease and visit healthcare services than women. Interviewed participants (both male and female) attributed this directly to men's ability to work and support their families.

> *"Because at home, he [the man] is the pillar. And if he had something, he wouldn't be able to work. And if he would see that his children should work [to provide for the family], he would get more worried. (CM3, female, age 45)"*
> *"(A Roma man may be like) "If I go to the hospital, they may find that I have a severe condition. I will not go, I will stay at home whatever happens" (...) Or they may fear the hospital and whenever they go they may feel bad. (CM15, female)"*

In general, (male) sex was a significant modifying variable for healthcare seeking practice, since all participants described an important role for men in the Roma community. Due to the majority of interviewed participants being women, this information was largely a secondary report. However, both CMs and KIs mentioned masculine elements of strength and pride to influence help-seeking behavior (norms).

> *"In younger ages, the man feels more vulnerable when more people learn that he has a problem (...) Especially if he has not had children yet, this gives him a bad reputation (...) If he is of older age and has had children, it's a matter of the position he has in"*
> *"the community (...) he will try to deal with it like "Ok, I have nothing", but he will always have the fear of losing his prestige. (KI1, female)"*

Perceived Barriers/Facilitators to Care

As illustrated in our theoretical framework, perceived barriers and facilitators were linked to health care seeking behavior in the studied Roma population. Namely, HPs and KIs perceived (health) illiteracy to be an important barrier to healthcare, limiting the population to understand information and to follow medical advice. Similarly, CMs indicated difficulty to seek medical care.

> *"This [illiteracy] (. . .) is important. . . it's often a reason for them to have fear, anxiety of cooperating [with HPs], low compliance, or not understanding instructions. For example (. . .) the importance of participating in a health education [activity] for respiratory diseases. (KI3, female)"*
> *"(. . .) they feel uncomfortable and disadvantaged because they are illiterate, to visit a [health] service and undergo some tasks. Their level of education, I think, draws them back because they feel uncomfortable and disadvantaged. (HP2, female)"*

Also, CMs reported that they experience discrimination in hospitals and receive limited explanations by doctors as significant factors contributing to their reluctance to seek care.

> *"(. . .) some people are racists! (. . .) Even when I go to hospitals, and I wait in line and it's my turn to go in, they will not call me, they will call their people, their acquaintances (. . .) That's why we got sick of hospitals... (CM14, female, age 55)"*

Community support during sickness was reported to be strong, such as CMs accompanying peers to healthcare services (subjective norm). The main facilitator to access care was the trust established over the years between the Roma population and the SCRMG facility, increasing HPs' and KIs' influence in supporting and motivating CMs in their health behavior.

> *"It's important that they [CMs] trust you. If they don't trust you, the problem will be the same. . . It is important for these people to trust this [entire] system around them. . . to have better contact and to accept the message. (HP1, male)"*

Limited financial means and poor living conditions were mentioned as key barriers preventing adoption of healthy practices, wherein financial constraints also limit the ability to overcome the physical distance to healthcare facilities.

DISCUSSION

Main Findings

This qualitative exploration of beliefs, perceptions, and behaviors among a Greek Roma population demonstrates five key themes related to CRDs. Concepts around asthma and COPD were introduced to interviewed participants through a vignette. The first emerging theme was related to the components of perceived disease identity and causes and reflected the limited awareness of CRDs and the harm of long-term exposure to risk factors. CM's beliefs and perceptions were predominantly related to acute symptoms. The second theme reflected the perceived severity and susceptibility to CRDs. Although respiratory health was perceived as important, it was reported that especially young Roma tend to believe that their personal susceptibility to CRD is low. The third theme was related to risk behaviors and related perceptions. Tobacco smoking was highly prevalent and intention to quit was low, although it was recognized as harmful for health. HAP due to widespread woodstove usage was attributed to financial constraints and poor living conditions. The fourth theme was related to healthcare seeking behavior and suggested the prioritization of daily needs over health issues. Providing for one's family and the perception of being strong were prioritized over personal health, particularly in men, unless symptoms were persistent and severe (norms). The fifth and last theme indicated the factors influencing access to care and healthcare seeking behavior. Health illiteracy, experienced discrimination, and financial constraints were the main barriers to care and behavioral change. Established trust and continuity of support services were key facilitators to healthcare seeking behavior.

Interpretation of Findings in Relation to Literature

Respiratory health was considered important by the Roma population of our study, yet did not result in an actual priority due to a lack of awareness about CRD and its risk factors, and a lack of means to reduce exposure. This is a finding that has not previously been reported. We also observed a low perceived disease susceptibility which, according to literature, likely contributes to late presentation at healthcare services and low engagement to preventive activities, especially among male Roma (44, 45). The particular concern about children's health found in this research has earlier been described similarly concerning immunization uptake (46). In line with other studies in different Roma populations, we identified an interplay of day-to-day priorities, ignorance of long-term implications, fear of a diagnosis, distance to healthcare facilities, low levels of health literacy, discrimination-fueled distrust of healthcare providers, and a strong masculine culture to negatively influence health behavior (45, 47–49). Our study is the first, however, to relate this interplay specifically to CRDs and potentially other chronic diseases.

In contrast to previous studies that report a socially bonding "norm" to reject healthcare standards (47, 50, 51), we found that this Roma population urged each other to seek medical care for persisting respiratory symptoms. This may indicate a certain trust developed over the years in local health services, and particularly in the SCRMG (44). Strikingly, this Roma population had more negative attitudes toward hospital care compared to the SCRMG. We noted that previous experiences shaped help-seeking behavior, wherein perceived discrimination negatively influenced behavior. It has been suggested that Roma "nonadherence norms" to medical recommendations are less likely to be present in environments that hold less anti-Roma views (52). This underlines the importance to support non-Roma HPs in their interaction with Roma, as expressed in previous studies (47–49, 51). Considering that most CRD-related care in Greece takes place in hospital settings (52), the importance of welcoming interactions should be duly noted. Identified barriers and facilitators to care, including perceived discrimination, low levels of health literacy, and established trust between the population and the SCRMG, corroborate previous evidence in other Roma populations (44–46, 53).

Furthermore, respiratory risk factors including smoking and HAP were highly prevalent in the Roma population of our study. We uncovered a close relationship between woodstove usage, financial constraints, limited ventilation, and poor living conditions. The negative health effects of the resulting HAP have been broadly reported in literature as a result of substandard infrastructure and poverty among Roma (16, 46, 53). In line with our findings, reluctance toward smoking cessation has been associated with emotional value, low health literacy, and a lack of confidence in effectiveness of risk reduction (54–56). Corresponding widespread smoking, especially in Roma men, is well-documented (15, 18, 19). Other studies have found smoking among Roma to be initiated at a young age and, alike HAP in our study, indicate a strong association with a lower socio-economic status (57, 58).

Strengths and Limitations

To the best of our knowledge, the perception of respiratory health and associated symptoms, such as coughing, by Roma has not been studied before. The main strength of our research is its multi-faceted exploration of an undocumented topic based on a well-theorized framework. To achieve an adequate representation of the topic, we collected data from three different stakeholder groups, while combining interviews and FGDs with observations. Transparency and validity in data analyses were promoted through careful thematic analysis by two independent researchers from different backgrounds.

Several limitations merit emphasis, however. First, since this was a hard-to-reach population, we were bound to the limited time available by the SCRMG-professionals to gain CMs' trust. Therefore, we did not achieve the desired sample diversity in terms of sex nor reach full data saturation or collect a sufficient number of questionnaires; the topics low perceived disease susceptibility, experienced discrimination in hospitals, and specifically Roma male views on health and health intervention participation would benefit from further exploration. Nevertheless, based on our current findings, we did not find indications for specific differences between males and females. Second, the sample's dependence on participants' opportunity and willingness possibly created selection bias toward more collaborative Roma. To a certain degree, triangulating interview and FGD data with observations and field notes accounted for this effect. In addition, the multidisciplinary team and inclusion of views from multiple informant groups helped to limit this potential bias. Third, given the reported diversity of Roma as ethnic group, data of this study reflecting one camp, and the qualitative study design, findings cannot be generalized. Rather, they should be interpreted with caution and in relation to previously published work (19, 37). Finally, given that the Roma population is culturally distinguished from the rest of FRESH AIRs' low-resource implementation settings, our theoretical framework would have been benefited by a sociology of health perspective, especially with regard to social contexts of poverty or strong ethnic differentiation.

Implications for Research, Policy, and Practice

Our study demonstrated typical beliefs, perceptions, and behaviors among a Greek Roma population, which should be considered during provision of care to ensure equity in access to and quality of care. Our findings suggest that successful approaches to combat CRD among Roma would benefit from following a multidisciplinary approach starting from the population and expanding toward the societal and public health level, as elaborated below. Similar approaches could be considered for other chronic diseases and vulnerable or low-resource populations. Such strategies would be both relevant and timely, as structural primary care reforms have been unfolding in Greece and the urgency of Roma inclusion has been underlined by the recent EU Roma strategic framework (27). Above all, our findings emphasize the importance to address these local beliefs and behaviors to combat the poor health outcomes of Europe's largest ethnic minority, which was also disproportionately hit by the coronavirus pandemic (8, 15, 16, 25, 26, 45).

At the population level, there seems to be a general need to improve CRD-related health literacy among Roma. Improved awareness of the harmful effects of HAP and other risk factors should be developed to aid conceptualization of CRDs and to weigh long-term implications and daily priorities. This is particularly important, considering that COPD poses a significant burden in Greece (20, 21, 59, 60) and taking into account Roma's vulnerability to CRDs (8–10). Although improvement of health literacy in itself is an important focus, policy makers should go beyond and actively work with cultural differences related to health and local reality (53, 56, 61, 62). This can only happen in collaboration with Roma: leveraging trust established by local mediators and strengthening relationships between Roma communities and primary care services (49, 63). For example, a Hungarian primary care programme involved non-professional health mediators of Roma ethnicity to promote access to care and was recently positively evaluated (64). CRD-specific examples of population-level interventions to increase CRD-related health literacy through increasing awareness can be drawn from FRESH AIR's work in global low-resource settings. Such approaches have used implementation science to culturally adapt and implement evidence-based cascading train-the-trainer awareness-raising interventions regarding the damaging effects of smoke from tobacco and HAP. These interventions start by identifying context-specific factors that may drive effectiveness and engage the whole community and relevant stakeholders to their development and delivery. Their effects on knowledge increase have been proven promising (65, 66).

In addition, a strong masculine culture was reflected in this study by all interviewed participants (regardless of their sex) and this may be a point of attention. Although further studies would be beneficial to better understand male perspectives on health behavior, previous studies have also indicated a strong culture of masculinity to hamper smoking cessation and participation in cancer screening among Roma elsewhere in Europe (56, 67). It has also been suggested that Roma are much less likely to support tobacco control measures than non-Roma of similar socioeconomic status (54, 67). Although evidence is limited, implementation strategies of risk-reduction progammes, such as smoking cessation, may benefit from taking into account such different attitudes and employing culturally acceptable methods to address the population.

Expanding toward the societal and public health level, Roma's living conditions and access to care need to be addressed. Structural improvement of (financial) access to better housing and cleaner fuels is necessary to enable behavioral change and reduce HAP. Within the studied population, socio-economic status was an important modifying variable for health seeking behavior. As appears from reporting by the EU Agency for Fundamental Rights, Roma, such as the population residing in the camp subject of this study, face precarious work, limited state support in provision of needs such as electricity, and uncertainty of long-term residence (25). Own investments in improvement of living conditions can therefore be perceived as too expensive, especially in the light of an uncertain duration of stay.

Furthermore, community support centers can be short of (medical) personnel and subject to fluctuating funding (69). Although this theme was not discussed in the Results section of this manuscript, additional quotes and context information (**Appendix 5**) do refer to the subject of fluctuating financial means. Only using available funds to facilitate direct care provision may be insufficient. Public health outreaches have been found to reinforce disengagement and do not alter the underlying system shaping health behavior (45, 53). Therefore, to facilitate adequate help-seeking behavior, healthcare efforts could benefit from taking a systemic approach, including measures to improve mutual cultural awareness between Roma and non-Roma (62, 70). A community-based participatory approach has been suggested as an effective way to foster Roma-involvement in readjustment of community agendas and allocation of resources based on local priorities (51, 69). As time investment can be substantial and expensive, university involvement or public-private partnerships have been suggested as successful enablers (68, 70).

CONCLUSION

Respiratory health does not seem an actual priority for the studied Roma population, despite its reported perceived importance. Health illiteracy and awareness of long-term effects of CRDs and their risk factors, such as HAP, need to be enhanced. To enable adequate help-seeking behavior and behavioral change benefiting health, implementation strategies of CRD-related interventions could increase their success by including investments in establishing trust, fostering community engagement, identifying ways to minimize the effects of financial constraints, and ensuring welcoming interactions between Roma and non-Roma. Strong relationships between Roma communities and primary care providers, such as the municipal support center and Roma mediator in this study, can act as an important facilitator. These findings can be used to design more successful context-driven implementation strategies for evidence-based respiratory health interventions for the studied Roma population and potentially for other Greek or European Roma populations. This study's approach and our findings may be relevant, not only for CRD and Roma, but also for other chronic diseases and similarly vulnerable/low-resource populations.

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 7th Health Region of Crete (6951; 27/05/2016) and the Leiden University Medical Center Medical Ethical Committee (P16.063; 04/15/2016). The patients/participants provided their written informed consent to participate in this study.

FRESH AIR COLLABORATORS

Pham Le An, Azamat Akylbekov, Andy Barton, Antonios Bertsias, Pham Duong Uyen Binh, Job FM van Boven, Dennis Burges, Lucy Cartwright, Liza Cragg, Tran Ngoc Dang, Ilyas Dautov, Berik Emilov, Irene Ferarrio, Frederik A van Gemert, Ben Hedrick, Le Huynh Thi Cam Hong, Nick Hopkinson, Elvira Isaeva, Rupert Jones, Corina de Jong, Sanne van Kampen, Winceslaus Katagira, Bruce Kirenga, Jesper Kjærgaard, Rianne MJJ van der Kleij, Janwillem Kocks, Le Thi Tuyet Lan, Tran Thanh Duv Linh, Kim Xuan Loan, Maamed Mademilov, Andy McEwen, Patrick Musinguzi, Rebecca Nantanda, Grace Ndeezi, Sophia Papadakis, Hilary Pinnock, Jillian Pooler, Maarten J Postma, Anja Poulsen, Pippa Powell, Nguyen Nhat Quynh, Susanne Reventlow, Sally Singh, Talant Sooronbaev, Jaime Correia de Sousa, James Stout, Marianne Stubbe Østergaard, Aizhamal Tabyshova, Tran Diep Tuan, James Tumwine, Le Thanh Van, Nguyen Nhu Vinh, Simon Walusimbi, Louise Warren, Sian Williams.

AUTHOR CONTRIBUTIONS

MA participated in local study development and contributed to data collection and analysis. MA and EBre analysed the data, co-wrote the first and subsequent versions of the manuscript. EBra designed the study, coordinated and conducted data collection, gave input throughout the entire data analysis, and writing process. IT participated in the writing of the original FRESH AIR proposal, provided scientific input for local study development, implementation, and results interpretation DS-P, VC, AK, and CP participated in local study development, data collection, and data interpretation. CP, RK, MC, NC, and RR advised throughout the study design and data collection. CL was the country lead for the FRESH AIR project, providing overall supervision and scientific input for study design, implementation, data interpretation, and writing of the manuscript. NC was the principal investigator of the overall FRESH AIR project, an original author of the FRESH AIR proposal, and contributed to the development of the study protocol. All authors critically reviewed the manuscript and approved its final version.

FUNDING

This study was funded by the EU Research and Innovation program Horizon2020 (Health, Medical research and the challenge of aging) under grant agreement number 680997 and trial registration number NTR5759. The funders had no role in study design, data collection, data analysis, data interpretation, or writing of the report.

ACKNOWLEDGMENTS

We sincerely thank the employees of the Support Center for Roma and Minority Groups of the Municipality of Heraklion for their vital contribution in the conduction of this research. We also thank the Roma community of Heraklion for their participation in this study. We also thank Mr. Antonios Bertsias and Mr. Myron Galenianos, team members of the Clinic of Social and Family Medicine, School of Medicine, University of Crete, for their valuable assistance in the implementation of FRESH AIR. We acknowledge Sian Williams, Lisa Cragg, and the International Primary Care Respiratory Group (IPCRG) for their continuous support and leadership in the execution of this work.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2022.812700/full#supplementary-material

REFERENCES

1. Forum of International Respiratory Societies. *The Global Impact of Respiratory Disease*. 2nd ed. Sheffield: European Respiratory Society. (2017).
2. GBD Chronic Respiratory Disease Collaborators. Prevalence and attributable health burden of chronic respiratory diseases, 1990-2017: a systematic analysis for the Global Burden of Disease Study (2017). *Lancet Respir Med*. (2020) 8:585–96.
3. World Health Organization. *Chronic obstructive pulmonary disease (COPD)*. (2017). Available online at: https://www.who.int/news-room/fact-sheets/detail/chronic-obstructive-pulmonary-disease-(copd).
4. World Health Organization. *Asthma*. (2020). Available online at: https://www.who.int/news-room/fact-sheets/detail/asthma.
5. Eisner MD, Anthonisen N, Coultas D, Kuenzli N, Perez-Padilla R, Postma D, et al. An official American Thoracic Society public policy statement: Novel risk factors and the global burden of chronic obstructive pulmonary disease. *Am J Respir Crit Care med*. (2010) 182:693–718. doi: 10.1164/rccm.200811-1757ST
6. Pathak U, Gupta NC, Suri JC. Risk of COPD due to indoor air pollution from biomass cooking fuel: a systematic review and meta-analysis. *Int J Environ Health Res*. (2020) 30:75–88. doi: 10.1080/09603123.2019.1575951
7. Cohen AJ, Brauer M, Burnett R, Anderson HR, Frostad J, Estep K, et al. Estimates and 25-year trends of the global burden of disease attributable to ambient air pollution: an analysis of data from the Global Burden of Diseases Study (2015). *Lancet*. (2017) 389:1907–18. doi: 10.1016/S0140-6736(17)30505-6
8. European Commission. *Report on the Health Status of the Roma Population in the EU and monitoring Data Collection in the Area of Roma Health in the Member States*. Publications Office of the European Union (2014).
9. Marmot M, Allen J, Bell R, Bloomer E, Goldblatt P. Consortium for the European Review of Social Determinants of Health and the Health Divide. WHO European review of social determinants of health and the health divide. *Lancet*. (2012) 380:1011–29. doi: 10.1016/S0140-6736(12)61228-8
10. Mihailov D. *The Health Situation of Roma Communities: Analysis of The Data From The UNDP/World Bank/EC Regional Roma Survey (2011)*. Bratislava: United Nations Development Programme. (2012).
11. European Commission. Roma integration in the EU. (2020). Available online at: https://ec.europa.eu/info/policies/justice-and-fundamental-rights/combatting-discrimination/roma-and-eu/roma-integration-eu_en.
12. Casillas-Clot J, Pereyra-Zamora P, Nolasco A. Determinants of disability in minority populations in Spain: a nationwide study. *Int J Environ Res Public Health*. (2021) 18:3537. doi: 10.3390/ijerph18073537

13. Vincze F, Földvári A, Pálinkás A, Sipos V, Janka EA, Ádány R, et al. Prevalence of chronic diseases and activity-limiting disability among Roma and non-Roma people: a cross-sectional, census-based investigation. *Int J Environ Res Public Health.* (2019) 16:3620. doi: 10.3390/ijerph16193620
14. Belak A, Geckova AM, van Dijk JP, Reijneveld SA. Health-endangering everyday settings and practices in a rural segregated Roma settlement in Slovakia: a descriptive summary from an exploratory longitudinal case study. *BMC Public Health.* (2017) 17:128. doi: 10.1186/s12889-017-4029-x
15. Cook B, Ferris Wayne G, Valentine A, Lessios A, Yeh E. Revisiting the evidence on health and health care disparities among the roma: a systematic review 2003-2012. *Int J Public Health.* (2013) 58:885–911. doi: 10.1007/s00038-013-0518-6
16. Pappa E, Chatzikonstantinidou S, Chalkiopoulos G, Papadopoulos A, Niakas D. Health-related quality of life of the roma in greece: the role of socio-economic characteristics and housing conditions. *Int J Environ Res Public Health.* (2015) 12:6669–81. doi: 10.3390/ijerph120606669
17. Majdan M, Coman A, Gallová E, Duricová J, Kállayová D, Kvaková M, et al. Assessment of the indoor environment and implications for health in Roma villages in Slovakia and Romania. *Cent Eur J Public Health.* (2012) 20:199–207. doi: 10.21101/cejph.a3719
18. Kaditis AG, Gourgoulianis K, Tsoutsou P, Papaioannou AI, Fotiadou N, Messini C, et al. Spirometric values in Gypsy (Roma) children. *Respir Med.* (2008) 102:1321–28. doi: 10.1016/j.rmed.2008.03.025
19. Rodriguez NS, Derecho NR. *Health and The Roma Community, Analysis of the Situation in Europe*. Madrid: Fundación Secretariado Gitano Health Area. (2009).
20. Kotsiou OS, Zouridis S, Kosmopoulos M, Gourgoulianis KI. Impact of the financial crisis on COPD burden: Greece as a case study. *Eur Respir J.* (2018) 27:170106. doi: 10.1183/16000617.0106-2017
21. Mitsiki E, Bania E, Varounis C, Gourgoulianis KI, Alexopoulos E. Characteristics of prevalent and new COPD cases in Greece: the GOLDEN study. *Int J Chron Obstruct Pulmon Dis.* (2015) 10:1371–82. doi: 10.2147/COPD.S81468
22. European Commission. *Report on the Implementation of National Roma Integration Strategies*. Brussels: Publications Office of the European Union (2019).
23. Economou C. *Barriers and Facilitating Factors in Access to Health Services in Greece*. Copenhagen: WHO Regional Office for Europe (2015).
24. European Union Agency for Fundamental Rights. *Second European Union Minorities and Discrimination Survey: Roma - Selected Findings*. Luxembourg: Publications Office of the European Union (2016).
25. European Union Agency for Fundamental Rights. *Coronavirus Pandemic in the EU - Impact on Roma and Travellers*. Luxembourg: Publications Office of the European Union (2020).
26. Armitage R, Nellums LB. COVID-19 and the gypsy, Roma and traveller population. *Public Health.* (2020) 185:48. doi: 10.1016/j.puhe.2020.06.003
27. European Commission. EU. *Roma Strategic Framework for Equality, Inclusion and Participation for 2020-2030*. Brussels. (2020).
28. Ridde V. Need for more and better implementation science in global health. *BMJ Global Health.* (2016) 1:e000115. doi: 10.1136/bmjgh-2016-000115
29. Damschroder LJ, Aron DC, Keith RE, Kirsh SR, Alexander JA, Lowery JC. Fostering implementation of health services research findings into practice: a consolidated framework for advancing implementation science. *Implement Sci.* (2009) 4:50. doi: 10.1186/1748-5908-4-50
30. Durlak JA, DuPre EP. Implementation matters: a review of research on the influence of implementation on program outcomes and the factors affecting implementation. *Am J Community Psychol.* (2008) 41:327–50. doi: 10.1007/s10464-008-9165-0
31. Nilsen P, Bernhardsson S. Context matters in implementation science: a scoping review of determinant frameworks that describe contextual determinants for implementation outcomes. *BMC Health Serv Res.* (2019) 19:189. doi: 10.1186/s12913-019-4015-3
32. Arora VS, Kühlbrandt C, McKee M. An examination of unmet health needs as perceived by Roma in Central and Eastern Europe. *Eur J Public Health*. (2016) 26:737–42. doi: 10.1093/eurpub/ckw004
33. Brakema EA, van der Kleij RMJJ, Poot CC, Chavannes N, Tsiligianni I, Walusimbi S, et al. A systematic approach to context-mapping to prepare for health interventions: development and validation of the SETTING-tool in four countries. *BMJ Global Health.* (2021) 6:e003221. doi: 10.1136/bmjgh-2020-003221
34. Cragg L, Williams S, Chavannes N, FRESH AIR. An implementation research project funded through Horizon 2020 exploring the prevention, diagnosis and treatment of chronic respiratory diseases in low-resource settings. *NPJ Prim Care Resp Med.* (2016) 26:1603. doi: 10.1038/npjpcrm.2016.35
35. 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
36. National Commission for Human Rights. *Annual Report (2008)*. Athens: NCHR. (2009).
37. Andrioti D, Kotrotsou C, Tsakatara V. The medical social centres in support of Roma in Greece. *Divers Equal Health Care.* (2013) 10:95–9. Available online at:here: https://www.primescholars.com/articles/the-medical-social-centres-in-support-of-roma-in-greece-94539.html
38. Rosenstock IM, Strecher VJ, Becker MH. Social learning theory and the health belief model. *Health Educ Behav.* (1988) 15:175–83. doi: 10.1177/109019818801500203
39. Kleinman A. *Patients and Healers in the Context of Culture: An Exploration of the Borderland Between Anthropology, Medicine, and Psychiatry*. Berkeley: University of California (1980). doi: 10.1525/9780520340848
40. Ajzen I. The theory of planned behaviour. *Organ Behav Hum Decis Process.* (1991) 50:170–211. doi: 10.1016/0749-5978(91)90020-T
41. Beebe J. *The Sage Encyclopedia of Qualitative Research Methods; Rapid Assessment Process*. Thousand Oaks: SAGE Publications. (2008).
42. Braun V, Clarke V. Using thematic analysis in psychology. *Qual Res Psychol.* (2006) 3:77–101. doi: 10.1191/1478088706qp063oa
43. Berlin J, Smith D, Newton P. "It's because it's cancer, not because you're a Traveller"-exploring lay understanding of cancer in English Romany Gypsy and Irish Traveller communities. *Eur J Oncology Nursing.* (2018) 34:49–54. doi: 10.1016/j.ejon.2018.02.010
44. McFadden A, Siebelt L, Gavine A, Atkin K, Bell K, Innes N, et al. Gypsy, Roma and Traveller access to and engagement with health services: a systematic review. *Eur J Public Health*. (2018) 28:74–81. doi: 10.1093/eurpub/ckx226
45. Jackson C, Bedford H, Cheater FM, Condon L, Emslie C, Ireland L, et al. Needles, Jabs and Jags: a qualitative exploration of barriers and facilitators to child and adult immunisation uptake among gypsies, travellers and Roma. *BMC Public Health*. (2017) 17:254. doi: 10.1186/s12889-017-4178-y
46. Belak A, Madarasova AM, van Dijk JP, Reijneveld SA. Why don't segregated Roma do more for their health? An explanatory framework from an ethnographic study in Slovakia. *Int J Public Health.* (2018) 63:1123–31. doi: 10.1007/s00038-018-1134-2
47. van Cleemput P, Parry G, Thomas K, Peters J, Cooper C. Health-related beliefs and experiences of gypsies and travellers: a qualitative study. *J Epidemiol Community Health.* (2007) 61:205–10. doi: 10.1136/jech.2006.046078
48. Lämsä R, Castaneda AE, Weiste A, Laalo M, Koponen P, Kuusio H. The role of perceived unjust treatment in unmet needs for primary care among Finnish Roma adults. *Int J Environ Res Public Health.* (2020) 17:5825. doi: 10.3390/ijerph17165825
49. Condon LJ, Salmon D. 'You likes your way, we got our own way': gypsies and travellers' views on infant feeding and health professional support. *Health Expect.* (2015) 18:784–95. doi: 10.1111/hex.12214
50. Pavlikova B, Freel L, van Dijk JP. To comply or not to comply: Roma approach to health laws. *Int J Environ Res Public Health.* (2020) 17:3087. doi: 10.3390/ijerph17093087
51. Belak A, Madarasova Geckova A, van Dijk JP, Reijneveld SA. What Roma nonadherence is likely and what drives it? Reply to Broz and Nunes. *Int J Public Health.* (2019) 64:805–7. doi: 10.1007/s00038-019-01250-z
52. Tsiligianni I. Respiratory health in Greece. *Lancet Respir Med.* (2020) 8:e65–6. doi: 10.1016/S2213-2600(20)30276-9
53. Petek D, Patič DR, Svab I, Lolić D. Attitudes of Roma toward smoking: qualitative study in Slovenia. *Croat Med J.* (2006) 47:344–7.
54. Stewart DW, Adams CE, Cano MA, Correa-Fernández V, Li Y, Waters AJ, et al. Associations between health literacy and established

predictors of smoking cessation. *Am J Public Health.* (2013) 103:e43–e9. doi: 10.2105/AJPH.2012.301062

55. Condon L, Curejova J, Morgan DL, Miles G, Fenlon D. Knowledge and experience of cancer prevention and screening among Gypsies, Roma and Travellers: a participatory qualitative study. *BMC Public Health.* (2021) 21:360. doi: 10.1186/s12889-021-10390-y
56. Sárváry A, Kósa Z, Jávorné RE, Gyulai A, Takács P, Sándor J, et al. Socioeconomic status, health related behaviour, and self-rated health of children living in Roma settlements in Hungary. *Cent Eur J Public Health.* (2019) 27:24–31. doi: 10.21101/cejph.a4726
57. Merzah M, Kósa Z, Sándor J, Natae S, Pikó P, Ádány R, et al. Roma socioeconomic status has a higher impact on smoking behaviour than genetic susceptibility. *Int J Environ Res Public Health.* (2021) 18:3206. doi: 10.3390/ijerph18063206
58. Tzanakis N, Anagnostopoulou U, Filaditaki V, Christaki P, Siafakas N. COPD group of the hellenic thoracic society. Prevalence of COPD in Greece. *Chest.* (2004) 125:892–900. doi: 10.1378/chest.125.3.892
59. Hanssens LGM, Devisch I, Lobbestael J, Cottenie B, Willems S. Accessible health care for Roma: a gypsy's tale a qualitative in-depth study of access to health care for Roma in Ghent. *Int J Equity Health.* (2016) 15. doi: 10.1186/s12939-016-0327-7
60. Belak A, Veselska ZD, Geckova AM, van Dijk JP, Reijneveld SA. How well do health-mediation programs address the determinants of the poor health status of Roma? A longitudinal case study. *Int J Environ Res Public Health.* (2017) 14:1569. doi: 10.3390/ijerph14121569
61. Svobodova I, Bobakova DF, Boskova L, Veselska ZD. How to improve access to health care for Roma living in social exclusion: a concept mapping study. *Int J Equity Health.* (2021) 20:61. doi: 10.1186/s12939-021-01396-4
62. de Graaf P, Rotar Pavlič D, Zelko E, Vintges M, Willems S, Hanssens LGM. Primary care for the Roma in Europe: positon paper of the European forum for primary care. *Zdr Varst.* (2016) 55:218–24. doi: 10.1515/sjph-2016-0030
63. Katona C, Gutási E, Papp M, Varga O, Kósa K. Facilitating equal access to primary care for all: work experiences of health mediators in a primary health care model programme in Hungary. *BMC Fam Pract.* (2020) 21:212. doi: 10.1186/s12875-020-01281-z
64. Brakema EA, van der Kleij R.M., Vermond D, van Gemert FA, Kirenga B. Let's stop dumping cookstoves in local communities. It's time to get implementation right. *NPJ Prim Care Resp Med.* (2020) 30:3. doi: 10.1038/s41533-019-0160-8
65. Brakema EA, van der Kleij RMJJ, Poot CC, An PL, Anastasaki M, Crone M, et al. Mapping low-resource contexts to prepare for lung health interventions in four countries (FRESH AIR): a mixed-method study. *Lancet Global Health.* (2022) 10:E63–76. doi: 10.1016/S2214-109X(21)00456-3
66. Paulik E, Nagymajtényi L, Easterling D, Rogers T. Smoking behaviour and attitudes of Hungarian Roma and non-Roma population towards tobacco control policies. *Int J Public Health.* (2011) 56:485–91. doi: 10.1007/s00038-011-0252-x
67. European Union Agency for Fundamental Rights. *Poverty and Employment: The Situation of Roma in 11 EU Member States.* Luxembourg. (2014).
68. Miranda DE, Garcia-Ramirez M, Balcazar FE, Suarez-Balcazar Y, A. Community-based participatory action research for roma health justice in a deprived district in Spain. *Int J Environ Res Public Health.* (2019) 16:3722. doi: 10.3390/ijerph16193722
69. Bobakova DF. Challenges for research, policy and practice in the field of Roma health. *Int J Public Health.* (2019) 64:645–6. doi: 10.1007/s00038-019-01254-9
70. Boskova L, Madarasova Geckova A, van Dijk JP, Reijneveld SA. Appropriate employment for segregated roma: mechanisms in a public–private partnership project. *Int J Environ Res Public Health.* (2020) 17:3588. doi: 10.3390/ijerph17103588

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: All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

Copyright © 2022 Anastasaki, van Bree, Brakema, Tsiligianni, Sifaki-Pistolla, Chatzea, Crone, Karelis, van der Kleij, Poot, Reis, Chavannes and Lionis. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

Frontiers in Public Health

ORIGINAL RESEARCH
published: 09 May 2022
doi: 10.3389/fpubh.2022.796937

Empirical Analysis of Health Assessment Objective and Subjective Methods on the Determinants of Health

Abdou Khadre Dit Jadir Fall[1,2]*, *Florence Migot-Nabias*[1] *and Najoua Zidi*[2]

[1] *Université Paris Cité, Institut de Recherche pour le Développement (IRD), Mère et Enfant en Milieu Tropical (MERIT), Paris, France,* [2] *Laboratoire d'Economie Dionysien (LED), EA 3391, Université Paris VIII, Saint-Denis, France*

Background: There are several methods for assessing health status. The aims of this study were to investigate the empirical differences between health assessment objective and subjective methods, to identify a possible long-term relationship between methods and health determinants and the influence of these methods on the perceived level of risk according to health determinants.

Methods: Using data from 1970 to 2018 in the United States, health status was assessed by perception of health, absence from work due to self-reported illness, life expectancy at birth and mortality rate. Health determinants were tobacco and alcohol consumptions, number of physicians per 1,000 persons, stay in hospitalization unit, curative care, release of greenhouse gases, per capita gross domestic product (GDP) and urbanization. The differences between health objective and subjective assessment methods were investigated through a Generalized linear model, a structural break date of health methods was investigated by Chow test and the long-term relationship between health assessment methods and health determinants by Engle and Granger cointegration test.

Results: Tobacco consumption was associated with a decrease of life expectancy while no long-term causal relationship was found between them. There was a positive correlation between alcohol consumption and perception of good health with a long-term causal relationship. Although per capita GDP positively influenced life expectancy, there was no cointegration between them. The release of greenhouse gases was positively correlated with both the absence from work due to self-reported illness and the perception of good health. Finally, curative care was associated with a decrease of mortality and absence from work due to self-reported illness and an increase of life expectancy and perception of good health while hospitalization is positively correlated with mortality and negatively correlated with life expectancy with a long-term causal relationship. Finally, the number of physicians per 1,000 persons was not correlated with health assessment methods used.

OPEN ACCESS

Edited by:
Miodraga Stefanovska-Petkovska, Universidade de Lisboa, Portugal

Reviewed by:
Andreia Silva Costa, ISAMB & ESEL, Portugal
Guvenc Kockaya, ECONiX Research, Analysis and Consultancy Plc., Turkey

***Correspondence:**
Abdou Khadre Dit Jadir Fall
fallka@yahoo.fr

Specialty section:
This article was submitted to Life-Course Epidemiology and Social Inequalities in Health, a section of the journal Frontiers in Public Health

Received: *18 October 2021*
Accepted: *28 March 2022*
Published: *09 May 2022*

Citation:
Fall AKDJ, Migot-Nabias F and Zidi N (2022) Empirical Analysis of Health Assessment Objective and Subjective Methods on the Determinants of Health. Front. Public Health 10:796937. doi: 10.3389/fpubh.2022.796937

Conclusion: Our results highlight the influence of health assessment methods on the determinants of health and the fact that the perceived risk of health determinants changes according to the method used. Thus, the impact of health assessment methods must be considered in order to prioritize the determinants of health.

Keywords: health determinant, objective and subjective health assessment methods, empirical analysis, cointegration, long-term causal relationship

INTRODUCTION

WHO defines health as a complete state of physical, mental and social wellbeing and not merely as the absence of disease or infirmity (1). Questions remain on how to properly measure health status. According to Ware (2), health status assessment is useful to evaluate the efficiency or effectiveness of medical interventions, the quality of care and population need. Several methods are used to assess the health status.

There are the subjective methods based on the perception of health status and self-assessment questionnaires. Generally, the perception of health is based on graduated responses such as "very good," "good," "average," "bad," and "very bad." While the self-assessment questionnaires select one or more health dimensions such as morbidity, heart difficulties, high blood pressure… They are usually constructed using methods according to the subjects and studies. Several concepts have been developed in relation to these methods such as the subjective wellbeing. It is based on positive concepts including happiness, life satisfaction, morale, self-esteem, autonomy dimensions (3). A large number of systematic reviews have been published as regards these questionnaires measuring a specific concept in a specific population group (4–7). However, there is still a bias related to the reproducibility, reliability and validity of methods and results. Engström and Holmlund (8) showed that individuals in the low socioeconomic group tended to underestimate their need of dental care, while according to Maddox and Douglass (9), health status self-estimation is credible, effective and tends to be a better predictor of health status in the future. Wolinsky and Johnson (10) showed that seniors who report poor health are more likely to die than those who report good health. Finally, according to Barsky et al. (11), there was a correlation between the perception of health and aggregate medical morbidity, psychiatric morbidity, functional disability and hypochondriacal attitudes. These studies, despite the questions about reproducibility and reliability, showed that subjective methods can be considered as a reliable criterion in assessing health status.

Health status is, also, measured using pathological or clinical measures such as signs, symptoms, blood pressure, temperature … Indeed, health status is assessed based on reported, diagnosed diseases and the frequency of chronic diseases. Several concepts and theories have been developed in connection with this method (12). Among the concept, there is the functional ability which focused on impairment (loss or abnormality of psychological, physiological or anatomical structure or function), disability (restriction or lack of ability to perform an activity), handicap, and mental health. Mental health is measured by the SF-36 Mental Health Dimension Score indicator called MH (13–15).

There is heterogeneity in the assessment of health status. Measurement methods can be ranged from the most general evaluating biological parameters, to the most specific focused on particular aspects such as disability or mental health.

The first objective of our study was to assess the empirical differences between the two health assessment methods generally used, namely the objective and subjective methods (16, 17). The indicators for the subjective method were the perception of health status and the absence from work due to self-reported illness and the indicators for the objective method were the life expectancy at birth and the mortality rate. The influence of these methods is analyzed on the same determinants of health status selected among medical determinants (physicians per 1,000 persons, average length of stay in hospitalization units and the bed occupancy rates in curative care), non-medical determinants (alcohol and tobacco consumption), population (urbanization), economic [per capita Gross Domestic Product (GDP)], and environmental (release of greenhouse gases) variables.

The second objective of our study was to evaluate a possible existence of a long-term relationship (a correlation over time) between perception of health status, absence from work due to self-reported illness, life expectancy at birth and mortality rate, and health determinants.

For this purpose, this study used data from the United States. This choice is explained by several reasons. First of all, the United States hosts the most expensive healthcare system in the world, with strong disparities, and a health insurance system heavily dependent on employment which excludes the unemployed (18–22). Moreover, some individuals combine several jobs but are still categorized as poor, and named the "working poor" (23–25). Ross et al. (26) showed a negative relationship between older working poor and the receiving of preventive care while Miller et al. (27) showed that low income and insured men are under-diagnosed and under-treated for prostate cancer. At the opposite, Mahal et al. (28) confirmed the fact that insured men with prostate cancer were more likely to be treated and to survive compared to non-insured.

Thus, this study can target the most important determinants which emerge depending on the method used. Consequently, it would be interesting to better appreciate the individual's health status for better care and reimbursement from insurance. Moreover, the risk perceived according to the determinants of health can modify the behavior of the individual toward these determinants and consequently his general health status.

METHODS

Study Design

The study was conducted using data, collected from databases OECD, World bank and Perspective monde, in the United States during the period 1970–2018.

Health status was evaluated by objective indicators including life expectancy at birth (average number of years that a newborn is expected to live if current mortality rates continue to apply) and mortality rate (number of deaths in the year per 1,000 people and estimated at mid-year). The subjective indicators were the absence from work due to self-reported illness (number of days lost per person per year due to an absence from work resulting for a self-reported illness) and the perception of health status (percentage of the population aged 15 and over in good health).

Non-medical health determinants were cigarette consumption [annual consumption of tobacco (cigarettes, cigars) in grams per person aged 15 and over] and alcohol consumption [annual consumption of pure alcohol (beer, wine, spirits, others) in liters per person aged 15 and over]. Health care resources were the number of physicians per 1,000 persons, the average length of stay in hospitalization units (calculated by dividing the number of bed-days by the number of discharges during the year) and the percentage of available beds in curative care (curative care). Curative care comprises health care contacts during which the principal intent is to relieve symptoms of illness or injury, to reduce the severity of an illness or injury, or to protect against exacerbation and/or complication of an illness or injury that could threaten life or normal functions.

The environmental variable was represented by the carbon dioxide emissions (metric tons per capita). Emissions were defined as the release of greenhouse gases or precursors of greenhouse gases into the atmosphere over an area and over a period of time. The calculation was made here by dividing carbon dioxide emissions in metric tons (1,000 kg) by the total number of inhabitants.

The last variables were the per capita gross domestic product (US dollars GDP/capita) and the percentage of population living in agglomerations counting more than one million residents.

Statistical Analysis and Econometric Methods

The statistical analysis was carried out using the Gretl software version. First of all, data of health assessment methods and health determinants are presented. Then, the empirical differences between health assessment methods and health determinants were assessed using Generalized linear model (GLM) [recommended against autocorrelation and heteroscedasticity problems] which estimated the following equations:

Health status, determined by "absence from work due to self-reported illness; perception of health status; life expectancy at birth; mortality rate" is correlated to "tobacco consumption, alcohol consumption, number of physicians per 1,000 persons, average length of stay in hospitalization, bed occupancy rates in curative care, carbon dioxide emission, per capita GDP, urbanization."

Then, the stability of health assessment methods for which data showed a change in the study period (life expectancy and mortality methods) was tested for different periods through a Chow test in order to determine a structural break date in their evolution. For this purpose, first of all, a Quandt Likelihood Ratio (QLR) test is performed in order to look for a break date. Then, a Chow test is performed using a Fisher (F) test. Finally, the means of the tested variables between before and after the break date were compared through a Mann-Whitney test.

Finally, the cointegration test was used to evaluate a long-term relationship between control variables and health assessment methods (29, 30). In this study, the Engle-Granger cointegration test between two variables was used. For this purpose, the following steps have been covered:

The stationarity (order of integration) of the variables was tested through a Dickey Full Augmented (ADF) test. The variables were differentiated in a case of no stationarity. Two variables are, potentially, cointegrated if they have the same order of integration. In this case, the Engel and Granger's method was applied to study the cointegration between the two variables. First of all, we checked that the unit root hypothesis was not rejected for the individual variables tested (step 1). Then, we checked that the unit root hypothesis was rejected for the residues of the cointegration regression (step 2 and 3) in a case of cointegration. Finally, in case of cointegration, an error correction model (ECM) was estimated with the linear variable (differentiated variable) (step 4). If the residue (e) was significant and negative, therefore, there was a long-term relationship between the two variables tested.

TABLE 1 | Characteristics of health status and determinants of health in the United States during the 1970–2018 period.

	Mean	Min–Max
Health assessment subjective method		
Absence from work due to self-reported illness (Day)	4.67	3.5–5.60
Perception of health status (%)	87.88	86–88.90
Health assessment objective method		
Life expectancy at birth (Year)	75.77	70.90–78.90
Mortality rate (%)	8.59	7.90–9.50
Non-medical determinants of health		
Tobacco consumption (gram)	2245.28	1061–3606
Alcohol consumption (liter)	9.12	8.1–10.4
Health care resources		
Physicians per 1,000 inhabitants	2.05	1,20–2,71
Average length of stay in hospitalization (Day)	8.51	6.10–14.9
Rate of available beds in curative care (%)	67.77	61.50–78
Economic variable		
Per capita GDP (USD/capita)	30546.31	5234.3–65280.7
Population		
Urbanization (%)	77.20	73.60–82.26
Environment variable		
Carbon dioxide emissions (metric tons/capita)	19.35	15.50–22.51

TABLE 2 | Generalized linear model results comparing subjective and objective measurement methods of health in the United States in 1970–2018.

Health status	**Subjective method**		**Objective method**	
Health determinants	**Absence from work due to illness**	**Perception of Good health**	**Life expectancy at birth**	**Mortality rate**
Tobacco consumption	Coef (std. err.): 0.199 (0.130)	−0.010 (0.014)	−0.011 (0.006)	−0.013 (0.0511)
	P. value: 0.127	0.470	**0.075***	0.791
Alcohol consumption	Coef (std. err.): – 0.116 (0.304)	0.067 (0.040)	0.0149 (0.014)	−0.096 (0.119)
	P. value: 0.703	**0.100***	0.317	0.418
Physicians per 1000 persons	Coef (std. err.): 0.042 (0.173)	−0.006 (0.017)	−0.013 (0.008)	0.048 (0.068)
	P. value: 0.808	0.688	0.110	0.476
Stay in hospitalization units	Coef (std. err.): 0.747 (0.225)	0.045 (0.070)	−0.054 (0.011)	0.200 (0.088)
	P. value: **0.001*****	0.516	**0.0001*****	**0.023****
Available beds in curative care	Coef (std. err.): – 1.153 (0.330)	−0.106 (0.039)	0.030 (0.016)	−0.260 (0.129)
	P. value: **0.0001*****	**0.008*****	**0.060***	**0.045****
Per capita GDP	Coef (std. err.): 0.187 (0.095)	0.006 (0.021)	0.023 (0.004)	−0.040 (0.037)
	P. value: **0.050****	0.762	**0.0001*****	0.273
Urbanization	Coef (std. err.): – 1.167 (1.289)	0.358 (0.630)	0.013 (0.063)	0.237 (0.504)
	P. value: 0.365	0.570	0.831	0.638
Carbon dioxide emission	Coef (std. err.): 0.568 (0.255)	0.121 (0.042)	−0.018 (0.012)	0.004 (0.043)
	P. value: **0.026****	**0.004*****	0.129	0.912

****P ≤ 0.01, **P ≤ 0.05, *P ≤ 0.10; In bold, significant P value.*

TABLE 3 | Chow test in the period 1970–2018.

	Life expectancy at birth	**Mortality rate**
Breaking point identified by the QLR test	2001 $F_{(9,21)} = 6.088$	2000 $F_{(9,21)} = 4.897$
Chow test	$F_{(9,21)} = 6.088$	$F_{(9,21)} = 4.897$
	P $F_{(9,21)} > 6.0851 = \mathbf{0.0003}^{***}$	P $F_{(9,21)} > 4.897 = \mathbf{0.0012}^{***}$
Mean comparison	Mean 1970–2000: 74.37	Mean 1970–1999: 8.79
	Mean 2002–2018: 78.25	Mean 2001–2018: 8.26
Mann–Whitney *U* test	$P < \mathbf{0.0001}^{***}$	$P < \mathbf{0.0001}^{***}$

****P ≤ 0.01; In bold, significant P value.*

RESULTS

Characteristics of Health Status and Determinants

According to **Table 1**, life expectancy at birth remained relatively moderate around 76 years in the United States despite an average annual absence from work of 5 days and a large majority of population aged 15 and over reporting being in good health (88%). A mean quantity of 2,245 g of tobacco (cigarettes, cigars) and 9 L of pure alcohol (beer, wine, spirits, others) were consumed during the study period. On average, patients spent 8 days in hospital while 2/3 of beds were available on curative care. A large part of the population (77%) lived in cities, emitted 19 tons of carbon dioxide per year with a relatively high GDP per capita (30 546 USD).

Association Between Health Determinants and Subjective Measurement Method

According to **Table 2**, the GDP per capita, the carbon dioxide emission and the stay in hospital units were positively correlated to the absence from work due to self-reported illness ($P = 0.050$, $P = 0.026$, and $P = 0.001$, respectively) while more beds available in curative care was negatively correlated with the absence from work related to self-reported illness care ($P = 0.0001$). There was a positive correlation between release of greenhouse gases and perception of health ($P = 0.004$) as well as with alcohol consumption ($P = 0.100$). For its part, tobacco did not appear to have an impact on both the absenteeism from work due to self-reported illness and the perception of health status. Finally, results showed that the perception of good health increased with the curative care ($P = 0.008$).

Association Between Health Determinants and Objective Measurement Method

The stay in hospitalization units was negatively correlated with life expectancy at birth as well as tobacco consumption ($P = 0.0001$ *and* $P = 0.075$, respectively) while there was a positive correlation between the availability of bed in curative care as well as per capita GDP and life expectancy ($P = 0.060$ *and* $P = 0.0001$, respectively). More beds available in curative care was negatively correlated with the mortality rate while there was a positive correlation between the stay in hospitalization units and the mortality rate ($P = 0.045$ *and* $P = 0.023$, respectively).

Structural Break Tests

As shown in **Table 3**, QLR test showed a break date in 2001 regarding the life expectancy at birth, confirmed by the Chow test. The Mann—Whitney *U* test confirmed an increase of the

TABLE 4 | Engle Granger cointegration test between health status perception and alcohol consumption and bed occupancy rate in the period 1982–2018.

	Ln perception of health status	Ln alcohol consumption	Ln available beds in curative care
Dickey-Fuller tests on first difference			
Null hypothesis of unit root: a = 1			
Test without constant			
T statistics	−4.920	−3.078	−3.897
P	*P* < 0.0001	0.002	*P* < 0.0001
Test with constant			
T statistics	−4.877	−3.070	−3.970
P	0.0003	0.028	0.0015
Test with constant and time trend			
T statistics	−4.947	−3.076	−3.975
P value	**0.0016*****	0.111	**0.009*****
Engel and Granger cointegration test			
Step 1: Testing a unit root			
Without constant	*P* value: 0.908	*P* value: 0.528	*P* value = 0.336
Step 2: cointegration regression			
Dependent variable		Coef. 2.054	Coef. 1.071
Health status perception		***P* value: <0.0001*****	***P* value: <0.0001*****
Step 3: Dickey-Fuller regression			
Lag 1			
Null hypothesis of unit root: a = 1		**P. asymptotic value: 0.045****	**P. asymptotic value: <0.0001*****
Step 4: Error correction model			
Dependent variable		First difference ln alcohol consumption:	First difference ln Available beds in curative care
First difference Health status perception		*Coefficient:* −0.011	*Coefficient:* −0.00013
		P value: 0.707	*P* value: 0.804
		e_1	**e_1**
		Coefficient: **−0.347**	*Coefficient:* **−0.345**
		P value: **0.004*****	*P* value: **0.007*****
		d_lnpes_1	d_lnpes_1
		Coefficient: 0.345	*Coefficient:* 0.199
		P value: **0.026****	*P* value: 0.180

***P ≤ 0.01, **P ≤ 0.05; In bold, significant P value.

life expectancy after 2001 through the mean comparison between the two periods. Indeed, before the break date, the mean age of life expectancy was 74 and 78 years after. The same pattern was observed with mortality rate through a decrease of the mortality rate after the break date 2000.

Long-Term Relationship Between Health Assessment Methods and Health Determinants

In **Tables 4–6**, only variables with the same order of integration, so a possibility of a long-term relationships, were shown.

Absence from work due to illness, stay in hospitalization units, curative care and per capita GDP series were all integrated in order 1. However, according to the Engle and Granger cointegration test, there was not a long-term relationship between absence from work due to illness and the health determinants stay in hospitalization units, curative care and per capita GDP series (results not shown).

Even, according to **Table 4**, there was a long-term relationship between perception of health status and curative care (*P* value of Dickey-Fuller regression < 0.0001) as well as alcohol consumption (*P* value of Dickey-Fuller regression = 0,045). Correction Error Model showed a negative and significant coefficient (e) for both.

Engle and Granger cointegration test showed a cointegration between mortality rate and curative care (*P* value of Dickey Fuller regression = 0.038) while the Correction Error Model showed a negative but not significant coefficient (e). Thus, there was not a long-term causal relationship between the mortality rate and curative care (**Table 5**).

Finally, cointegration test showed a long-term causal relationship between the life expectancy and stay at hospitalization unit. Indeed, the *P* value of Dickey Fuller regression was significant (*P* = 0.002) and the residue (e) was negative and significant (**Table 6**).

DISCUSSION

This article investigated the empirical differences between health assessment objective and subjective methods on health

TABLE 5 | Engle Granger cointegration test between mortality rate and bed occupancy rate in the period 1970–2018.

	Ln mortality rate	**Ln available beds in curative care**
Augmented Dickey-Fuller tests on first difference Null hypothesis of unit root: a = 1		
Test without constant		
T statistics	−8.259	−3.897
P value	***P* < 0.0001*****	***P* < 0.0001*****
Test with constant		
T statistics	−8.278	−3.970
P value	***P* < 0.0001*****	**0.0015****
Test with constant and time trend		
T statistics	−8.652	−3.975
P value	***P* < 0.0001*****	**0.009*****
Engel and Granger cointegration test		
Step 1: Testing a unit root		
Without constant	*P* value: 0.314	*P* value: 0.336
Step 2: Cointegration regression		
Dependent variable		Coef. 0.510
Mortality rate		***P* value: <0.00001*****
Step 3: Dickey-Fuller regression		
Lag 1 Null hypothesis of unit root: a = 1		**P. asymptotic value: 0.038*****
Step 4: Error correction model		
Dependent variable		First difference ln available beds in curative care:
First difference LN mortality rate		*Coefficient:* −0.001 *P* value: 0.320 **e_1** *Coefficient:* −0.112 *P* value: 0.109 d_lnmortality rate_1 *Coefficient:* −0.137 *P* value: 0.340

$^{***}P \leq 0.01$, $^{**}P \leq 0.05$; In bold, significant P value.

TABLE 6 | Engle Granger cointegration test between life expectancy and stay in hospitalization unit in the period 1970–2018.

	Ln life expectancy	**Ln stay in hospitalization unit**
Augmented Dickey-Fuller Tests on First difference Null hypothesis of unit root: a = 1		
Test without constant		
T statistics	−2.447	−3.089
	0.013	0.002
Test with constant		
T statistics	−5.935	−3.520
P value	***P* < 0.0001*****	**0.011*****
Test with constant and time trend		
T statistics	−6.815	−3.704
P value	***P* < 0.0001*****	**0.031*****
Engel and Granger cointegration test		
Step 1: Testing a unit root		
With constant	***P* value: 0.020****	*P* value: 0.430
Step 2: Cointegration		
Dependent variable	Const (*p* value) of regression: 4.575	Coef. −0.118
Life expectancy at birth	**(<0.00001***)**	***P* value: <0.00001*****
Step 3: Dickey-Fuller regression		
Lag 1 Null hypothesis of unit root: a = 1		**P. asymptotic value: 0.095***
Step 4: Error correction model		
Dependent variable		First difference ln stay in hospitalization:
First difference life expectancy		*Coefficient:* −0.061 *P* value: **0.002***** e_1 *Coefficient:* –**0.148** *P* value: **0.076** d_life expectancy_1 *Coefficient:* 0.146 *P* value: 0.309

$^{***}P \leq 0.01$, $^{**}P \leq 0.05$, $^{*}P \leq 0.10$; In bold, significant P value.

determinants and a possible existence of a long-term relationship between them in the United States.

We found an increase of the life expectancy and a decrease of the mortality rate in the 2000s with specific dates highlighted by QLR, Chow and Mann-Whitney tests. These results were in line with those of Woolf and Schoomaker (31) and Mokdad et al. (32). However, this increase in life expectancy is not as elevated as in countries such as France or the United Kingdom. Access to healthcare may play a key factor insofar as in the United States, this access is conditioned by employment. Despite the fact that European countries have a high per capita GDP, we can hypothesize an influence of GDP in these countries but not in the United States.

Indeed, Swift (33) estimated that total GDP and per capita GDP exerted a significant influence on life expectancy for most European countries (1% increase in life expectancy resulting from 6% increase in total GDP) while according to Zaman et al. (34), the relationship between GDP and life expectancy may be explained by a direct relationship between GDP and health government expenditure. However, in the United States, government's intervention is limited. Thus, the finding that there was no long-term relationship between life expectancy and per capita GDP (results not shown) in the

United States was in line with this. Moreover, the United States is the richest state in the world but it remains far behind other states in terms of life expectancy, ranging for example at the 18th position for life expectancy of women, among the 30 OECD (Organization for Economic Co-operation and Development) countries, slightly above Greece, Korea, and Mexico (35).

Our results showed that life expectancy in the United States was negatively associated with tobacco consumption while there was not a long-term causal relationship between them in the period 1970–2018 (results not shown). Indeed, many papers concluded that the most relevant health indicators for poor populations from the United states were obesity, alcohol and tobacco consumption (35, 36). Holford et al. (37) estimated 17.6 million deaths related to smoking from 1964 to 2012, in the continuity of what Rogers and Powell-Griner (38) described formerly with a higher life expectancy for no smokers compared to former smokers and for former smokers compared to current smokers. Mokdad et al. (32) considered tobacco consumption, high body mass index and alcohol consumption as the top risk factors for diminished life expectancy.

Compared to European countries, tobacco consumption is a concern, although it remains moderate in Scandinavian countries (17% in Denmark, 16% in Finland, 15% in Norway, and 11% in Sweden) to which is attributed a reputation for better health systems in link with better levels of health indicators (39).

The results showing a long-term positive relationship between alcohol consumption and perception of health may be related to the indicator used and also the personal beliefs. Thus, according to Chang et al. (40), 80% of people in Helsinki believe that drinking red wine is healthier than drinking beer or spirits. Also, Strandberg et al. (41) reported that red wine drinkers had a 34% lower mortality rate than beer or vodka drinkers. So, on the one hand, there is a popular belief in the health benefits of certain types of alcohol and on the other, the fact that no alcohol consumption can prevent serious illnesses related to its consumption, therefore for avoiding early mortalities. This paradox was a perfect illustration of the differences found for some health determinants according to the method used for their assessment. The same pattern was found for the influence of releasing greenhouse gases on the absence from work due to self-reported illness and on the perception of health status.

On the one hand, the finding of a positive effect may be explained by the fact that individuals move, go to their occupation (work, leisure) therefore improve their wellbeing because they feel healthy. On the other hand, a negative effect may be deduced in case of illness due to pollution, that will prevent people from working and hence increase their absence from work due to self-reported illness.

The finding (i) that the stays at hospitalization unit was negatively associated with life expectancy and positively associated with mortality rate and (ii) that curative care was related to increased life expectancy and perception of good health and to decreased mortality and absence from work due to self-reported illness as well as was rather expected. Over the long term, receiving curative care can reduce the severity of an illness or injury, protect against exacerbation and/or complication of an illness or injury while the longer a hospitalization stay is, the greater the risk of mortality, as reflecting a serious case of concern. Moreover, the finding that receiving curative care influenced perception of health care status was explained by the positive virtues of receiving curative care.

Limitation

It may be interesting to conduct this study in a multi-country panel to confirm the results observed in the United States or to constitute a comparative panel between poor and rich countries in order to compare the influence of health determinants according to the status of countries. Also, cointegration studies are more interesting over long periods.

CONCLUSION

To our knowledge, no work previously investigated the empirical differences between health assessment objective and subjective methods with a long-term causal relationship analysis. Our results highlight the fact that the determinants of health change according to the health status assessment method used, with an impact on the long-term relationship between health indicators of the methods and health determinants. Non-medical determinants were the most affected. For examples, tobacco consumption was associated to a decrease of life expectancy but there was not a long-term relationship while there was a positive correlation between alcohol consumption and perception of good health with a long-term causal relationship. Air pollution was positively correlated to absence from work du to self-reported illness and perception of good health while there were no long-term relationships between them. In contrast, whatever the methods, medical determinants play an important role. Thus, the impact of health assessment methods must be considered in order to prioritize the determinants of health.

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.

AUTHOR CONTRIBUTIONS

AF, FM-N, and NZ wrote the article. All authors reviewed the manuscript and approved it for publication.

ACKNOWLEDGMENTS

The authors are grateful to Stéphane Rossignol (Professor at Université Paris VIII, LED EA 3391) for his reading and corrections.

REFERENCES

1. World Health Organization. *Constitution of the World Health Organization IN: Basic Documents and Geneva WHO* (1948).
2. Ware JE Jr, Brook RH, Davies AR, Lohr KN. Choosing measures of health status for individuals in general populations. *Am J Public Health.* (1981) 71:620–5. doi: 10.2105/AJPH.71.6.620
3. Bowling A. *Measuring Health: A Review of Quality-Of-Life Measurement Scales. 3rd Edn*. Maidenhead: Open University Press (2005). p. 211.
4. Daker-White G. Reliable and valid self-report outcome measures insexual (dys) function: a systematic review. *Arch Sex Behav.* (2002) 31:197e209. doi: 10.1023/a:1014743304566
5. De Boer MR, Moll AC, de Vet HC, Terwee CB, Volker-Dieben HJ, van Rens G. H. Psychometric properties of vision-related quality oflife questionnaires: a systematic review. *Ophthalmic Physiol Opt*. (2004) 24:257–73. doi: 10.1111/j.1475-1313.2004.00187.x
6. Hallin P, Sullivan M, Kreuter M. Spinal cord injury and quality of lifemeasures: a review of instrument psychometric quality. *Spinal Cord.* (2000) 38:509–23. doi: 10.1038/sj.sc.3101054
7. Edwards B, Ung L. Quality of life instruments for caregivers of patients with cancer. *Cancer Nurs.* (2002) 25:342–9. doi: 10.1097/00002820-200210000-00002
8. Engström S, Holmlund A. Self-estimated oral and general health are related and associated with clinically investigated dental health. *Swed Dent J.* (2011) 35:169–75.
9. Maddox GL, Douglass EB. Self-assessment of health: a longitudinal study of elderly subjects. *J Health Soc Behav.* (1973) 14:87–93. doi: 10.2307/2136940
10. Wolinsky FD, Johnson RJ. Perceived health status and mortality among older men and women. *J Gerontol.* (1992) 47:S304–S312. doi: 10.1093/geronj/47.6.S304
11. Barsky AJ, Cleary PD, Klerman GL. Determinants of perceived health status of medical outpatients. *Soc Sci Med.* (1992) 34:1147–54. doi: 10.1016/0277-9536(92)90288-2
12. Guillemin F, Bombardier C, Beaton D. Cross-cultural adaptation of health-related quality of life measures: literature review and proposed guidelines. *J Clin Epidemiol*. (1993) 46:1417–32. doi: 10.1016/0895-4356(93)90142-N
13. Strand BH, Dalgard OS, Tambs K, Rognerud M. Measuring the mental health status of the Norwegian population: a comparison of the instruments SCL-25, SCL-10, SCL-5 and MHI-5 (SF-36). *Nord J Psychiatry*. (2003) 57:113–8. doi: 10.1080/08039480310000932
14. Clements-Nolle K, Marx R, Guzman R, Katz M. HIV prevalence, risk behaviors, health care use, and mental health status of transgender persons: implications for public health intervention. *Am J Public Health.* (2001) 91:915. doi: 10.2105/AJPH.91.6.915
15. Ware JE Jr, Manning WG Jr, Wells KB, Duan N, Newhouse JP. Health status and the use of outpatient mental health services. *Am Psychol.* (1984) 39:1090–100. doi: 10.1037/0003-066X.39.10.1090
16. Couffinhal A, Dourgnon P, Tubeuf S. *Outils de mesure des inégalités de santé : Quelques débats d'actualité (Santé, Société et Solidarité, Edition 2)* (2004). doi: 10.3406/oss.2004.1008
17. Devaux M, Jusot F, Sermet C, Tubeuf S. Hétérogénéité sociale de déclaration de l'état de santé et mesure des inégalités de santé. *Revue française des affaires sociales.* (2008) 1, 29–47. doi: 10.3917/rfas.081.0029
18. Zimlichman E, Henderson D, Tamir O, Franz C, Song P, Yamin, et al. Health care–associated infections: a meta-analysis of costs and financial impact on the US health care system. *JAMA Intern Med.* (2013) 173:2039–204. doi: 10.1001/jamainternmed.2013.9763
19. Andersen RM, Rice TH, Kominski GF. *Changing the US Health Care System: Key Issues in Health Services Policy and Management*. John Wiley and Sons (2011).
20. Ruggie M. The paradox of liberal intervention: health policy and the American welfare state. *Am J Sociol.* (1992) 97:919–44. doi: 10.1086/229856
21. Sowell T. *Black Rednecks and White Liberals: Hope, Mercy, Justice and Autonomy in the American Health Care System*. Encounter Books (2009).
22. Wall AL. *Health Care Systems in Liberal Democracies*. Psychology Press (1996).
23. Duncan GJ, Huston AC, Weisner TS. *Higher Ground: New Hope for the Working Poor and Their Children*. Russell Sage Foundation (2007).
24. Roberto E. *Commuting to Opportunity: The Working Poor and Commuting in the United States* (2008).
25. Zuberi D. *Differences That Matter: Social Policy and the Working Poor in the United States and Canada*. Seattle, WA; Vancouver, BC: Cornell University Press (2006). doi: 10.7591/9781501711251
26. Ross JS, Bernheim SM, Bradley EH, Teng HM, Gallo WT. Use of preventive care by the working poor in the United States. *Prev Med.* (2007) 44:254–9. doi: 10.1016/j.ypmed.2006.11.006
27. Miller DC, Litwin MS, Bergman J, Stepanian S, Connor SE, Kwan L, et al. Prostate cancer severity among low income, uninsured men. *J Urol.* (2009) 181:579–84. doi: 10.1016/j.juro.2008.10.010
28. Mahal BA, Aizer AA, Ziehr DR, Hyatt AS, Lago-Hernandez C, Chen YW, et al. The association between insurance status and prostate cancer outcomes: implications for the Affordable Care Act. *Prostate Cancer Prostatic Dis.* (2014) 17:273–9. doi: 10.1038/pcan.2014.23
29. Engle RF, Granger CWJ. Cointegration and error correction: representation, estimation, and testing, econometrica. *Econom Soc.* (1987) 55: 251–76. doi: 10.2307/1913236
30. Johansen S. Estimation and hypothesis testing of cointegartion vectors in gaussian vector autoregressive models. *Econometrica*. (1991) 59:1551–80. doi: 10.2307/2938278
31. Woolf SH, Schoomaker H. Life expectancy and mortality rates in the United States, 1959-2017. *JAMA*. (2019) 322:1996–2016. doi: 10.1001/jama.2019.16932
32. Mokdad AH, Ballestros K, Echko M, Glenn S, Olsen HE, Mullany E, et al. The state of US health, 1990-2016: burden of diseases, injuries, and risk factors among US states. *JAMA*. (2018) 319:1444–72. doi: 10.1001/jama.2018.0158
33. Swift R. The relationship between health and GDP in OECD countries in the very long run. *Health Econ.* (2011) 20:306–22. doi: 10.1002/hec.1590
34. Zaman SB, Hossain N, Mehta V, Sharmin S, Mahmood SAI. An association of total health expenditure with GDP and life expectancy. *J Med Res Innov*. (2017) 1:AU7–12. doi: 10.15419/jmri.72
35. Munnell AH, Hatch. Robert E, Lee JG. *Why is Life Expectancy So Low in the United States?* Center for Retirement Research at Boston College (2004).
36. Stewart ST, Cutler DM, Rosen AB. Forecasting the effects of obesity and smoking on US life expectancy. *N Engl J Med.* (2009) 361:2252–60. doi: 10.1056/NEJMsa0900459
37. Holford TR, Meza R, Warner KE, Meernik C, Jeon J, Moolgavkar SH, et al. Tobacco control and the reduction in smoking-related premature deaths in the United States, 1964–2012. *JAMA.* (2014) 311:164–71. doi: 10.1001/jama.2013.285112
38. Rogers RG, Powell-Griner E. Life expectancies of cigarette smokers and nonsmokers in the United States. *Soc Sci Med.* (1991) 32:1151–9. doi: 10.1016/0277-9536(91)90092-Q
39. Maryse G. La santé des populations. *L'Europe en Formation*. (2015) 377:79–95. doi: 10.3917/eufor.377.0079
40. Chang KJ, Thach ML, Olsen J. Wine and health perceptions: exploring the impact of gender, age and ethnicity on consumer perceptions of wine and health. *Wine Econ Policy.* (2016) 5:105–13. doi: 10.1016/j.wep.2016.09.001
41. Strandberg TE, Strandberg AY, Salomaa VV, Pitkälä K, Tilvis RS, Miettinen TA. Alcoholic beverage preference, 29-year mortality, and quality of life

in men in old age. *J Gerontol A Biol Sci Med Sci.* (2007) 62:213–8. doi: 10.1093/gerona/62.2.213

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: All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

Copyright © 2022 Fall, Migot-Nabias and Zidi. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

frontiers | Frontiers in Public Health

ORIGINAL RESEARCH
published: 10 May 2022
doi: 10.3389/fpubh.2022.852971

Public Views About Involvement in Decision-Making on Health Data Sharing, Access, Use and Reuse: The Importance of Trust in Science and Other Institutions

Ngozi Nwebonyi[1,2], *Susana Silva*[3,4] *and Cláudia de Freitas*[1,2,5,6]*

[1] Laboratório para a Investigação Integrativa e Translacional em Saúde Populacional (ITR), Porto, Portugal, [2] Departamento de Ciências da Saúde Pública e Forenses e Educação Médica, Faculdade de Medicina da Universidade do Porto (FMUP), Porto, Portugal, [3] Departamento de Sociologia, Instituto de Ciências Sociais, Universidade do Minho, Braga, Portugal, [4] Centro em Rede de Investigação em Antropologia, Universidade do Minho, Braga, Portugal, [5] EPIUnit - Instituto de Saúde Pública, Universidade do Porto, Porto, Portugal, [6] Centre for Research and Studies in Sociology, University Institute of Lisbon (ISCTE-IUL), Lisbon, Portugal

Background: Data-intensive and needs-driven research can deliver substantial health benefits. However, concerns with privacy loss, undisclosed surveillance, and discrimination are on the rise due to mounting data breaches. This can undermine the trustworthiness of data processing institutions and reduce people's willingness to share their data. Involving the public in health data governance can help to address this problem by imbuing data processing frameworks with societal values. This study assesses public views about involvement in individual-level decisions concerned with health data and their association with trust in science and other institutions.

Methods: Cross-sectional study with 162 patients and 489 informal carers followed at two reference centers for rare diseases in an academic hospital in Portugal (June 2019–March 2020). Participants rated the importance of involvement in decision-making concerning health data sharing, access, use, and reuse from "not important" to "very important". Its association with sociodemographic characteristics, interpersonal trust, trust in national and international institutions, and the importance of trust in research teams and host institutions was tested.

Results: Most participants perceived involvement in decision-making about data sharing (85.1%), access (87.1%), use (85%) and reuse (79.9%) to be important or very important. Participants who ascribed a high degree of importance to trust in research host institutions were significantly more likely to value involvement in such decisions. A similar position was expressed by participants who valued trust in research teams for data sharing, access, and use. Participants with low levels of trust in national and international institutions and with lower levels of education attributed less importance to being involved in decisions about data use.

Conclusion: The high value attributed by participants to involvement in individual-level data governance stresses the need to broaden opportunities for public participation

OPEN ACCESS

Edited by:
Miodraga Stefanovska-Petkovska,
Universidade de Lisboa, Portugal

Reviewed by:
Brígida Riso,
Universidade de Lisboa, Portugal
Daniela Miranda,
Sevilla University, Spain

***Correspondence:**
Cláudia de Freitas
claudia.defreitas@ispup.up.pt

Specialty section:
This article was submitted to Life-Course Epidemiology and Social Inequalities in Health, a section of the journal Frontiers in Public Health

Received: *11 January 2022*
Accepted: *28 March 2022*
Published: *10 May 2022*

Citation:
Nwebonyi N, Silva S and de Freitas C (2022) Public Views About Involvement in Decision-Making on Health Data Sharing, Access, Use and Reuse: The Importance of Trust in Science and Other Institutions. Front. Public Health 10:852971. doi: 10.3389/fpubh.2022.852971

in health data decision-making, namely by introducing a meta consent approach. The important role played by trust in science and in other institutions in shaping participants' views about involvement highlights the relevance of pairing such a meta consent approach with the provision of transparent information about the implications of data sharing, the resources needed to make informed choices and the development of harm mitigation tools and redress.

Keywords: public involvement, data governance, trust, research trustworthiness, data sharing, data access, data reuse, rare diseases

INTRODUCTION

Health care quality improvement can be bolstered by data-intensive and needs-driven research (1). The use of big health data promises to transform biomedical and health care research and to deliver substantial public health benefits that range from disease risk prediction and prevention to the discovery of new therapies for untreatable health conditions, as are many rare diseases (2, 3). However, mounting reports of data breaches and mismanagement have generated concern for privacy loss, undisclosed surveillance, and discrimination (4–6). These concerns can undermine public trust in data processing organizations (e.g., governmental, care and research institutions), which is key in shaping public attitudes toward data sharing and use (7, 8). For instance, large-scale health data projects such as care.data and the Clinical Practice Research Datalink (CPRD) in the UK have failed to accomplish their goals because they could not achieve public trust and acceptability, despite promising benefits to health care and the public. Studies of the public's opinion suggest that these projects failed due to public concerns about informed consent, limited trust in data security and privacy, lack of communication on how data linkage would work, and the undisclosed involvement of commercial and private companies (7, 9, 10). Concerns such as these can evolve into a more generalized perceived lack of institutional trustworthiness, which can limit people's willingness to share their data for research and to concede to its (re)use (11–13). There is, therefore, an imminent need for optimizing governance strategies to promote safe, acceptable, and beneficial uses of data in health research.

International policy agencies recommend the involvement of the public to ensure that data processing frameworks are consistent with societal values and individuals' expectations for the protection and use of their data (14, 15). Public involvement is also substantiated by ethical arguments that center on the fair distribution of the benefits arising from data use (16, 17). Furthermore, it has been argued that public involvement exercises can help foster authentic dialogue between researchers and publics, enhance accountability among data stewards on the governance chain and increase research trustworthiness, all of which are vital for ensuring and sustaining public trust in science (7).

Public involvement in health data governance entails awareness raising, consultation, partnering with and/or empowering of members of the public to participate in research and governance practices and it can be set in motion through a variety of methods including deliberative polls, citizen juries, participatory appraisals, scenario-based workshops, and focus groups (18). Data holders can also participate via participant-led data cooperatives (e.g., Open Humans, PEER Network, MIDATA) that enable them to share and aggregate their data while keeping control over its uses (19–23).

At the individual level, public involvement can be fostered by enabling lay people to participate in decisions about particular aspects of data governance, including whether they want to share their data (*data sharing*), with whom they want to share their data with (*data access*), for what purposes it can be used (*data use*) and whether data can be shared for purposes other than those for which they were originally collected (*data reuse*). These individual-level decisions are typically enacted through different types of informed consent procedures (24, 25).

Broad consent offers data donors limited opportunities for decision-making beyond the initial decision of sharing data. In this type of consent potential data donors are asked consent to sharing data for purposes that may not yet be entirely specified but whose core aim is known to the public. As such, it differs from blanket consent in which shared data can be used without any restrictions (26, 27). Specific consent enables data donors to have more control over their data by enabling them to decide who uses the data and for what purposes, within the scope of a specific project or a set of similar research initiatives. Both broad and specific consent are requested at the moment people are asked to share data, usually at one single time-point. Dynamic consent, on the other hand, enables a higher degree of involvement in decision-making by allowing data donors to define and modify consent preferences over time, including decisions about the possible reuse of their data (28, 29). The latter implies the creation of interactive platforms that enable data subjects to be notified of requests to use their data and to be re-contacted to proceed with making a decision regarding consent (30, 31). Ploug and Holm (32) argue that adopting a dynamic consent approach can lead to the routinization of consent and even to "consent fatigue", as participants will likely receive large amounts of consent requests each requiring analysis of an entire project. Alternatively, they propose a meta consent model, which combines the broad and dynamic consent models with additional options for blanket consent and blanket refusal. In this type of model, data donors choose what type of consent (e.g., broad, blanket, dynamic, refusal) they would like to provide for the reuse of their data in future projects. Such a choice can be done both according to data type (e.g., electronic patient records, tissue, health data, non-health data) and to the context of data use (e.g., public vs. private, commercial vs. non-commercial, national vs. international). For instance, if data donors choose dynamic consent for health-data reuse by the commercial sector, they will be asked for specific consent each time data for a new project is requested in that domain. If instead they choose broad consent for electronic patient records reuse by public health institutions, they will be asked for consent only when a new project falls outside the scope of projects they already gave consent to (32).

Opportunities for public involvement have expanded substantially in the past decade and there is a growing interest in understanding whether patients, and other members of the public, value involvement in individual-level decisions about health data sharing, access, use and reuse (33–40). Ludman et al. (38) found that research participants wanted to decide whether their previously shared data could be submitted to a new database through active engagement in reconsenting procedures despite "their extraordinary trust in the research team" (32). Similarly, another study showed that patients would like to be re-contacted to decide on the reuse of their data and that not being given the opportunity to reconsent would be perceived as a threat to individual and group autonomy (33). Courbier et al. found that patients and their family members would like to keep control over their shared data and that about half would not delegate the decision about whom their data will be shared with to an ethics committee (34). And a study involving research participants in four European countries showed they were supportive of de-identified data reuse if they were involved in decision-making about data sharing and access, namely by retaining control to withdraw their data at any time (35).

Most existing studies address the multiple aspects of individual-level data governance independently and few have explored how trust in research initiatives influences the value bestowed by different publics on involvement in data decision-making (34, 38). In this study, we assess the views of rare disease patients and their informal carers about being involved in decisions regarding data sharing, access, use and reuse with a focus on the role played by trust in science and other institutions. Most rare diseases have no treatment and specific rare disease populations are very small and scattered geographically (34, 41). Data sharing within and between countries is therefore essential for enabling research that can advance the development of accurate diagnoses and therapies (42). However, this type of research requires a combination of genetic and phenotypic information which presents a high privacy risk for these patients and their relatives. Assessing rare disease patients and carers' views about involvement in decision-making concerned with their data can help in designing a data governance structure suitable to meet their needs and expectations from biomedical and health care research and to enhance the trustworthiness of institutions involved in research (16, 43).

MATERIALS AND METHODS

Participants

This observational and cross-sectional study is part of a mixed methods project focusing on public involvement in health data governance whose protocol is described elsewhere (44). For the purposes of this paper, participants include people with rare diseases and their informal carers who are both stakeholders directly involved in decisions regarding their own data sharing, access, use and reuse for biomedical and health care research. Participants were consecutively recruited from two Reference Centers for Rare Diseases at the University Hospital Center S. João (UHCSJ), in Portugal, between June 2019 and March 2020. Following a consultation, patients aged 12 years and above and their carers were handed a study information leaflet by a health professional. Subsequently, they were invited to participate in the study by a researcher who clarified any arising doubts or questions. Those who decided to participate were accompanied to a private setting, where they read and signed the informed consent. Underage participants who agreed to participate gave verbal consent and the informed consent form was signed by their legal representatives. All participants were asked to fill in a self-administered questionnaire individually.

Of the 728 people invited, 77 refused to take part in the study due to unwillingness to participate ($n = 37$), lack of time ($n = 34$), lack of consent from the legal tutor ($n = 3$), limited literacy ($n = 2$) and emotional distress following diagnosis ($n = 1$). In total, 651 people (162 patients and 489 carers) agreed to participate (response rate: 89.4%).

Data Collection

The structured questionnaire was developed by the research team based on a review of literature and existing instruments related to the research topic. The questionnaire was pretested by specialists with combined experience as professionals, informal carers and researchers (social and health sciences) and subsequently piloted by a group of patients and carers. The full questionnaire is available online [see (44)].

The assessment of the importance attributed by participants to involvement in decisions about their own health data sharing, access, use and reuse was based on the analysis of answers to four questions: 1) how important is it that you decide whether your data is shared for research purposes (*data sharing*); 2) how important is it that you decide whom your data is shared with (*data access*); 3) how important is it that you decide for what purposes your data is used for (*data use*); and, 4) how important is it that you decide whether your data can be used for purposes other than those for which it was initially collected (*data reuse*). The level of importance was rated using a 5-point Likert scale, ranging from "very important" to "not important" (range 1–5). For this analysis, the variables were categorized into "important" (including participants who answered "important" and "very important") and "other" (including "not important", "slightly important" and "moderately important"). This study included 637 participants

(159 patients and 478 carers), with available data on all the above-mentioned outcomes.

Data on sociodemographic characteristics (sex, age, educational level, marital status, occupation, and perceived income adequacy), as well as participants' involvement with patient organizations were collected. Occupations were classified according to the Portuguese Classification of Occupations 2011 (45) and grouped into four categories: (1) upper-white-collar, including executive civil servants, industrial directors and executives, professionals and scientists, middle management and technicians; (2) lower-white-collar, including administrative and related workers, service and sales workers; (3) blue-collar, which includes farmers and skilled agricultural workers, fisheries workers, skilled workers, craftsmen and similar, machine operators and assembly workers, and unskilled workers; and (4) other, including students, unemployed, domestic workers, participants on disability pension or on paid/unpaid leave, retired and informal carers or members of a foster family. Perceived income adequacy was measured through the question "When thinking of your household income, would you say that your household is able to make ends meet?". Participants could check one of the following answer categories: insufficient, caution with expenses, enough to make ends meet, and comfortable.

Interpersonal trust, trust in national institutions and trust in international institutions were measured through ten self-administered questions based on the European Social Survey (ESS) rated on a scale from 0 to 10. Interpersonal trust was measured by three questions: "Generally speaking, would you say that most people can be trusted or that you can't be too careful in dealing with people?"; "Do you think that most people would try to take advantage of you if they got the chance, or would they try to be fair?"; and "Would you say that most of the time people try to be helpful or that they are mostly looking out for themselves?". As reported in another study (46) principal component analysis to these three questions produced a single component, explaining 70% of the variance. Institutional trust was measured by asking participants how they trusted national institutions such as a country's parliament, the legal system, the police, politicians, and political parties, as well as international institutions, namely the European Parliament and the United Nations. Principal component analysis of the dataset shows that the variables are well suited for constructing two indexes, one for trust in national institutions and another for trust in international institutions. The total score of the rating scales is divided by the number of valid responses to make the indexes ranging from 0 to 10, with higher scores indicating higher levels of trust.

The views of patients and carers about the importance of trust in research host institutions and in research teams in decisions regarding data sharing were assessed using a 5-point Likert scale ranging from "not important" to "very important" (range: 0–4) for the question: "There are some aspects people consider important to decide if they will share their health data for scientific research. If you had to make such decision, how important would you rate the following aspects: (1) trust in the institution hosting the research; (2) trust in the team conducting the research". For this analysis, the answers were dichotomized as "very important" and "other" (all other answers).

Data Analysis

Categorical variables are presented as counts and proportions, while continuous variables were summarized as medians and interquartile range (P25–P75). The Chi-square test or the Fisher exact test, as well as the Mann-Whitney test were used, as appropriate, to assess the associations and mean differences between the explanatory variables and the outcomes. Statistical significance was set at a value of $p < 0.01$. The statistical analyses were performed using the software IBM SPSS Statistics for Windows, version 27.0 (IBM Corp., Armonk, N.Y., USA).

RESULTS

The characteristics of the participants and their views about involvement in decision-making on health data sharing, access, use, and reuse are presented in **Tables 1** and **2**, respectively. Most participants attained 12 or less years of education (75.6%) and were not involved with patient's organizations (94.9%). Almost 80% of the carers were female, while over 53% of the patients were male. Carers were older (>30 years) than patients (87.2 vs. 15.1%) and more frequently married or living with a partner (78.0 vs. 9.5%). More than half of carers perceived their income as insufficient (56.2%), while 64.6% of patients considered it comfortable/enough to make ends meet. About three quarters of the carers and two thirds of the patients perceived trust in research host institutions and trust in research teams as very important issues when making decisions about sharing data. Participants presented low levels of trust in national institutions (Median [P25–P75] 3.5 [1.8–5.2]), increasing slightly for trust in international institutions (Median [P25–P75] 5.0 [2.5–7.0]) and interpersonal trust (Median [P25–P75] 4.7 [3.0–6.7]).

Most participants considered it important or very important to be involved in decisions concerned with health data sharing (85.1%), access (87.1%), use (85%) and reuse (79.9%). This trend was observed among both patients and carers (**Table 2**).

Carers and older participants stated more frequently the importance of being involved in decision-making regarding data sharing and data access (**Table 3**). More educated participants revealed a statistically significant tendency to attribute more importance to participation in decisions about data use, while participants with the lowest levels of trust in national and international institutions (Median [P25–P75]: 2.2[0.8–4.0]) and 3.0[1.0–5.0], respectively) were less likely to value such type of involvement. Participants who considered trust in research host institutions as very important rated higher the importance of being involved in decisions about data sharing, data access, data use, and data reuse. A similar position was primarily expressed by participants who valued trust in research teams for data sharing, data access, and data use.

DISCUSSION

The majority of people affected by rare diseases who were surveyed placed a high value on opportunities for involvement

TABLE 1 | Characterization of the participants, stratified by people with rare diseases and their informal carers.

Participants	**Total (*N* = 637)**	**Patients (*n* = 159)**	**Carers (*n* = 478)**
Sex, *n* (%)			
Female	453 (71.1)	75 (47.2)	378 (79.1)
Male	184 (28.9)	84 (52.8)	100 (20.9)
Age (years), *n* (%)			
<18	92 (14.6)	92 (57.9)	-
18–30	103 (16.4)	43 (27.0)	60 (12.8)
>30	434 (69.0)	24 (15.1)	410 (87.2)
Educational level (years), *n* (%)			
≤12	476 (75.6)	151 (95.6)	325 (68.9)
>12	154 (24.4)	7 (4.4)	147 (31.1)
Marital status, *n* (%)			
Married/living with partner	384 (60.9)	15 (9.5)	369 (78.0)
Other	247 (39.1)	143 (90.5)	104 (22.0)
Occupation, *n* (%)			
Upper white-collar	148 (24.6)	5 (3.2)	143 (32.3)
Lower white-collar	116 (19.3)	8 (5.1)	108 (24.4)
Blue-collar	87 (14.5)	9 (5.7)	78 (17.6)
Other	250 (41.6)	136 (86.1)	114 (25.7)
Perceived income adequacy, *n* (%)			
Insufficient/Caution with expenses	315 (51.3)	51 (35.4)	264 (56.2)
Enough to make ends meet/comfortable	299 (48.7)	93 (64.6)	206 (43.8)
Involvement in patient organizations, *n* (%)			
No	598 (94.9)	154 (98.1)	444 (93.9)
Yes	32 (5.1)	3 (1.9)	29 (6.1)
Trust in research host institution, *n* (%)			
Very important	461 (73.6)	107 (67.7)	354 (75.6)
Other	165 (26.4)	51 (32.3)	114 (24.4)
Trust in research team, *n* (%)			
Very important	448 (71.5)	103 (65.6)	345 (73.4)
Other	179 (28.5)	54 (34.4)	125 (26.6)
Trust in national institutions, Md (P25-P75)	3.5 (1.8–5.2)	4.5 (2.0–6.0)	3.4 (1.8–5.0)
Trust in international institutions, Md (P25-P75)	5.0 (2.5–7.0)	6.5 (3.0–8.0)	5.0 (2.0–7.0)
Interpersonal trust, Md (P25-P75)	4.7 (3.0–6.7)	4.7 (2.4–6.7)	4.7 (3.0–6.4)

In each variable, the total may not add 637 participants, 159 patients or 478 carers due to missing values. The proportions may not add 100 due to rounding.

in decisions about health data sharing, access, use and reuse (ranging between 80–87%). These views differ from those of other publics such as people with diabetes among whom less than 50% considered important to decide what type of data can be shared and with whom (33). However, they are echoed by rare diseases communities across Europe who expressed a strong desire in keeping control over their shared data throughout the data processing cycle (80%) (34). Difficulties in obtaining diagnoses, the absence of cures, and oftentimes of treatment, inspire a firm commitment on the part of rare disease patients and their carers toward advancing research, which is further strengthened by a perceived need to optimize the use of scarce biospecimens and research resources (34, 36, 40, 42, 47). These challenges may explain rare diseases participants' eagerness to engage in decisions about how their data should be governed. Playing an active role in deciding what data can be shared, with whom and for which purposes can help to not only reorient governance frameworks to become more commensurate with their values and preferences, but also directly impact their lives, and those of future generations, by driving research and care to meet their specific needs (34, 40).

Our findings also show a strong positive association between the value attributed to trust in science and the value attributed to public involvement in data governance. Participants who ascribed a high degree of importance to trust in research institutions when choosing whether to share their data were significantly more likely to value involvement across the full spectrum of aspects related with individual-level data governance (data sharing, access, use and reuse). A similar pattern was found for trust in researchers and involvement in decisions about data sharing, access and use. These findings resonate with Aitken et al. (7) argument that public involvement is one out

TABLE 2 | Participants' views about involvement in decision-making regarding health data sharing, access, use and reuse.

Involvement in decision-making regarding	Total (*N* = 637) *n* (%)	Patients (*n* = 159) *n* (%)	Carers (*n* = 478) *n* (%)
Data sharing			
Not important	14 (2.2)	6 (3.8)	8 (1.7)
Slightly important	14 (2.2)	6 (3.8)	8 (1.7)
Moderately important	67 (10.5)	28 (17.6)	39 (8.2)
Important	286 (44.9)	62 (39.0)	224 (46.9)
Very important	256 (40.2)	57 (35.8)	199 (41.6)
Data access			
Not important	13 (2.0)	7 (4.4)	6 (1.3)
Slightly important	15 (2.4)	7 (4.4)	8 (1.7)
Moderately important	54 (8.5)	26 (16.4)	28 (5.9)
Important	265 (41.6)	55 (34.6)	210 (43.9)
Very important	290 (45.5)	64 (40.3)	226 (47.3)
Data use			
Not important	7 (1.1)	2 (1.3)	5 (1.0)
Slightly important	17 (2.7)	7 (4.4)	10 (2.1)
Moderately important	71 (11.1)	26 (16.4)	45 (9.4)
Important	271 (42.5)	62 (39.0)	209 (43.7)
Very important	271 (42.5)	62 (39.0)	209 (43.7)
Data reuse			
Not important	13 (2.0)	6 (3.8)	7 (1.5)
Slightly important	19 (3.0)	6 (3.8)	13 (2.7)
Moderately important	96 (15.1)	31 (19.5)	65 (13.6)
Important	255 (40.0)	67 (42.1)	188 (39.3)
Very important	254 (39.9)	49 (30.8)	205 (42.9)

The proportions may not add 100 due to rounding.

a set of institutional arrangements that are central in ensuring the trustworthiness of research, which, in turn, is required to foster public trust in science. Scientific initiatives guided by participatory ideals privilege reciprocity and acknowledge participants' expectations, needs and agency, not least by facilitating a people-centered approach to consent that enables data donors to choose from blanket, broad or dynamic consent models (i.e. meta consent) (32). Dynamic consent approaches afford participants an ongoing opportunity to decide the conditions in which their data can be shared, accessed, used and reused, over time and across a range of research initiatives and settings. These approaches also contribute to the establishment of ongoing communication with, and feedback from, researchers that can give rise to more substantive participatory initiatives (e.g., public deliberation exercises; public engagement in data access committees) (29, 48–51). Such participatory initiatives carry potential to increase research transparency and to promote accountability by enabling researchers and diverse publics to come together and build dialogic relationships that are essential for uncovering existing concerns and imbuing systems of data governance with public values and the mechanisms needed to ensure checks and balances, oversight and redress for misconduct (7, 52, 53). However, while public involvement can enhance research trustworthiness, (7, 54, 55), a minimum level of public trust in science has to be present for public involvement to unfold (49). Our study corroborates these findings by showing that rare diseases patients and their carers are significantly more likely to value involvement in health data governance when they hold trust in science in high regard.

Following a wider international trend (56–58), the Portuguese population has reported relatively high levels of trust in science (56). Yet, its level of trust in other institutions, including the European Parliament, national government, and the legal and health care systems, tends to be substantially lower (59–61). Trust in national institutions is related to citizens' perception of how effective institutions are in attending to their needs. For example, in Portugal, citizens who perceive government to be less effective and trustworthy are also less satisfied with the health system (59). Participants in our study also expressed low trust in national institutions and, to a lesser extent, in international institutions. Importantly, our study further shows that participants with the lowest levels of trust in national and international institutions attributed significantly less importance to getting involved in decision-making about how their data can be used. This trend may find explanation in the idea that public involvement is unlikely to inspire reciprocal partnerships and lead to transformative change in institutions perceived to be opaque, irresponsive, and unaccountable (62). Effecting change that is transformative requires the development of trusting relationships between institutional stakeholders and lay members of the public, the ability to accommodate and build on different types of knowledge and expertise and a thorough commitment to attending to the needs, and responding to the concerns, of the various parties involved (63). Where institutions fail to cultivate trust, incentives for involvement may wane or disappear altogether (49). Participatory exercises demand time, skills, and the confidence that the efforts made are grounded on transparent information and can foster the change needed to engender meaningful partnerships and ensure accountability (7). When these conditions are not met the drive for participation tends to plummet.

Carers and older participants in our study were more prone to value involvement in decisions about data sharing and data access. These findings align with those of an international survey carried out with people affected by rare diseases that found that participants identifying as patient representatives and older respondents were both more likely to perceive health-related information as sensitive and to want to retain control over who accesses their information, how and why (34).

Finally, our study shows that participants with lower levels of education attributed significantly less importance to involvement in decision-making about the purposes for which their data can be used. This finding may be pointing to an unequal distribution of the resources needed to make informed decisions about data use (e.g., health and digital literacy, access to digital devices, communication and negotiation skills). Big data, machine

TABLE 3 | Factors influencing participants' views about involvement in decision-making regarding health data sharing, access, use and reuse.

	Total	Data sharing		Data access		Data use		Data reuse	
		Important[a] ***n* (%)**	**Other[b]** ***n* (%)**	**Important[a]** ***n* (%)**	**Other[b]** ***n* (%)**	**Important[a]** ***n* (%)**	**Other[b]** ***n* (%)**	**Important[a]** ***n* (%)**	**Other[b]** ***n* (%)**
	637	542 (85.1)	95 (14.9)	555 (87.1)	82 (12.9)	542 (85.1)	95 (14.9)	509 (79.9)	128 (20.1)
Type of participant									
Patient	159	119 (74.8)*	40 (25.2)*	119 (74.8)*	40 (25.2)*	124 (78.0)	35 (22.0)	116 (73.0)	43 (27.0)
Carer	478	423 (88.5)*	55 (11.5)*	436 (91.2)*	42 (8.8)*	418 (87.4)	60 (2.6)	393 (82.2)	85 (17.8)
Sex									
Female	453	393 (86.8)	60 (13.2)	405 (89.4)	48 (10.6)	394 (87.0)	59 (13.0)	371 (81.9)	82 (18.1)
Male	184	149 (81.0)	35 (19.0)	150 (81.5)	34 (18.5)	148 (80.4)	36 (19.6)	138 (75.0)	46 (25.0)
Age (years)									
<18	92	67 (72.8)*	25 (27.2)*	66 (71.7)*	26 (28.3)*	69 (75.0)	23 (25.0)	69 (75.0)	23 (25.0)
18-30	103	86 (83.5)*	17 (16.5)*	93 (90.3)*	10 (9.7)*	88 (85.4)	15 (14.6)	85 (82.5)	18 (17.5)
>30	434	384 (88.5)*	50 (11.5)*	389 (89.6)*	45 (10.4)*	379 (87.3)	55 (12.7)	350 (80.6)	84 (19.4)
Educational level (years)									
≤12	476	397 (83.4)	79 (16.6)	404 (84.9)	72 (15.1)	389 (81.7)*	87 (18.3)*	372 (78.2)	104 (21.8)
>12	154	141 (91.6)	13 (8.4)	145 (94.2)	9 (5.8)	148 (96.1)*	6 (3.9)*	133 (86.4)	21 (13.6)
Marital status									
Married/living with partner	384	342 (89.1)	42 (10.9)	348 (90.6)	36 (9.4)	338 (88.0)	46 (12.0)	318 (82.8)	66 (17.2)
Others	247	197 (79.8)	50 (20.2)	202 (81.8)	45 (18.2)	200 (81.0)	47 (19.0)	188 (76.1)	59 (23.9)
Occupation									
Upper white-collar	148	137 (92.6)	11 (7.4)	137 (92.6)	11 (7.4)	138 (93.2)	10 (6.8)	119 (80.4)	29 (19.6)
Lower white-collar	116	99 (85.3)	17 (14.7)	102 (87.9)	14 (12.1)	98 (84.5)	18 (15.5)	91 (78.4)	25 (21.6)
Blue-collar	87	69 (79.3)	18 (20.7)	76 (87.4)	11 (12.6)	70 (80.5)	17 (19.5)	68 (78.2)	19 (21.8)
Other	250	204 (81.6)	46 (18.4)	205 (82.0)	45 (18.0)	205 (82.0)	45 (18.0)	199 (79.6)	51 (20.4)
Perceived income adequacy									
Insufficient/Caution with expenses	315	265 (84.1)	50 (15.9)	281 (89.2)	34 (10.8)	265 (84.1)	50 (15.9)	253 (80.3)	62 (19.7)
Enough to make ends meet/comfortable	299	259 (86.6)	40 (13.4)	253 (84.6)	46 (15.4)	258 (86.3)	41 (13.7)	238 (79.6)	61 (20.4)
Involvement with patients' organizations									
No	598	508 (84.9)	90 (15.1)	522 (87.3)	76 (12.7)	508 (84.9)	90 (15.1)	477 (79.8)	121 (20.2)
Yes	32	31 (96.9)	1 (3.1)	29 (90.6)	3 (9.4)	31 (96.9)	1 (3.1)	29 (90.6)	3 (9.4)
Trust in research host institution									
Very important	461	412 (89.4)*	49 (10.6)*	423 (91.8)*	38 (8.2)*	421 (91.3)*	40 (8.7)*	385 (83.5)*	76 (16.5)*
Other	165	121 (73.3)*	44 (26.7)*	121 (73.3)*	44 (26.7)*	111 (67.3)*	54 (32.7)*	115 (69.7)*	50 (30.3)*
Trust in research team									
Very important	448	401 (89.5)*	47 (10.5)*	410 (91.5)*	38 (8.5)*	405 (90.4)*	43 (9.6)*	369 (82.4)	79 (17.6)
Other	179	132 (73.7)*	47 (26.3)*	135 (75.4)*	44 (24.6)*	128 (71.5)*	51 (28.5)*	131 (73.2)	48 (26.8)
Trust in national institutions, Md (P25-P75)	3.5 (1.8–5.2)	3.8 (1.8–5.4)	2.4 (1.0–3.8)	3.8 (1.8–5.3)	2.6 (1.3–5.0)	3.8 (2.0–5.4)*	2.2 (0.8–4.0)*	3.6 (1.8–5.2)	3.2 (1.6–5.3)
Trust in international institutions, Md (P25–P75)	5.0 (2.5–7.0)	5.0 (2.5–7.0)	3.5 (1.0–5.5)	5.0 (2.5–7.0)	3.0 (1.0–7.0)	5.0 (2.5–7.0)*	3.0 (1.0–5.0)*	5.0 (2.5–7.0)	4.5 (2.0–7.0)
Interpersonal trust, Md (P25–P75)	4.7 (3.0–6.7)	4.7 (3.0–6.7)	3.7 (2.0–6.0)	4.7 (3.0–6.3)	5.0 (2.3–6.7)	4.7 (3.0–6.3)	3.7 (2.0–6.7)	4.7 (3.0–6.3)	4.8 (3.0-6.7)

*[a] Includes participants who answered "important" and "very important"; [b] includes participants who answered, "not important", "slightly important" and "moderately important"; *p < 0.001. In each variable, the total may not add 637 participants, 159 patients or 478 carers due to missing values. The proportions may not add 100 due to rounding.*

learning and artificial intelligence have contributed to expand the purposes of biomedical and healthcare research to a multitude of fast-evolving fields (64). Increasingly, research endeavors focus on issues that lay people may not be familiar with and feel wary to express opinions about (e.g., gene therapy) (65). Disregard for the needs of publics who are less equipped to assess the value and risks of cutting-edge research and care can contribute to reduce trust and avert their participation. Moreover, it can reinforce a long-lasting pattern of exclusion found across the European Region where minority and socioeconomically disadvantaged groups have been systematically under-represented in health research, as well as in the participatory spaces created to involve lay people in its design and implementation (17, 66, 67).

Assessing and attending to consent preferences and offering time and support to anyone expected to make informed decisions is essential (68, 69). However, with the exponential growth of data sources and data uses, informal support may not be sufficient to enable informed consent (44). As argued by Fiske et al. (65), it is necessary to make way for a new group of professionals—health information counselors—who can advise on the far-reaching implications of data decisions and assist in addressing arising ethical, legal and social challenges and dilemmas that often extend beyond the individual sphere (e.g., the right to choosing not to know and, thus, to decline the return of incidental research findings that may identify a predisposition for late-onset genetic diseases with implications for the offspring) (70). Health information counseling services may be especially relevant for decisions concerned with the use of one's data for purposes other than those for which it was originally collected. The reuse of health data can occur in contexts with norms and values different from those upheld in research and care settings and which are more often subject to "data trust deficits" (71). Commercial settings such as direct-to-consumer genetic testing companies are one such example where values such as transparency and reciprocity may be overridden by economic interest (e.g., patenting consumer data that was first shared under the pretense that it would be used to democratize genomics) [see (72)]. Elucidating on the ethical, legal and social implications of sharing data for research, care, commercial and other secondary purposes is of critical importance to reduce resource gaps, inform lay people's expectations, empower them to make informed decisions and promote the trustworthiness of data processing organizations.

STRENGTHS AND LIMITATIONS

This study offers three major contributions. First, it is one of a few studies to assess public views about involvement in all key dimensions of individual-level data governance and to enable the identification of differences in the importance attributed to participation in decision-making concerned with health data sharing, access, use, and reuse. Another major contribution relates to the examination of its association with various types of trust and sociodemographic variables. Finally, data collection was carried out over an extended recruitment period of 10 months and participants were consecutively invited to participate at two reference centers for rare diseases located in an academic hospital center that oversees patients from the entire Northern Health Region of Portugal. Nevertheless, recruitment in one region limits the generalizability of the results and thus inferences for the general rare diseases population should be performed with caution. Furthermore, the value attributed to opportunities for involvement in decisions about health data sharing, access, use and reuse may be overestimated in this particular setting, as the reference centers have a strong academic orientation and are involved with rare diseases European Reference Networks. Many of the patients and carers surveyed have been involved in data sharing for national and international research projects and are experienced in decision-making concerned with their health data. However, this specific context might entail power-asymmetric relationships which may influence research participation and the data collected (73). The recruitment of participants in non-academic and in private settings would enable an enriching comparison. Finally, further qualitative and quantitative research is warranted to uncover participants' motivations and expectations regarding involvement in individual-level data governance, as well as to provide an in-depth understanding on the factors that contribute to foster and sustain public trust in research carried out in health care institutions.

CONCLUSION

The high value attributed by participants to involvement in individual-level data governance stresses the need to rethink opportunities for public participation in health data decision-making. Broadening the consent options currently on offer to people affected by rare diseases to include mechanisms that allow them to choose between broad, blanket and dynamic consent models according to the type of data requested and the context in which that request is made deservers thorough consideration. Trust in science and other institutions played an important role in shaping our participants' views about involvement. Accordingly, the adoption of a meta consent approach (32) would likely need to be accompanied by the provision of transparent information about the implications of data sharing, assistance with obtaining the resources needed to make informed choices and the development of harm mitigation tools and redress.

DATA AVAILABILITY STATEMENT

The datasets generated and analyzed for this study are not publicly available due to a confidentiality agreement securing participants' privacy and anonymity, but they are available from the corresponding author upon reasonable request.

AUTHOR CONTRIBUTIONS

CF conceptualized the study. SS and CF designed the data analysis strategy. NN performed data analysis. NN and CF wrote the first draft of the manuscript. NN, SS, and CF contributed to the interpretation of data, critically reviewed previous versions of the manuscript, and approved the final version. All authors contributed to the article and approved the submitted version.

FUNDING

This work was funded by FEDER through the Operational Programme for Competitiveness and Internationalisation and national funding from the Foundation for Science and Technology—FCT (Portuguese Ministry of Science, Technology and Higher Education) (Ref. POCI-01–0145-FEDER-032194), under the project Public and patient involvement in health data governance: a people-centred approach to data protection in genetic diseases (Ref. FCT PTDC/SOC-SOC/32194/2017) and the Unidade de Investigação em Epidemiologia—Instituto de Saúde Pública da Universidade do Porto (EPIUnit) (Ref. UIDB/04750/2020), Laboratório para a Investigação Integrativa e Translacional em Saúde Populacional (ITR) (LA/P/0064/2020), the individual contract grant DL57/2016/CP1336/CT0001 (CF) and the individual contract grant IF/01674/2015 (SS).

ACKNOWLEDGMENTS

We are grateful to all participants who collaborated in this study, as well as to the health professionals of University Hospital Centre São João who collaborated in participants' recruitment. We are also thankful to the research team of project DATAGov, and in particular to Maria João Baptista, Elisa Leão Teles, Helena Machado, Mariana Amorim, and Tiago Maia. This manuscript was developed in the scope of a Master Thesis in Public Health led by NN.

REFERENCES

1. Rumsfeld JS, Joynt KE, Maddox TM. Big data analytics to improve cardiovascular care: promise and challenges. *Nat Rev Cardiol.* (2016) 13:350–9. doi: 10.1038/nrcardio.2016.42
2. Flores M, Glusman G, Brogaard K, Price ND, Hood L. P4 medicine: how systems medicine will transform the healthcare sector and society. *P Med.* (2013) 52:565–76. doi: 10.2217/pme.13.57
3. Benson M. Clinical implications of omics and systems medicine: focus on predictive and individualized treatment. *J Int Med.* (2016) 279:229–40. doi: 10.1111/joim.12412
4. Prainsack B. The political economy of digital data: introduction to the special issue. *Pol Stud.* (2020) 41:439–46. doi: 10.1080/01442872.2020.1723519
5. Pollock AM, Roderick P. Trust in the time of markets: protecting patient information. *Lancet.* (2014) 383:1523–4. doi: 10.1016/S0140-6736(14)60727-3
6. Wadmann S, Hoeyer K. Dangers of the digital fit: Rethinking seamlessness and social sustainability in data-intensive healthcare. *Big Data Soc.* (2018) 5:1–13. doi: 10.1177/2053951717752964
7. Aitken M, Cunningham-Burley S, Pagliari C. Moving from trust to trustworthiness: Experiences of public engagement in the Scottish Health Informatics Program. *Sci Pub Pol.* (2016) 43:713–23. doi: 10.1093/scipol/scv075
8. Trinidad SB, Fullerton SM, Bares JM, Jarvik GP, Larson EB, Burke W. Genomic research, and wide data sharing: Views of prospective participants. *Genet Med.* (2010) 12:486–95. doi: 10.1097/GIM.0b013e3181e38f9e
9. Van Staa TP, Goldacre B, Buchan I, Smeeth L. Big health data: the need to earn public trust. *BMJ.* (2016) 354:i3636. doi: 10.1136/bmj.i3636
10. Williams GA, Fahy N. Building and maintaining public trust to support the secondary use of personal health data. World Health Organization. Regional Office for Europe. *Eurohealth*. (2019) 25:7–10. Available online at: https://apps.who.int/iris/handle/10665/332521 (accessed December 10, 2021).
11. Oderkirk J, Ronchi E. Governing data for better health and healthcare. OECD: OECD Observer (2018). p. 1–4. Available online at: http://oe.cd/1JT (accessed December 10, 2021).
12. MORI I. The One-Way Mirror: Public attitudes to commercial access to health data. Wellcome Trust (2016). Available online at: https://www.ipsos-mori.com/researchpublications/publications/1803/Commercial-access-to-health-data.aspx (accessed on December 10, 2021)
13. Milne R, Morley KI, Almarri MA, Anwer S, Atutornu J, Baranova EE, et al. Demonstrating trustworthiness when collecting and sharing genomic data: public views across 22 countries. *Gen Med.* (2021) 13:1–12. doi: 10.1186/s13073-021-00903-0
14. OECD. Recommendation of the Council on Health Data Governance. (2016). Available online at: https://www.oecd.org/health/health-systems/Recommendation-of-OECD-Council-on-Health-Data-Governance-Booklet.pdf (accessed December 10, 2021).
15. OECD. Health data governance: privacy, monitoring and research – policy brief. (2015). Available online at: https://www.oecd.org/health/health-systems/Health-Data-Governance-Policy-Brief.pdf (accessed December 10, 2021).
16. Kaye J, Terry SF, Juengst E, Coy S, Harris JR, Chalmers D, et al. Including all voices in international data sharing governance. *Hum Gen.* (2018) (1):13–9. doi: 10.1186/s40246-018-0143-9
17. Fiske A, Prainsack B, Buyx A. Meeting the needs of underserved populations: setting the agenda for more inclusive citizen science of medicine. *J Med Ethics.* (2019) 45:617–22. doi: 10.1136/medethics-2018-105253
18. Aitken M, Tully MP, Porteous C, Denegri S, Cunningham-Burley S, Banner N, et al. Consensus statement on public involvement and engagement with data intensive health research. *Int J Pop Data Sci.* (2019) 4:1–6. doi: 10.23889/ijpds.v3i4.837
19. Kariotis T, Ball MP, Greshake Tzovaras B, Dennis S, Sahama T, Johnston C, et al. Emerging health data platforms: From individual control to collective data governance. *Data Pol.* (2020) 2:e13–25. doi: 10.1017/dap.2020.14
20. Buyx A, Savio L, del Prainsack B, Vo H. Every participant is a PI Citizen science and participatory governance in population studies. *Int J Epid.* (2017) 46:377–84. doi: 10.1093/ije/dyw204
21. Vayena E, Blasimme A. Biomedical big data: new models of control over access, use and governance. *J Bioethic Inquiry.* (2017) 14:501–13. doi: 10.1007/s11673-017-9809-6
22. Hafen E, Kossmann D, Brand A. Health data cooperatives - Citizen empowerment. *Methods Inf Med.* (2014) 53:82–6. doi: 10.3414/ME13-02-0051
23. Riso B, Tupasela A, Vears DF, Felzmann H, Cockbain J, Loi M, et al. Ethical sharing of health data in online platforms – which values should be considered? *Life Sci Soc Policy.* (2017) 13:12–39. doi: 10.1186/s40504-017-0060-z
24. Beier K, Schweda M, Schicktanz S. Taking patient involvement seriously: A critical ethical analysis of participatory approaches in data-intensive medical research. *BMC Med Inform Decis Mak.* (2019) 19:90–100. doi: 10.1186/s12911-019-0799-7
25. Kaye J, Curren L, Anderson N, Edwards K, Fullerton SM, Kanellopoulou N, et al. From patients to partners: participant-centric initiatives in biomedical research. *Nat Rev Genet.* (2012) 13:371–6. doi: 10.1038/nrg3218
26. Goisauf M, Durnová AP. From engaging publics to engaging knowledges: Enacting "appropriateness" in the Austrian biobank infrastructure. *Public Underst Sci.* (2019) 28:275–89. doi: 10.1177/0963662518806451
27. Wendler D. Broad versus blanket consent for research with human biological samples. *Hastings Cent Rep.* (2013) 43:3–4. doi: 10.1002/hast.200
28. Budin-Ljøsne I, Teare HJA, Kaye J, Beck S, Bentzen HB, Caenazzo L, et al. Dynamic Consent: A potential solution to some of the challenges of modern biomedical research. *BMC Med Ethics.* (2017) 18:1. doi: 10.1186/s12910-016-0162-9
29. Kaye J, Whitley EA, Lund D, Morrison M, Teare H, Melham K. Dynamic consent: A patient interface for twenty-first century research networks. *Eur J Hum Genet.* (2015) 23:141–6. doi: 10.1038/ejhg.2014.71
30. Teare HJA, Morrison M, Whitley EA, Kaye J. Towards 'Engagement 20': insights from a study of dynamic consent with biobank participants. *Digital Health.* (2015) 1:1–13. doi: 10.1177/2055207615605644
31. Thiel DB, Platt J, Platt T, King SB, Fisher N, Shelton R, et al. Testing an online, dynamic consent portal for large population biobank research. *Pub Health Gen.* (2015) 18:26–39. doi: 10.1159/000366128
32. Ploug T, Holm S. Meta consent: A flexible and autonomous way of obtaining informed consent for secondary research. *BMJ.* (2015) 7:h2146. doi: 10.1136/bmj.h2146

33. Shah N, Coathup V, Teare H, Forgie I, Nicola G, Tue G, et al. Motivations for data sharing — views of research participants from four European countries: a DIRECT study. *Eur J Hum Genet.* (2019) 27:721–9. doi: 10.1038/s41431-019-0344-2
34. Courbier S, Dimond R, Bros-facer V. Share and protect our health data: an evidence-based approach to rare disease patients' perspectives on data sharing and data protection - quantitative survey and recommendations. *Orphanet J Rare Dis.* (2019) 14:175–90. doi: 10.1186/s13023-019-1123-4
35. Shah N, Coathup V, Teare H, Forgie I, Giordano GN, Hansen TH, et al. Sharing data for future research — engaging participants' views about data governance beyond the original project: a DIRECT Study. *Genet Med.* (2019) 21:1131–8. doi: 10.1038/s41436-018-0299-7
36. McCormack P, Kole A, Gainotti S, Mascalzoni D, Molster C, Lochmüller H, et al. "You should at least ask". The expectations, hopes and fears of rare disease patients on large-scale data and biomaterial sharing for genomics research. *Eur J Hum Genet.* (2016) 24:1403–8. doi: 10.1038/ejhg.2016.30
37. Riordan F, Papoutsi C, Reed JE, Marston C, Bell D, Majeed A. Patient and public attitudes towards informed consent models and levels of awareness of Electronic Health Records in the UK. *Int J Med Inf.* (2015) 84:237–47. doi: 10.1016/j.ijmedinf.2015.01.008
38. Ludman EJ, Fullerton SM, Spangler L, Trinidad SB, Fujii MM, Jarvik GP, et al. Glad you asked: participants' opinions of re-consent for DBGAP data submission. *J Emp Res Hum Ethics.* (2010) 5:9–16. doi: 10.1525/jer.2010.5.3.9
39. Bell EA, Ohno-Machado L, Grando MA. Sharing my health data: a Survey of data sharing preferences of healthy individuals. *AMIA Annu Symp Proc.* (2014) 14:1699–108.
40. Darquy S, Moutel G, Lapointe AS, D'Audiffret D, Champagnat J, Guerroui S, et al. Patient/family views on data sharing in rare diseases: study in the European LeukoTreat project. *Eur J Hum Genet.* (2015) 24:338–43. doi: 10.1038/ejhg.2015.115
41. Boulanger V, Schlemmer M, Rossov S, Seebald A, Gavin P. Establishing patient registries for rare diseases: Rationale and challenges. *Pharmaceut Med.* (2020) 34:185–90. doi: 10.1007/s40290-020-00332-1
42. De Freitas C, dos Reis V, Silva S, Videira PA, Morava E, Jaeken J. Public and patient involvement in needs assessment and social innovation: A people-centred approach to care and research for congenital disorders of glycosylation. *BMC Health Serv Res.* (2017) 17:682–93. doi: 10.1186/s12913-017-2625-1
43. Thorogood A. International data sharing and rare disease: the importance of ethics and patient involvement. In: *Rare Diseases.* (2020); p- 1-17. IntechOpen, London. doi: 10.5772/intechopen.91237
44. De Freitas C, Amorim M, MacHado H, Leão Teles E, Baptista MJ, Renedo A, et al. Public and patient involvement in health data governance (DATAGov): protocol of a people-centred, mixed-methods study on data use and sharing for rare diseases care and research. *BMJ Open.* (2021) 11:e044289. doi: 10.1136/bmjopen-2020-044289
45. Instituto Nacional Estatística - Classificação Portuguesa das Profissões. (2010). Available online at: https://www.ine.pt/xurl/pub/107961853 (accessed December 11, 2021).
46. Zmerli S, Newton K. Social trust and attitudes toward democracy. *Public Opin Q.* (2008) 72:706–24. doi: 10.1093/poq/nfn054
47. Dwyer AA, Quinton R, Morin D, Pitteloud N. Identifying the unmet health needs of patients with congenital hypogonadotropic hypogonadism using a web-based needs assessment: Implications for online interventions and peer-to-peer support. *Orphanet J Rare Dis.* (2014) 9:83–94. doi: 10.1186/1750-1172-9-83
48. Murtagh MJ, Blell MT, Butters OW, Cowley L, Dove ES, Goodman A, et al. Better governance, better access: Practising responsible data sharing in the METADAC governance infrastructure. *Hum Genomics.* (2018) 12:24. doi: 10.1186/s40246-018-0154-6
49. Kraft SA, Cho MK, Gillespie K, Halley M, Varsava N, Ormond KE, et al. Beyond consent: Building trusting relationships with diverse populations in precision medicine research. *Am J Bioeth.* (2018) 8:3–20. doi: 10.1080/1526516120181431322
50. Spencer K, Sanders C, Whitley Edgar A, Lund D, Kaye J, William et al. Patient perspectives on sharing anonymized personal health data using a digital system for dynamic consent and research feedback: A qualitative study. *JMIR.* (2016) 18:e66. doi: 10.2196/jmir.5011
51. Burgess MM. From "trust us" to participatory governance: deliberative publics and science policy. *Pub Understanding of Sci.* (2014) 23:48–52. doi: 10.1177/0963662512472160
52. O'Doherty KC, Burgess MM, Edwards K, Gallagher RP, Hawkins AK, Kaye J, et al. From consent to institutions: Designing adaptive governance for genomic biobanks. *Soc Sci Med.* (2011) 73:367–74. doi: 10.1016/j.socscimed.2011.05.046
53. Ursin L, Ytterhus B, Christensen E, Skolbekken JA. If you give them your little finger, they'll tear off your entire arm: losing trust in biobank research. *Medicine, Health Care Phil.* (2020) 23:565–76. doi: 10.1007/s11019-020-09969-w
54. Kerasidou A. Trust me, I'm a researcher!: The role of trust in biomedical research. *Med Health Care and Philos.* (2016) 20:43–50. doi: 10.1007/s11019-016-9721-6
55. Moodley K, Singh S. "It's all about trust": reflections of researchers on the complexity and controversy surrounding biobanking in South Africa. *BMC Med Ethics.* (2016) 17:57–66. doi: 10.1186/s12910-016-0140-2
56. Gallup. How does the world feel about science and health? Wellcome Global Monitor (2019). Available online at: https://cms.wellcome.org/sites/default/files/wellcome-global-monitor-2018.pdf (accessed January 10, 2022).
57. Funk C, Hefferon M, Kennedy B, Johnson C. Trust and mistrust in Americans' views of scientific experts. Pew Res Center (2019). Available online at: https://www.pewresearch.org/science/2019/08/02/trust-and-mistrust-in-americans-views-of-scientific-experts/ (accessed January 10, 2022).
58. Castell S, Charlton A, Clemence M, Pettigrew N, Pope S, Quigley A, et al. *Public Attitudes to science 2014.* Ipsos MORI Social Research Institute, London. (2014).
59. Asensio M. The political legitimacy of the healthcare system in Portugal: insights from the European Social Survey. *Healthcare.* (2021) 9:202–23. doi: 10.3390/healthcare9020202
60. Dotti Sani GM, Magistro B. Increasingly unequal? The economic crisis, social inequalities, and trust in the European Parliament in 20 European countries. *Euro J Pol Res.* (2016) 55:246–64. doi: 10.1111/1475-6765.12126
61. Torcal M. The decline of political trust in Spain and Portugal: economic performance or political responsiveness? *Am Behav Sci.* (2014) 58:1542–67. doi: 10.1177/0002764214534662
62. Gaventa J. 'Towards participatory governance: assessing the transformative possibilities'. In: Hickey S, Mohan G (eds) *Towards participatory governance: assessing the transformative possibilities' Participation: From tyranny to transformation.* Zed Books Ltd, New York (2004) p. 25–41.
63. De Freitas C, Martin G. Inclusive public participation in health: Policy, practice, and theoretical contributions to promote the involvement of marginalised groups in healthcare. *Soc Sci Med.* (2015) 135:31–9. doi: 10.1016/j.socscimed.2015.04.019
64. Obermeyer Z, Emanuel EJ. Predicting the Future — Big data, machine learning, and clinical medicine. *The N Eng J Med.* (2016) 375:1216–9. doi: 10.1056/NEJMp1606181
65. Fiske A, Buyx A, Prainsack B. Health Information Counselors: a new profession for the age of big data. *Acad Med.* (2019) 94:37–41. doi: 10.1097/ACM.0000000000002395
66. MacFarlane A, Ogoro M, de Freitas C, Niranjan V, Severoni S, Waagensen E. Migrants' involvement in health policy, service development and research in the WHO European Region: a narrative review of policy and practice. *Trop Med & Inter Health.* (2021) 26:1164–76. doi: 10.1111/tmi.13643
67. World Health Organisation. More than numbers — evidence for all. European Health report. (2018). Available online at: https://www.euro.who.int/__data/assets/pdf_file/0003/380478/HEALTH_REPORT_HIGHLIGHTS_2018_EN.PDF (accessed December 22, 2021).
68. Baía I, de Freitas C, Samorinha C, Provoost V, Silva S. Dual consent? Donors' and recipients' views about involvement in decision-making on the use of embryos created by gamete donation in research. *BMC Med Ethics.* (2019) 20:90–6. doi: 10.1186/s12910-019-0430-6

69. Gainotti S, Turner C, Woods S, Kole A, McCormack P, Lochmüller H, et al. Improving the informed consent process in international collaborative rare disease research: effective consent for effective research. *Eur J Hum Genet.* (2016) 24:1248–54. doi: 10.1038/ejhg.2016.2
70. Kleiderman E, Knoppers BM, Fernandez C, Boycott KM, Ouellette G, Wong-Rieger D, et al. Returning incidental findings from genetic research to children: views of parents of children affected by rare diseases. *J Med Ethics.* (2014) 40:691–6. doi: 10.1136/medethics-2013-101648
71. Shah N, Johansson JV, Haraldsdóttir E, Bentzen HB, Coy S, Mascalzoni D, et al. Governing health data across changing contexts: A focus group study of citizen's views in England, Iceland, and Sweden. *Int J Med Inf.* (2021) 156:104623. doi: 10.1016/j.ijmedinf.2021.104623
72. Sterckx S, Cockbain J, Howard H, Huys I, Borry P. "Trust is not something you can reclaim easily": patenting in the field of direct-to-consumer genetic testing. *Genet Med.* (2013) 15:382–7. doi: 10.1038/gim.2012.143
73. Hoeyer K. 'Science is really needed—that's all I know": informed consent and the non-verbal practices of collecting blood for genetic research in northern Sweden. *Genet Soc.* (2010) 22:229–244. doi: 10.1080/1463677032000147199

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: All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

Copyright © 2022 Nwebonyi, Silva and de Freitas. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

frontiers | Frontiers in Public Health

ORIGINAL RESEARCH
published: 19 May 2022
doi: 10.3389/fpubh.2022.832090

Association Between Residence Status Regularization and Access to Healthcare for Undocumented Migrants in Switzerland: A Panel Study

Julien Fakhoury[1,2]*, **Claudine Burton-Jeangros**[1,2], **Liala Consoli**[1,2], **Aline Duvoisin**[1,3] and **Yves Jackson**[4]

[1] Swiss National Center of Competence in Research (NCCR) "LIVES – Overcoming Vulnerability: Life Course Perspectives", University of Geneva, Geneva, Switzerland, [2] Institute of Sociological Research, University of Geneva, Geneva, Switzerland, [3] Center for the Interdisciplinary Study of Gerontology and Vulnerability, University of Geneva, Geneva, Switzerland, [4] Geneva University Hospital and University of Geneva, Geneva, Switzerland

Background: Switzerland has a universal healthcare system. Yet, undocumented migrants face barriers at different levels that hinder their access to healthcare services. The aim of this study is to assess whether undocumented migrants' healthcare utilization improves with residence status regularization.

Methods: We used two-wave panel data from the Parchemins study, a study exploring the impact of regularization on undocumented migrants' health in Geneva, Switzerland. First wave data were collected between 2017 and 2018, second wave data between 2019 and 2020. At baseline, the sample consisted of 309 undocumented migrants, recruited after the implementation of a temporary regularization policy in Geneva. We distributed them into two groups according to their residence status 12 months before the second data collection [regularized vs. undocumented (controls)]. Using as dependent variable the number of medical consultations within two distinct 12-months periods (the first before regularization, the second after regularization), we conducted multivariable regression analyses applying hurdle specification to identify factors enhancing healthcare utilization. Then, we estimated first-difference panel models to assess change in healthcare utilization along regularization. Models were adjusted for demographic, economic and health-related factors.

Results: Of the 309 participants, 68 (22%) were regularized. For the 12 months before regularization, these migrants did not significantly differ in their healthcare utilization from the controls. At this stage, factors increasing the odds of having consulted at least once included being a female (aOR: 2.70; 95% CI: 1.37–5.30) and having access to a general practitioner (aOR: 3.15; 95% CI: 1.62–6.13). The factors associated with the number of consultations apart from underlying health conditions were the equivalent disposable income (aIRR per additional CHF 100.-: 0.98; 95% CI: 0.97–1.00) and having access to a general practitioner (aIRR: 1.45; 95% CI: 1.09–1.92). For the 12 months after regularization, being regularized was not associated with higher odds of having consulted

OPEN ACCESS

Edited by:
Angelo D'Errico,
Azienda Sanitaria Locale TO3, Italy

Reviewed by:
Marte Karoline Råberg Kjøllesdal,
Norwegian Institute of Public Health (NIPH), Norway
Ankita Siddhanta,
Independent Researcher, Houston, United States
Silvano Gabriele Cella,
University of Milan, Italy

***Correspondence:**
Julien Fakhoury
julien.fakhoury@unige.ch

Specialty section:
This article was submitted to Life-Course Epidemiology and Social Inequalities in Health, a section of the journal Frontiers in Public Health

Received: 09 December 2021
Accepted: 11 April 2022
Published: 19 May 2022

Citation:
Fakhoury J, Burton-Jeangros C, Consoli L, Duvoisin A and Jackson Y (2022) Association Between Residence Status Regularization and Access to Healthcare for Undocumented Migrants in Switzerland: A Panel Study. Front. Public Health 10:832090. doi: 10.3389/fpubh.2022.832090

at least once. However, among participants who consulted at least once, regularized ones reported higher counts of medical consultations than controls (3.7 vs. 2.6, $p = 0.02$), suggesting a positive impact of regularization. Results from the first-difference panel models confirmed that residence status regularization might have driven migrants' healthcare utilization (aβ: 0.90; 95% CI: 0.31-1.77).

Conclusions: This study supports the hypothesis that residence status regularization is associated with improved healthcare utilization among undocumented migrants. Future research is needed to understand the mechanisms through which regularization improves undocumented migrants' use of healthcare services.

Keywords: undocumented migrants, healthcare utilization, access to healthcare, policy, residence status regularization

BACKGROUND

Over the past few years,the International Community reaffirmed on multiple occasions its commitment toward universal health coverage (UHC), defined by the World Health Organization as ensuring the right of everyone to access quality healthcare in times of need and at an affordable price (1–3). Notably, UHC was framed as one of the main millennium and sustainable development goals adopted by the United Nations (1–3). Yet, undocumented migrants, i.e. migrants settled in a host country without a valid residence authorization, often face legal restrictions that limit their access to healthcare. For instance, in Europe, only few countries provide undocumented migrants with an access to regular primary and secondary healthcare (4). Most restrict undocumented migrants' entitlement to emergency services, sometimes at a substantial cost that prevents them from seeking care (4).

These structural (legal, administrative) restrictions cumulate with barriers to healthcare at the individual level. The Behavioral model, a theoretical framework specifically developed to explain healthcare access and utilization, differentiates the predisposing from the enabling factors. The predisposing factors refer to sociodemographic characteristics such as age, gender, ethnicity, level of acculturation or immigration status. The enabling factors encompass the persons' resources to access healthcare and cover its costs (5, 6). In the case of undocumented migrants, factors linked to their lack of residence status such as fear of denunciation, social marginalization, stigmatization or limited financial resources have consistently been found to hinder their access to healthcare (5, 7–11). For instance, in Switzerland, Germany and Denmark, undocumented pregnant women tended to avoid or delay pre-natal consultations due to lack of knowledge about the healthcare system, socioeconomic hardships or fear of being identified as undocumented (12–14). Undocumented migrants in Denmark also reported poor language proficiency and lack of social relationships with Danish people as major barriers to healthcare (9). These barriers are not specific to the European context but also exist in other parts of the world, such as in South Asia. For example, a study in India found that undocumented Bangladeshi and Nepalese migrants did not frequently use the local healthcare systems due to social exclusion and fear of identification or discrimination (15). Overall, compared to regular migrants, studies conducted in several European countries showed that undocumented migrants were less likely to seek healthcare (8, 12, 16).

On the other hand, undocumented migrants cumulate risk factors for poor health that may increase their needs for care. They show a high prevalence of multiple chronic conditions, which has been found to be a key determinant of their healthcare demand (17). They frequently occupy physically demanding jobs and are exposed to abuses on the labor market and to workplace violations (18–21). They have consistently been shown to be at higher risk of infections such as sexually transmitted diseases (22–25). Their often limited life opportunities and social interactions increase their risk of suffering from psychological distress (26). Overall, studies conducted across Europe showed that undocumented migrants consistently reported poorer physical and mental health than regular migrants or native residents (27–29).

This discrepancy between undocumented migrants' health needs and their effective use of the healthcare system may contribute to the development of serious yet preventable health problems among this population. In turn, this could lead to higher medical costs borne by healthcare systems and society as a whole (30). Designing efficient policies to bridge this gap is therefore of crucial medical, public health and economic importance (31, 32). A study among undocumented women in Utah, United States (US), suggested that public policies aiming at favoring undocumented migrants' social integration increased their healthcare utilization (33). In California, the enrolment of undocumented children in health insurance plans improved their access to and their use of medical and dental care (34). In view of this evidence, one may expect inclusive policies such as residence status regularization to contribute to the alleviation of structural and individual barriers to healthcare for undocumented migrants, resulting in improved healthcare access and increased utilization. However, evidence regarding

---

Abbreviations: 95% CI, 95% confidence interval; aβ, adjusted beta coefficient; aIRR, adjusted incidence risk ratio; aOR, adjusted odds ratio; CHF, Swiss Franc; €, Euro; HUG, Geneva University Hospital; IRR, incidence risk ratio; OR, odds ratio; NGO's, non-governmental associations; SD, standard deviation; SHS, Swiss Health Survey; USD, Dollar US; UHC, Universal health coverage.

the impact of residence status change on undocumented migrants' healthcare utilization is scarce. In Europe, to the best of our knowledge, no study has addressed the association between residence status regularization and use of healthcare services. This article attempts to fill this gap using longitudinal data from the Parchemins Study, a study evaluating the impact of the residence status regularization on undocumented migrants' living conditions and health in Geneva, Switzerland. More specifically, this paper tests whether residence status regularization leads to increased healthcare utilization among undocumented migrants.

METHODS

Setting

According to the latest estimates, the Canton of Geneva (population 500,000 inhabitants), Switzerland, is home to 10,000–15,000 undocumented migrants (35). Most of these migrants are well-established workers who lack valid residence authorization (undocumented economic migrants). Failed asylum seekers account for a small share (36). The healthcare system in Switzerland is universal but requires the individual purchase of a mandatory private insurance. Against payment of a premium of CHF 375—(344 €/405 USD) per month on average, this insurance covers a wide range of preventive, curative as well as rehabilitation services, provided that the patient has first paid a fixed deductible ranging from CHF 300—(250 €/324 USD) to CHF 2,500—(2,300 €/2,698 USD) entirely out of pocket. According to the Swiss legislation, undocumented migrants are entitled to and obliged, like any other resident, to take out a private health insurance upon 3 months of residence in the country. However, compliance with this obligation is only verified for individuals with a valid residence authorization.

In the Canton of Geneva, only 13–16% undocumented migrants are effectively insured (16% in our sample) (26, 37). Barriers faced by undocumented migrants to the purchase of an insurance include fear of denunciation, limited awareness of their rights or insufficient economic resources (36). In order to broaden healthcare access and utilization for undocumented and uninsured population, the Geneva University Hospital (HUG) dedicated a health center that provides a comprehensive range of subsidized medical services. Free medical consultations are also organized in the community, where general practitioners serve voluntarily as family doctors for undocumented migrants.

In 2017–2018, the Canton of Geneva implemented a two-year pilot policy called "Operation Papyrus". Its aim was to grant undocumented economic migrants renewable residence authorizations upon the following strict requirements: (1) no previous application for asylum, (2) a continuous stay in Geneva for 10 years (5 years for parents of school-aged children), (3) financial independence, (4) basic French proficiency, and (5) absence of criminal record. Meeting these criteria, jointly agreed upon by the local authorities, trade unions and non-governmental associations, guaranteed migrants who applied for regularization to be granted a residence authorization. Policy implementation also involved trade unions and non-governmental associations (NGO's) with a mandate from the local authorities (1) to act as gatekeepers and (2) to assist eligible migrants throughout the regularization process.

After regularization, migrants had 3 months to enroll into a health insurance scheme. Once insured, they could no longer consult at the HUG dedicated unit for undocumented and uninsured population but could access to the whole range of services within the regular healthcare system. In this context, two hypotheses are tested here. On the one hand, regularization is expected to increase the uptake of the mandatory health insurance scheme, resulting in improved healthcare utilization among regularized migrants. On the other hand, regularized migrants could face difficulties in meeting the healthcare costs generated by the Swiss mandatory health insurance while also losing access to the HUG dedicated unit for uninsured population, which in turn could hamper their healthcare utilization (38).

Study Design

This is a longitudinal, observational study based on two-wave data collected within the larger framework of the Parchemins Study. The Parchemins Study protocol can be consulted elsewhere (38).

Participants

Participants were recruited in Geneva between October 2017 and December 2018. At baseline, the sampled population consisted of undocumented economic migrants who (1) were aged 18 or more, (2) were not nationals of a European Union or European Free-Trade Association member state, (3) had never been asylum seekers, and (4) had been residing continuously in Geneva for at least 3 years. It included migrants who had been regularized within 3 months prior to their participation, a timeframe deemed too short to allow significant shifts due to regularization in their living conditions.

Recruitment strategies were set up in order to ensure a convenience sample as diverse as possible, taking into account that undocumented economic migrants are hard-to-reach. The main strategy consisted of face-to-face recruitment at two different settings: (1) during sessions organized by mandated trade unions and NGO's to assist undocumented economic migrants with their application for regularization (85%) and (2) in the waiting room of the HUG dedicated unit for undocumented and uninsured population (15%). Secondary strategies included snowball sampling and advertising through social networks.

Before they participated in the first data collection, all participants were ensured confidentiality orally and in writing and were asked to fill in an informed consent form. Those who consented to participate in the second data collection were asked for their phone numbers and e-mail addresses, so that we could recontact them approximately 12 months later. We then prioritized phone contact for the follow-up strategy and resorted to e-mail messages in case of non-response (38).

Data Collection

Data were collected face-to-face by trained investigators, using a mobile tablet with a pre-loaded questionnaire (Computer-Assisted Personal Interviews). The questionnaire consisted of measurements of variables related to participants' (1) sociodemographic characteristics and residence status, (2) living conditions, (3) health and access to healthcare, (4) economic and financial situation, and (5) social relationships. It was specifically developed for the purpose of longitudinal data collection in the context of the Parchemins Study and was used for the first and second data collections, with slight adjustments before the latter. The use of the same questionnaire for both data collections allowed for comparisons over time of the same variables measured at different time points.

The questionnaire was translated into the four main languages spoken by undocumented economic migrants in Geneva (French, English, Portuguese and Spanish). It was completed at the University of Geneva or at a place chosen by the participants, in their preferred language. The participants entered their responses in the mobile tablet with the assistance of the investigators. First wave data were gathered between October 2017 and December 2018 and second wave data between March 2019 and February 2020. On average, the time elapsed between the first and second personal interviews was 15 months.

The Ethics Committee of the Geneva Canton, Switzerland, approved the study protocol (CCER 2017-00897).

Variables

Measure of Healthcare Utilization

Healthcare utilization was measured using a discrete variable, the self-reported number of consultations to a medical doctor in the previous 12 months, which is an indicator widely used in the literature (39–42). The number of consultations ranged from 0 to 10+ (10+ meaning "10 consultations or more"). Medical consultations encompassed visits to a general practitioner, specialist, psychiatrist or gynecologist, but excluded consultations with a dentist, which are not covered by health insurance in Switzerland, and visits to emergency rooms. Given that participants in the regularized group had obtained a residence permit at most for 3 months at the moment of the first data collection, we assumed that the number of consultations reported for the 12 months prior to the first personal interviews reflected participants' healthcare utilization while undocumented.

Residence Status

Residence status regularization was our main exposure of interest. We categorized participants into two groups, based on the evolution of their residence status between the two data collection periods (**Figure 1**). The regularized group encompassed migrants that had been regularized 12 months or more prior to their second wave personal interviews. This categorization allowed us to explore within-individual covariance over time between healthcare utilization and residence status regularization. Indeed, we assumed that over the 12 months prior to their second wave personal interviews, these regularized migrants had insurance and faced fewer constraints on seeking healthcare as compared to when they were undocumented. Alternatively, the control group included (1) migrants who remained undocumented at the time of the second data collection and (2) migrants who got a residence authorization <12 months prior to their second wave personal interviews. We merged this latter subgroup with the undocumented participants to limit temporality bias, since we could not determine whether the medical visits that they reported occurred prior to their regularization—that is, if they occurred despite facing barriers to healthcare related to their lack of legal status—or after they had been regularized.

Covariates

Covariates were selected following the Behavioral model (5) and taking into account the eligibility criteria for regularization in the context of the Operation Papyrus.

As predisposing covariates, we included age, sex, country of origin (Latin America as reference, Asia, Eastern Europe, Africa), the duration of stay in Geneva (in years) and the level of oral French proficiency (Good as reference, Fair, Poor). These two latter variables were specifically included to reduce the risk of confounding bias, since they could predict residence status regularization in the context of the Operation Papyrus.

We used the equivalent disposable income [per CHF 100—units (95€/100 USD)], which was also a regularization criterion, the transfer of remittances (Yes vs. No) as well as having access to a general practitioner (GP) (Yes vs. No) as measures of enabling factors. Sending remittances was included as a complementary measure of the financial resources. Specifically, since remittances are often budgeted for relatives living in the country of origin or abroad, sending remittances might reduce participants' financial resources available in the event of healthcare needs. Having access to a GP was measured asking participants if they had a doctor to whom they could go for most of their health problems, regardless of whether the doctor was employed at the HUG or operating elsewhere. Despite a hypothesized mediating effect, we did not include health insurance as an enabling covariate in our analyses due to collinearity issues.

Health needs factors were measured using (1) the presence of comorbidity, defined as the self-report of 3 or more somatic chronic conditions (43) and (2) the self-report of depression or anxiety. The chronic conditions used to define comorbidity were selected in accordance with the Swiss Health Survey (SHS) (see 2.5.4 Secondary data source for a description of SHS). They included: asthma, any chronic lung disease, any heart disease, hypertension, a stroke, chronic joint pain, chronic back pain, chronic neck pain, diabetes, cholesterol, osteoporosis, any allergy, any liver disease and any kidney disease (44).

Age, the enabling factors and the variables related to the health needs were measured at baseline and at follow-up. Sex, origin, the duration of stay in Geneva and the level of oral proficiency in French were only measured at baseline.

Secondary Data Source

To compare participants' healthcare utilization with the general population's use, a secondary random sample of 580 natives and legal residents in Geneva, comparable in terms of age range

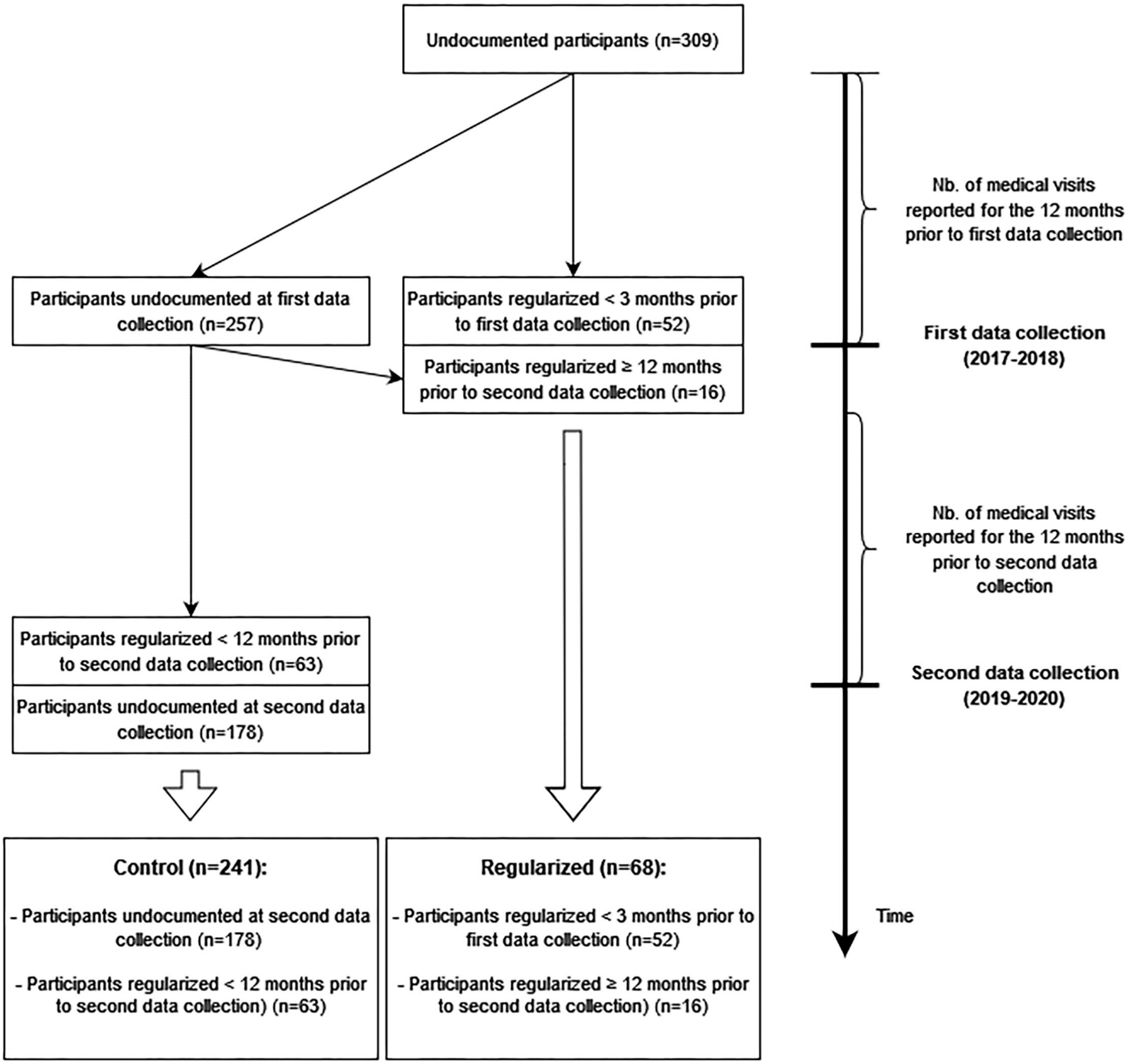


FIGURE 1 | Assignment of participants to residence status groups over time.

and occupational status, was drawn from the 2017 Swiss Health Survey (SHS). The SHS is a survey held every 5 years whose aim is to describe the health status as well as the healthcare consumption of the Swiss population (44).

Statistical Analyses

Categorical variables are presented as absolute numbers and relative percentages. Continuous variables are presented as means and standard deviations (SD). Cross-sectional comparisons across residence status groups were made using the Chi-square test or the Mann-Whitney's U-test, as appropriate. Changes over time in the outcome, the enabling and the health needs factors were measured using the McNemar's Chi-square test or the Wilcoxon's Signed-Rank Test, as appropriate. Statistical significance was set at 0.05.

First, we ran bivariate analyses to compare participants' healthcare utilization at baseline with that of the general population in Geneva. The comparison was not adjusted for predisposing and enabling covariates, since the 2017 SHS did not include fully comparable measures.

In a second step, we conducted univariate and multivariate count regression analyses to identify the factors associated with healthcare utilization among participants for the two 12-months periods studied; first, for the 12 months prior to the first data

TABLE 1 | Sociodemographic characteristics of the study participants, stratified by residence status ($N = 309$).

	Total ($N = 309$)	Control group ($N = 241$)	Regularized group ($N = 68$)	*p*-value
Female	225 (72.8%)	171 (71%)	54 (79.4%)	0.166
Age[a]	43.9 (10.3)	43.5 (10.5)	45.7 (9.1)	0.086
Origin				0.048
Latin America	200 (64.7%)	146 (60%)	54 (79.4%)	
Africa	17 (5.50%)	15 (6.2%)	2 (2.9%)	
East Asia	68 (22%)	59 (24.5%)	9 (13.2%)	
Eastern Europe	24 (7.8%)	21 (8.7%)	3 (4.4%)	
Duration in Geneva[a]	11.9 (4.8)	11.3 (5%)	13.7 (3.8)	<0.001
Oral French proficiency				0.005
Good	133 (43%)	97 (40.2%)	36 (52.9%)	
Fair	116 (37.5%)	88 (36.5%)	28 (41.2%)	
Poor	60 (19.5%)	56 (23.2%)	4 (5.9%)	

[a] *Presented as mean (SD).*

collection and second, for the 12 months before the second data collection. For the period prior to the first data collection, we were particularly interested in determining whether regularized migrants already differed from the controls in terms of medical visits. For the 12 months before the second data collection, we specifically looked for an association between regularization and healthcare utilization.

We used hurdle models to account for zero-inflation and over-dispersion in the outcome. Hurdle models also allowed emphasizing two distinct processes underlying healthcare utilization. The first process distinguished users from non-users of healthcare services, i.e., modeled the odds of reporting at least one medical visit, using logit regressions (the hurdle parts). Results of the hurdle parts are presented as odds ratio (OR) and 95% confidence intervals (95% CI) for univariate regressions and as adjusted odds ratio (aOR) and 95% CI for multivariate regressions. The second process assessed the factors associated with higher counts of medical consultations among healthcare users using truncated-at-zero negative binomial regressions (the truncated parts). Results of the truncated parts are presented as incidence risk ratios (IRR) and 95% CI for univariate regressions and as adjusted incidence risk ratios (aIRR) and 95% CI for multivariate regressions.

In a third step, we estimated panel models using the first-difference estimator to assess change in healthcare utilization associated with regularization. The first-difference estimator controlled for time-invariant unobserved heterogeneity and thus allowed for the exploration of within-individual covariance over time. Results of the first-difference panel models are presented as adjusted beta coefficients (aβ) and 95% CI. All the analyses were run using R (version 4.0).

RESULTS

Sample Description

This study included 309 participants, predominantly women (72.8%) originating from Latin America (64.7%) or East Asia (22%) (**Table 1**). Of these 309 participants, 68 (22%) belonged to the regularized group. At baseline, the mean age of the participants was 43.9 years (SD: 10.3). Regardless of the residence status, most participants reported at least a fair level of French proficiency (80.5%) and the mean duration of stay in Geneva was 11.9 years (SD: 4.8). Nonetheless, participants in the regularized group reported better French proficiency and had resided significantly longer in Geneva than those in the undocumented group.

At baseline, the mean equivalent disposable income was CHF 2348—(2157 €; 2539 USD) [SD: CHF 1159—(1064 €; 1253 USD)] the proportion of participants sending remittances to their home country was 69.6% and the proportion of participants having access to a GP was 35% (**Table 2**). The mean equivalent disposable income remained stable over time among regularized participants, while it slightly but significantly increased among undocumented ones. The proportion of participants sending remittances significantly decreased over time in the regularized group, but remained stable overall. In both groups, the number of participants who reported having access to a GP significantly increased.

While the proportion of participants suffering from comorbidity significantly rose from 16.5% (Wave 1) to 23.6% (Wave 2), the proportion of participants reporting depression or anxiety did not significantly change over time.

Medical Consultations in the 12 Months Before the First Data Collection

Figure 2 displays participants' self-reported number of medical consultations in the 12 months prior to the first data collection, as compared to the estimates for 2017 for the general population in Geneva. While all undocumented, participants reported significantly fewer consultations than the general population in Geneva, with the lower quartiles taking on the values of 0 and 1 respectively, the medians of 2 and 3 and the upper quartiles of 4 and 5 respectively. On average, participants reported 2.7 consultations compared to 3.6 for the general population.

Either in the univariate (OR: 0.91; 95% CI: 0.50–1.67) [**Table 3**, Hurdle part (1)] or multivariate analyses (aOR 0.88; 95% CI:

TABLE 2 | Levels of enabling and health needs factors at each period, stratified by residence status.

	Total (*N* = 309)		*p*-value	Control group (*N* = 241)		*p*-value	Regularized group (*N* = 68)		*p*-value
	Wave 1	Wave 2		Wave 1	Wave 2		Wave 1	Wave 2	
Equivalent disposable income (in CHF.-)	2,348 (1,159)	2,441 (1,075)	0.031	2,205 (1,209)	2,346 (1098)	0.018	2,854 (777)	2,777 (918)	0.872
Transfer of remittances	215 (69.6%)	210 (68%)	0.508	170 (70.5%)	173 (71.8%)	0.647	45 (66%)	37 (54.4%)	0.033
Having access to a GP	108 (35%)	153 (49.5%)	<0.001	76 (31.5%)	97 (40.2%)	0.006	32 (47.1%)	56 (82.4%)	<0.001
Comorbidity	51 (16.5%)	73 (23.6%)	0.003	42 (17.4%)	62 (25.7%)	0.004	9 (13.2%)	11 (16.2%)	0.727
Depression or anxiety	57 (18.4%)	62 (20.1%)	0.484	50 (20.7%)	59 (24.5%)	0.170	7 (10.3%)	3 (4.4%)	0.289

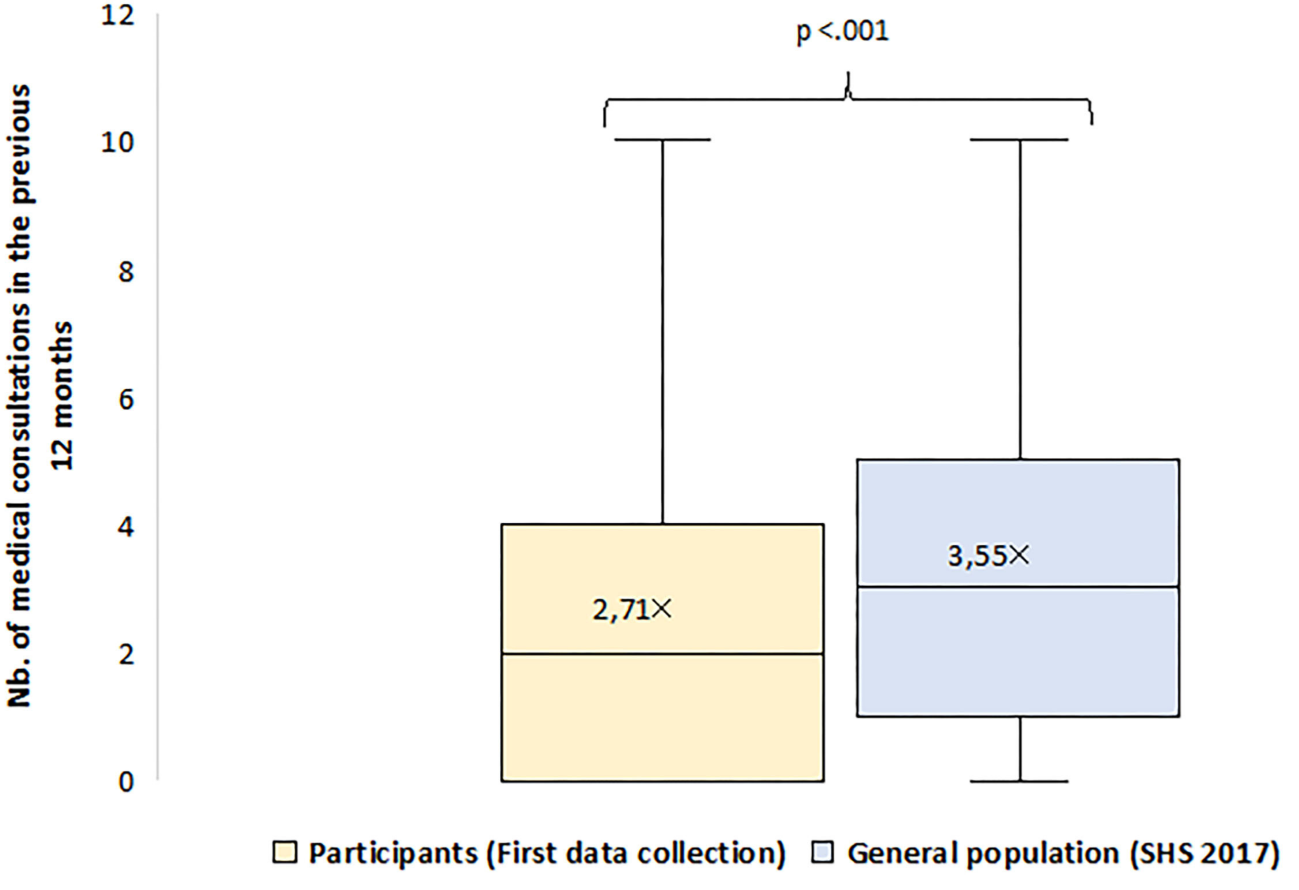


FIGURE 2 | Healthcare utilization of study participants (first data collection), as compared to the general population in Geneva.

0.44–1.77) [**Table 4**, Hurdle part (1)], regularized participants were not more likely to have had at least one medical consultation in the last 12 months than controls. In the multivariate analysis, only being a female (aOR: 2.70; 95% CI: 1.37–5.30), having access to a GP (aOR: 3.15; 95% CI: 1.62–6.13) and suffering from comorbidity (aOR: 6.01; 95% CI: 1.73–20.84) significantly increased the odds of having at least one medical consultation in the last 12 months.

Among participants who reported at least one medical consultation, regularized and control participants reported comparable counts of consultations (aIRR: 1.18; 95% CI: 0.83–1.66) [**Table 4**, Truncated part (1)]. In both univariate and multivariate analyses, having access to a GP (aIRR: 1.45; 95% CI: 1.09–1.92), suffering from comorbidity (aIRR: 1.46; 95% CI: 1.06–2.00) and reporting depression or anxiety (aIRR: 1.80; 95% CI: 1.29–2.51) were the only factors associated with more medical consultations. On the other hand, a higher equivalent disposable income was negatively associated with visits counts (aIRR per additional CHF 100.-: 0.98; 95% CI: 0.97–1.00).

TABLE 3 | Univariate associations between the number of consultations in the previous 12 months and predisposing, enabling and health needs factors.

	Consultations in the previous 12 months prior to the first wave personal interviews				**Consultations in the previous 12 months prior to the second wave personal interviews**			
	Hurdle part (1) (*N* = 309)		**Truncated part (1) (*N* = 227)**		**Hurdle part (2) (*N* = 309)**		**Truncated part (2) (*N* = 218)**	
	OR (95% CI)	***p*-value**	**IRR (95% CI)**	***p*-value**	**OR (95% CI)**	***p*-value**	**IRR (95% CI)**	***p*-value**
Regularized (ref. Controls)	0.91 (0.50, 1.67)	0.767	1.05 (0.72, 1.53)	0.781	1.21 (0.66, 2.21)	0.542	1.55 (1.09, 2.22)	0.016
Female (ref. Male)	2.92 (1.70, 5.01)	<0.001	1.05 (0.72, 1.54)	0.805	2.68 (1.58, 4.55)	<0.001	1.19 (0.80, 1.76)	0.390
Age (per additional year)	1.00 (0.98, 1.03)	0.958	1.00 (0.98, 1.01)	0.629	1.00 (0.98, 1.03)	0.725	1.00 (0.99, 1.02)	0.460
Origin: Asia (ref. Latin America)	0.50 (0.27, 0.93)	0.028	0.71 (0.48, 1.06)	0.095	0.71 (0.38, 1.30)	0.263	1.03 (0.70, 1.52)	0.869
Origin: Eastern Europe (ref. Latin America)	0.26 (0.11, 0.62)	0.002	1.29 (0.67, 2.48)	0.451	0.19 (0.08, 0.46)	<0.001	1.58 (0.74, 3.37)	0.239
Origin: Africa (ref. Latin America)	0.47 (0.17, 1.35)	0.163	1.60 (0.82, 3.12)	0.167	0.45 (0.16, 1.25)	0.126	1.63 (0.79, 3.36)	0.182
Oral proficiency in French: Fair (ref. Good)	1.15 (0.65, 2.01)	0.632	0.94 (0.66, 1.33)	0.713	1.28 (0.73, 2.23)	0.387	0.84 (0.60, 1.20)	0.342
Oral proficiency in French: Poor (ref. Good)	1.31 (0.65, 2.66)	0.448	0.95 (0.62, 1.44)	0.796	0.89 (0.46, 1.71)	0.729	0.78 (0.50, 1.22)	0.272
Duration in Geneva (per additional year)	0.98 (0.93, 1.03)	0.346	1.01 (0.98, 1.05)	0.416	0.98 (0.93, 1.04)	0.558	1.03 (0.99, 1.06)	0.107
Equivalent disposable income (per additional CHF 100.-)	0.97 (0.95, 0.99)	0.008	0.99 (0.97, 1.00)	0.035	0.94 (0.92, 0.97)	<0.001	0.99 (0.98, 1.01)	0.230
Transfer of remittances (ref. No transfer)	0.73 (0.41, 1.29)	0.270	0.77 (0.56, 1.07)	0.114	0.79 (0.47, 1.36)	0.399	0.87 (0.63, 1.22)	0.431
Having access to a GP (ref. No access to GP)	2.56 (1.41, 4.64)	0.002	1.28 (0.94, 1.75)	0.119	1.66 (1.01, 2.72)	0.045	2.05 (1.53, 2.76)	<0.001
Comorbidity (ref. Absence of comorbidity	7.06 (2.14, 23.35)	0.001	1.73 (1.22, 2.45)	0.002	2.89 (1.44, 5.79)	0.003	1.54 (1.11, 2.16)	0.011
Depression or anxiety (ref. Absence of depression and anxiety)	1.88 (0.90, 3.92)	0.092	1.88 (1.33, 2.65)	<0.001	1.55 (0.80, 2.98)	0.208	1.27 (0.87, 1.85)	0.187

TABLE 4 | Multivariate associations between the number of consultations in the previous 12 months and predisposing, enabling and health needs factors.

	Consultations in the previous 12 months prior to the first wave personal interviews				**Consultations in the previous 12 months prior to the second wave personal interviews**			
	Hurdle part (1) (*N* = 309)		**Truncated part (1) (*N* = 227)**		**Hurdle part (2) (*N* = 309)**		**Truncated part (2) (*N* = 218)**	
	aOR (95% CI)	***p*-value**	**aIRR (95% CI)**	***p*-value**	**aOR (95% CI)**	***p*-value**	**aIRR (95% CI)**	***p*-value**
Regularized (ref. Controls)	0.88 (0.44, 1.77)	0.716	1.18 (0.83, 1.66)	0.353	0.96 (0.46, 2.01)	0.904	1.50 (1.07, 2.09)	0.018
Female (ref. Male)	2.70 (1.37, 5.30)	0.004	1.21 (0.82, 1.79)	0.335	2.31 (1.20, 4.44)	0.012	1.32 (0.91, 1.91)	0.141
Age (per additional year)	0.99 (0.96, 1.02)	0.578	0.99 (0.97, 1.00)	0.135	0.99 (0.95, 1.02)	0.380	0.99 (0.98, 1.01)	0.369
Origin: Asia (ref. Latin America)	0.53 (0.26, 1.09)	0.083	0.74 (0.50, 1.08)	0.114	0.93 (0.45, 1.92)	0.835	1.09 (0.81, 1.56)	0.621
Origin: Eastern Europe (ref. Latin America)	0.62 (0.21, 1.82)	0.384	2.07 (1.06, 4.02)	0.032	0.40 (0.13, 1.18)	0.096	1.41 (0.70. 2.83)	0.335
Origin: Africa (ref. Latin America)	0.53 (0.16, 1.76)	0.302	1.32 (0.71, 2.46)	0.374	0.34 (0.10, 1.13)	0.077	1.97 (1.07, 3.64)	0.029
Oral proficiency in French: Fair (ref. Good)	1.05 (0.54, 2.04)	0.878	1.08 (0.80, 1.48)	0.607	1.01 (0.52, 1.96)	0.972	0.94 (0.69. 1.27)	0.671
Oral proficiency in French: Poor (ref. Good)	1.17 (0.47, 2.92)	0.730	1.07 (0.71, 1.60)	0.760	0.56 (0.24, 1.33)	0.191	0.95 (0.63, 1.45)	0.820
Duration in Geneva (per additional year)	0.96 (0.89, 1.03)	0.272	1.02 (0.99, 1.06)	0.246	0.98 (0.92, 1.05)	0.570	1.01 (0.97, 1.04)	0.717
Equivalent disposable income (per additional CHF 100.-)	0.98 (0.95, 1.00)	0.091	0.98 (0.97, 1.00)	0.019	0.94 (0.91, 0.97)	<0.001	0.98 (0.96, 0.99)	0.004
Transfer of remittances (ref. No transfer)	0.78 (0.40, 1.53)	0.470	0.93 (0.69, 1.26)	0.648	1.35 (0.69, 2.61)	0.382	1.22 (0.90, 1.65)	0.191
Having access to a GP (ref. No access to GP)	3.15 (1.62, 6.13)	<0.001	1.45 (1.09, 1.92)	0.011	2.43 (1.28, 4.61)	0.006	2.30 (1.70, 3.11)	<0.001
Comorbidity (ref. Absence of comorbidity)	6.01 (1.73, 20.84)	0.005	1.46 (1.06, 2.00)	0.019	2.61 (1.21, 5.65)	0.015	1.42 (1.03, 1.94)	0.027
Depression or anxiety (ref. Absence of depression and anxiety)	1.29 (0.56, 2.97)	0.547	1.80 (1.29, 2.51)	<0.001	0.84 (0.39, 1.83)	0.667	1.22 (0.87, 1.72)	0.251

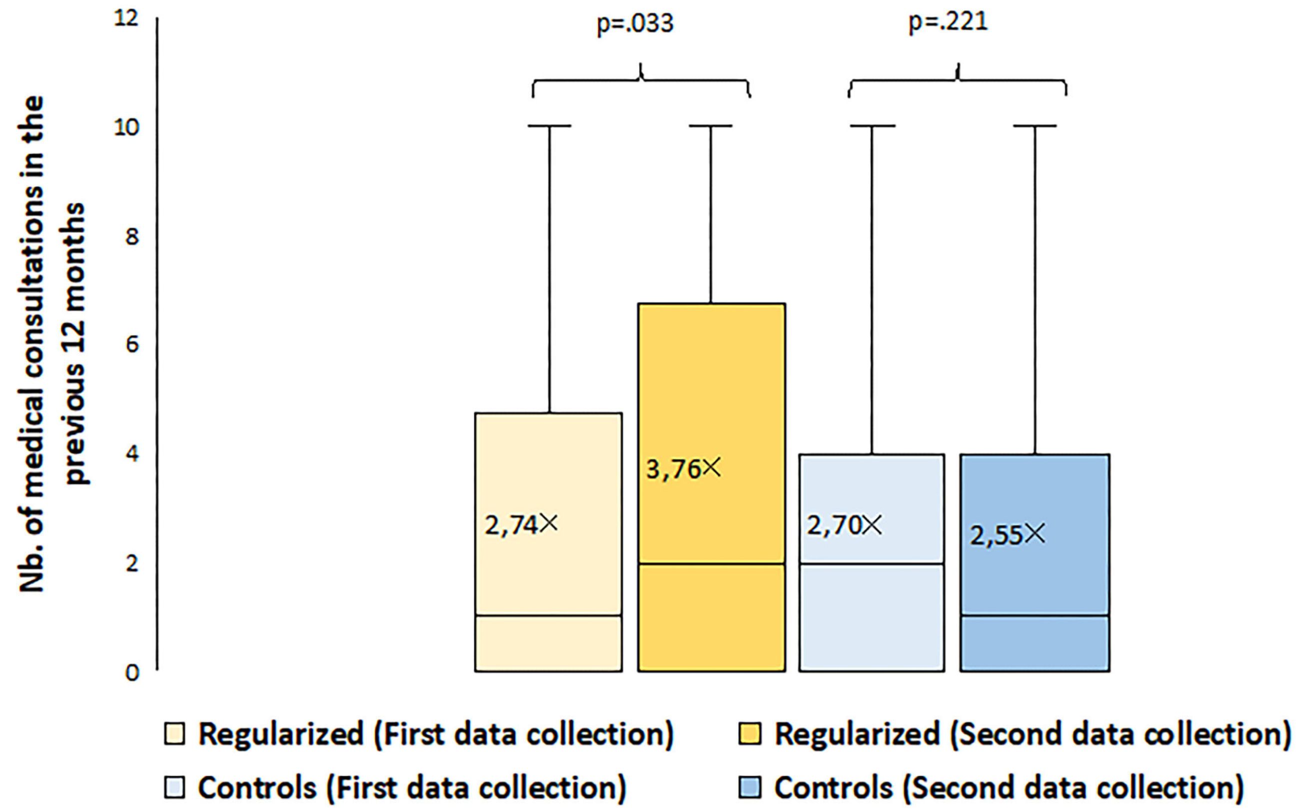


FIGURE 3 | Evolution of participants' healthcare utilization, stratified by residence status groups.

Medical Consultations in the 12 Months Before the Second Data Collection

While the number of medical consultations significantly increased between the first and second data collections among regularized participants, it remained stable in the control group, suggesting a positive relationship between regularization and healthcare utilization (**Figure 3**). Specifically, the average number of reported medical consultations significantly rose from 2.7 to 3.7 among the regularized group, while it non-significantly decreased from 2.7 to 2.6 in the control group.

Overall, factors associated with increased odds of having visited a medical doctor in the 12 months prior to the second data collection were consistent with the results of the first data collection. Specifically, participants in the control group were still as likely as regularized ones to have consulted at least once (aOR: 0.96; 95% CI: 0.46–2.01) In the multivariate analysis, being a female (aOR: 2.31; 95% CI: 1.20-4.44), having access to a GP (aOR: 2.43; 95% CI: 1.28-4.61), comorbidity (aOR: 2.61; 95% CI: 1.21-5.65) and a lower equivalent disposable income (aOR per additional CHF 100.-: 0.94; 95% CI: 0.91-0.97) significantly increased the odds of reporting at least one consultation.

However, among participants who visited a doctor at least once, regularized ones reported significantly higher counts of medical consultations than controls (IRR: 1.55; 95% CI: 1.09–2.22) [**Table 3**, Truncated part (2)]. This difference remained significant after adjustment for predisposing, enabling and health needs factors (aIRR: 1.50; 95% CI: 1.07, 2.09) [**Table 4**, Truncated part (2)]. A lower equivalent disposable income (aIRR per additional CHF 100.-: 0.98; 95% CI: 0.96–0.99), having access to a GP (aIRR: 2.30; 95% CI: 1.70–3.11) and suffering from comorbidity (aIRR: 1.42; 95% CI: 1.03–1.94) also remained significantly associated with higher counts of medical consultations.

Within-Individual Covariance Between Healthcare Utilization and Residence Status Regularization

Results from the first-difference panel model adjusted for time-varying enabling and health needs factors provided further support for a positive association at the individual level between residence status regularization and healthcare utilization. Indeed, regularization of residence status was associated with an increase in the number of medical consultations (aβ: 0.90; 95% CI: 0.31–1.77) (**Table 5**). The equivalent disposable income (aβ per additional CHF 100.-: −0.04; 95% CI: −0.07–0.00) as well as having access to a GP (aβ: 0.86; 95% CI: 0.15–1.56) were also significant predictors of change in the number of medical visits.

TABLE 5 | Within-individual variation in healthcare utilization according to residence status regularization, enabling and health needs factors.

	Change in the number of medical consultations	
	Beta coefficients (95% CI)	***p*-value**
Regularization (>12 months prior to the second wave personal interview) (ref. Undocumented or regularization < 12 months)	0.90 (0.31, 1.77)	0.043
Equivalent disposable income (per additional CHF 100.-)	−0.04 (−0.07, 0.00)	0.043
Transfer of remittances (ref. No transfer)	0.12 (−0.71, 0.94)	0.781
Having access to a GP (ref. No access to GP)	0.86 (0.15, 1.56)	0.017
Comorbidity (ref. Absence of comorbidity)	−0.15 (−0.98, 0.68)	0.723
Depression or anxiety (ref. Absence of depression and anxiety)	0.23 (−0.63, 1.09)	0.602
F-statistic	2.90	0.009
Within R-squared	0.05	

DISCUSSION

Using two-wave panel data, this study provides evidence of a positive association between healthcare utilization and residence status regularization among a sample of undocumented economic migrants in Geneva, Switzerland. For the 12-months period prior to the first data collection, these migrants reported significantly fewer medical consultations than the general population and at this stage, migrants in the regularized group did not differ in their healthcare utilization from those in the control group. However, while the number of visits increased after regularization among the former, it remained stable over the two data collections among the latter. This increase suggested a positive impact of regularization on healthcare utilization, as it led to significant cross-sectional differences between regularized and control participants, even after adjusting for predisposing, enabling and health needs factors. At the within-individual level, the first-difference panel model provided further evidence of a positive impact of regularization on healthcare utilization.

In Switzerland, undocumented migrants' main reasons for avoiding healthcare utilization do not differ from those cited in other European countries (29, 36, 45). Furthermore, factors associated with healthcare utilization among this population are similar to those found in the general population in Switzerland (46). Specifically, we found that women were more likely than men to visit a doctor and that having access to a GP was a key predictor of undocumented migrants' healthcare utilization. We also found that a lower equivalent disposable income was associated with higher medical visit counts, a result consistent with previous studies about undocumented migrants' health needs in Switzerland and elsewhere, which showed that the lack of financial resources increased the odds of reporting poor health (26, 47–49). While financial barriers might hinder healthcare utilization in times of need, especially in countries where the healthcare system is predominantly market-driven, the dedicated unit for undocumented population in Geneva might contribute to bridging the gap between migrants' health needs and their healthcare access, providing a contextual explanation for the negative relationship between income and healthcare utilization. Still, we found that compared to legal residents and natives in Geneva, undocumented migrants reported fewer medical consultations despite the availability of dedicated public health services. While we could not adjust for other factors nor provide a detailed picture of the type of healthcare sought by undocumented migrants, these results are in line with previous findings in various geographical settings. Studies led in England, Denmark, Portugal, Belgium or the Netherlands consistently showed that undocumented migrants were not as likely as the legal immigrants or the natives to consult in primary care services (8, 16, 50–52). In Germany, Castañeda found that undocumented migrants tended to avoid or delay consultations in specialist care services (53). De Jonge et al. made similar observations in the Netherlands, where they found that undocumented pregnant women attended fewer pre-natal visits than their documented counterparts, a finding in line with a similar study conducted in Geneva, Switzerland (54). Overall, our results support the hypothesis that regardless of the country of residence, undocumented migrants are more likely to be disadvantaged in the utilization of healthcare services (12).

In light of this consistent association between lack of residence status and limited use of healthcare services, we hypothesized that regularization could enhance migrants' healthcare utilization through a more secured residence status. In Europe, calls for effective policies and practices improving access to healthcare for undocumented migrants have multiplied in recent years (31, 32). Yet, policy recommendations issued so far mainly focused on the organization of the healthcare system. They rarely encompassed reforms in other areas, such as in migratory or labor policies, to promote healthcare for undocumented migrants (32). To our knowledge, Belgium is the only European country in which the provision of a residence status was explicitly suggested by a panel of experts as a policy instrument to facilitate undocumented migrants' access to treatment for specific infectious diseases, such as tuberculosis (32). To date, only qualitative interviews with regularized migrants suggested that positive effects of regularization policies encompassed improved access to various public benefits such as welfare, social insurances and healthcare (55). Using a quantitative approach, this paper thus bridges a gap. It supports that policies aiming at granting undocumented migrants residence authorizations might improve healthcare utilization for this population and, as a result, foster better health in this community. It also strengthens previous findings in other contexts such as in the US, where policies promoting undocumented migrants' social integration were found to have positive effects on their healthcare utilization (33, 34).

Several limitations should be considered when interpreting the results of this study. Overall, the different sizes of our residence status groups [Regularized ($N = 68$) and Controls ($N = 241$)] reduced statistical power and increased the margins of error. Yet, despite the increased margins of error, we still found a significant association between residence status regularization and healthcare utilization, not only at the between-individual level, but also at the within-individual one. This suggests a strong effect size, i.e., a strong relationship between regularization and healthcare utilization. Nonetheless, our sampled population may not be representative of the undocumented population in Geneva and, a fortiori, in Europe due to convenience sampling. More specifically, we explored the situation of a specific group of stable, well-established undocumented workers, whose socio-economic conditions and health needs are widely different from those of newly arrived migrants at the borders of Europe (56). Furthermore, since 15% of our participants were recruited in the HUG waiting rooms, the sample might have been biased toward healthcare users, leading to slight overestimation of undocumented migrants' healthcare utilization. Convenience sampling also hampers the generalizability of our results, since we cannot exclude unobserved residual confounding due to selection bias. However, the availability of longitudinal data on hard-to-reach migrants and the use of the first-difference estimator minimizes this risk of confounding to unobserved time-variant features. Still, the results should be subject to cautious interpretation regarding causality and the underlying mechanisms at stake. Of note, we could not assess whether the effect of the residence status per se was mediated by affiliation to a health insurance due to collinearity issues. More research is thus needed to better understand the mechanisms through which regularization improves undocumented migrants' use of healthcare services and the mid-to-long term impact of this better access on migrants' health.

CONCLUSION

This study supports the hypothesis that public policies aiming at granting undocumented migrants residence authorizations improve healthcare utilization for this population. It strengthens previous findings that highlighted the positive effects of public policies promoting migrants' inclusion on their use of healthcare services. More research is needed to understand the mechanisms through which regularization improves undocumented migrants' use of healthcare services.

DATA AVAILABILITY STATEMENT

The datasets presented in this article are not readily available because the datasets generated and/or analyzed during the current study are not publicly available due to the temporary embargo on data dissemination until 2023 required by the main funding agency of the study (Swiss National Fund for Scientific Research) but are available from the corresponding author on reasonable request. Requests to access the datasets should be directed to julien.fakhoury@unige.ch.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Ethics Committee of Geneva Canton, Switzerland (CCER 2017–00897). The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

YJ and CB-J designed the Parchemins study, the framework within which this article was elaborated. Data were collected by JF, AD, and LC. JF conducted all the analyses and drafted the manuscript. YJ, CB-J, AD, and LC proofread the manuscript. All authors approved the manuscript.

FUNDING

The Parchemins study was supported by the Geneva University School of Medicine, Fondation Safra, Geneva Directorate of Health, Geneva Directorate of Social Affairs, Swiss Federal Office of Public Health, the NCCR LIVES Project and the Swiss National Fund for Scientific Research (Grant 100017_182208). Funders had no role in the development of the study design, data collection, interpretation, and dissemination.

ACKNOWLEDGMENTS

Authors wish to thank Prof. Giovanni Ferro-Luzzi, all partner organizations, the Swiss Federal Office for Public Health and the Geneva Canton Departments of Health and of Social Affairs for their support in the implementation of this study.

REFERENCES

1. World Health Organization. *Primary Health Care on the Road to Universal Health Coverage.* Geneva: World Health Organization (2019).
2. European Union. *The European Pillar of Social Rights in 20 Principles.* Luxembourg: Publications office of the European Union (2017).
3. United Nation. *Transforming our World: The 2030 Agenda for Sustainable Development.* New York, NY: United Nation (2015).
4. Cuadra CB. Right of access to health care for undocumented migrants in EU: a comparative study of national policies. *Eur J Public Health.* (2012) 22:267–71. doi: 10.1093/eurpub/ckr049
5. Aday LA, Andersen R. A framework for the study of access to medical care. *Health Serv Res.* (1974) 9:208–20.
6. Gelberg L, Andersen RM, Leake BD. The Behavioral Model for Vulnerable Populations: application to medical care use and outcomes for homeless people. *Health Serv Res.* (2000) 34:1273–302. Available online at: https://pubmed.ncbi.nlm.nih.gov/10654830/

7. Legido-Quigley H, Pocock N, Tan ST, Pajin L, Suphanchaimat R, Wickramage K, et al. Healthcare is not universal if undocumented migrants are excluded. *BMJ.* (2019) 366:l4160. doi: 10.1136/bmj.l4160
8. Schoevers MA, Loeffen MJ, van den Muijsenbergh ME, Lagro-Janssen ALM. Health care utilisation and problems in accessing health care of female undocumented immigrants in the Netherlands. *Int J Public Health.* (2010) 55:421–8. doi: 10.1007/s00038-010-0151-6
9. Biswas D, Kristiansen M, Krasnik A, Norredam M. Access to healthcare and alternative health-seeking strategies among undocumented migrants in Denmark. *BMC Public Health.* (2011) 11:560. doi: 10.1186/1471-2458-11-560
10. Larchanché S. Intangible obstacles: health implications of stigmatization, structural violence, and fear among undocumented immigrants in France. *Soc Sci Med.* (2012) 74:858–63. doi: 10.1016/j.socscimed.2011.08.016
11. Poduval S, Howard N, Jones L, Murwill P, McKee M, Legido-Quigley H. Experiences among undocumented migrants accessing primary care in the United Kingdom: a qualitative study. *Int J Health Serv.* (2015) 45:320–33. doi: 10.1177/0020731414568511
12. Winters M, Rechel B, de Jong L, Pavlova M. A systematic review on the use of healthcare services by undocumented migrants in Europe. *BMC Health Serv Res.* (2018) 18:30. doi: 10.1186/s12913-018-2838-y
13. Wolff H, Epiney M, Lourenco AP, Costanza MC, Delieutraz-Marchand J, Andreoli N, et al. Undocumented migrants lack access to pregnancy care and prevention. *BMC Public Health.* (2008) 8:93. doi: 10.1186/1471-2458-8-93
14. Castañeda H. Paternity for Sale: *Med Anthropol Q.* (2008) 22:340–359. doi: 10.1111/j.1548-1387.2008.00039.x
15. Singh S, Siddhanta A. Cross border migration and HIV vulnerability in South Asian countries. *J Transm Dis Immun.* (2017) 1. doi: 10.21767/2573-0320.10005
16. Dias SF, Severo M, Barros H. Determinants of health care utilization by immigrants in Portugal. *BMC Health Serv Res.* (2008) 8:207. doi: 10.1186/1472-6963-8-207
17. Jackson Y, Paignon A, Wolff H, Delicado N. Health of undocumented migrants in primary care in Switzerland. *PLoS ONE.* (2018) 13:e0201313. doi: 10.1371/journal.pone.0201313
18. Ahonen EQ, Porthé V, Vázquez ML, García AM, López-Jacob MJ, Ruiz-Frutos C, et al. Project* for the I. A qualitative study about immigrant workers' perceptions of their working conditions in Spain. *J Epidemiol Community Health.* (2009) 63:936–42. doi: 10.1136/jech.2008.077016
19. Sousa E, Agudelo-Suárez A, Benavides FG, Schenker M, García AM, Benach J, et al. Immigration, work and health in Spain: the influence of legal status and employment contract on reported health indicators. *Int J Public Health.* (2010) 55:443–51. doi: 10.1007/s00038-010-0141-8
20. Benach J, Muntaner C, Delclos C, Menéndez M, Ronquillo C. Migration and "low-skilled" workers in destination countries. *PLoS Med.* (2011) 8:e1001043. doi: 10.1371/journal.pmed.1001043
21. Myhrvold T, Småstuen MC. The mental healthcare needs of undocumented migrants: an exploratory analysis of psychological distress and living conditions among undocumented migrants in Norway. *J Clin Nurs.* (2017) 26:825–39. doi: 10.1111/jocn.13670
22. De Vito E, de Waure C, Specchia ML, Ricciardi W. *Public Health Aspects of Migrant Health: A Review of the Evidence on Health Status for Undocumented Migrants in the European Region*. Copenhagen: WHO Regional Office for Europe (2015).
23. Sebo P, Jackson Y, Haller DM, Gaspoz J-M, Wolff H. Sexual and reproductive health behaviors of undocumented migrants in Geneva: a cross sectional study. *J Immigrant Minority Health.* (2011) 13:510–7. doi: 10.1007/s10903-010-9367-z
24. Wolff H, Lourenço A, Bodenmann P, Epiney M, Uny M, Andreoli N, et al. Chlamydia trachomatis prevalence in undocumented migrants undergoing voluntary termination of pregnancy: a prospective cohort study. *BMC Public Health.* (2008) 8:391. doi: 10.1186/1471-2458-8-391
25. Keygnaert I, Vettenburg N, Roelens K, Temmerman M. Sexual health is dead in my body: participatory assessment of sexual health determinants by refugees, asylum seekers and undocumented migrants in Belgium and the Netherlands. *BMC Public Health.* (2014) 14:416. doi: 10.1186/1471-2458-14-416
26. Fakhoury J, Burton-Jeangros C, Consoli L, Duvoisin A, Courvoisier D, Jackson Y. Mental health of undocumented migrants and migrants undergoing regularization in Switzerland: a cross-sectional study. *BMC Psychiatry.* (2021) 21:175. doi: 10.1186/s12888-021-03149-7
27. Kuehne A, Huschke S, Bullinger M. Subjective health of undocumented migrants in Germany–a mixed methods approach. *BMC Public Health.* (2015) 15:926. doi: 10.1186/s12889-015-2268-2
28. D'Egidio V, Mipatrini D, Massetti AP, Vullo V, La Torre G. How are the undocumented migrants in Rome? Assessment of quality of life and its determinants among migrant population. *J Public Health.* (2017) 39:440–6. doi: 10.1093/pubmed/fdw056
29. Chauvin P, Parizot I, Simonnot N. *Access to Healthcare for Undocumented Migrants in 11 European Countries*. Paris: Médecins du monde (2009).
30. Bozorgmehr K, Razum O. Effect of restricting access to health care on health expenditures among asylum-seekers and refugees: a quasi-experimental study in Germany, 1994–2013. *PLoS ONE.* (2015) 10:e0131483. doi: 10.1371/journal.pone.0131483
31. De Vito E, de Waure C, Specchia ML, Parente P, Azzolini E, Frisicale EM, et al. Are undocumented migrants' entitlements and barriers to healthcare a public health challenge for the European Union? *Public Health Rev.* (2016) 37:13. doi: 10.1186/s40985-016-0026-3
32. Dauvrin M, Derluyn I, Coune I, Verrept H, Lorant V. Towards fair health policies for migrants and ethnic minorities: the case-study of ETHEALTH in Belgium. *BMC Public Health.* (2012) 12:726. doi: 10.1186/1471-2458-12-726
33. Korinek K, Smith KR. Prenatal care among immigrant and racial-ethnic minority women in a new immigrant destination: exploring the impact of immigrant legal status. *Soc Sci Med.* (2011) 72:1695–703. doi: 10.1016/j.socscimed.2011.02.046
34. Howell E, Trenholm C, Dubay L, Hughes D, Hill I. The impact of new health insurance coverage on undocumented and other low-income children: lessons from three California counties. *J Health Care Poor Underserved.* (2010) 21:109–24. doi: 10.1353/hpu.0.0293.697
35. Morlok Mi, Oswald A, Meier H, Efionayi-Mäder D, Ruedin D, Bader D, Wanner P. *Les sans-papiers en Suisse en 2015*. Basel: BSS (2016)
36. Wyssmüller C, Efionayi-Mäder D. Undocumented migrants: their needs and strategies for accessing health care in Switzerland: Country Report on People & Practices. Bern: Swiss Forum for Population and Migration Studies (2011).
37. Wolff H, Stalder H, Epiney M, Walder A, Irion O, Morabia A. Health care and illegality: a survey of undocumented pregnant immigrants in Geneva. *Soc Sci Med.* (2005) 60:2149–54. doi: 10.1016/j.socscimed.2004.12.007
38. Jackson Y, Courvoisier DS, Duvoisin A, Ferro-Luzzi G, Bodenmann P, Chauvin P, et al. Impact of legal status change on undocumented migrants' health and well-being (Parchemins): protocol of a 4-year, prospective, mixed-methods study. *BMJ Open.* (2019) 9:e028336. doi: 10.1136/bmjopen-2018-028336
39. Da Silva RB, Contandriopoulos A-P, Pineault R, Tousignant P, A. Global approach to evaluation of health services utilization: concepts and measures. *Healthc Policy.* (2011) 6:e106–17. doi: 10.12927/hcpol.2011.22351
40. Ortega AN, Fang H, Perez VH, Rizzo JA, Carter-Pokras O, Wallace SP, et al. Health care access, use of services, and experiences among undocumented Mexicans and other Latinos. *Arch Intern Med.* (2007) 167:2354–60. doi: 10.1001/archinte.167.21.2354
41. Vargas Bustamante A, Fang H, Garza J, Carter-Pokras O, Wallace SP, Rizzo JA, et al. Variations in healthcare access and utilization among Mexican immigrants: the role of documentation status. *J Immigr Minor Health.* (2012) 14:146–55. doi: 10.1007/s10903-010-9406-9
42. Gimeno-Feliu LA, Pastor-Sanz M, Poblador-Plou B, Calderón-Larrañaga A, Díaz E, Prados-Torres A. Overuse or underuse? *Use of healthcare services among irregular migrants in a north-eastern Spanish region. Int J Equity Health.* (2021) 20:41. doi: 10.1186/s12939-020-01373-3
43. Fortin M, Stewart M, Poitras M-E, Almirall J, Maddocks H, A. Systematic review of prevalence studies on multimorbidity: toward a more uniform methodology. *Ann Fam Med.* (2012) 10:142–51. doi: 10.1370/afm.1337
44. Swiss Federal Office of Public Health. *Enquête suisse sur la santé 2017.* (2019). Available online at: https://www.bfs.admin.ch/bfs/fr/home/statistiken/gesundheit/erhebungen/sgb.html (accessed April 7, 2020).
45. Woodward A, Howard N, Wolffers I. Health and access to care for undocumented migrants living in the European Union: a scoping review. *Health Policy Plan.* (2014) 29:818–30. doi: 10.1093/heapol/czt061

46. Zufferey J. *La santé dans le canton de Genève: Résultats de l'Enquête suisse sur la santé 2017*. Neuchâtel: Observatoire suisse de la santé (2020).
47. Fakhoury J, Burton-Jeangros C, Guessous I, Consoli L, Duvoisin A, Jackson Y. Self-rated health among undocumented and newly regularized migrants in Geneva: a cross-sectional study. *BMC Public Health.* (2021) 21:1198. doi: 10.1186/s12889-021-11239-0
48. Jolivet A, Cadot E, Florence S, Lesieur S, Lebas J, Chauvin P. Migrant health in French Guiana: Are undocumented immigrants more vulnerable? *BMC Public Health.* (2012) 12:53. doi: 10.1186/1471-2458-12-53
49. Cloos P, Ndao EM, Aho J, Benoît M, Fillol A, Munoz-Bertrand M, Ouimet M-J, Hanley J, Ridde V. The negative self-perceived health of migrants with precarious status in Montreal, Canada: a cross-sectional study. *PLoS ONE.* (2020) 15:e0231327. doi: 10.1371/journal.pone.0231327
50. Ehmsen BK, Biswas D, Jensen NK, Krasnik A, Norredam M. Undocumented migrants have diverse health problems. *Dan Med J.* (2014) 61:A4897. Available online at: https://pubmed.ncbi.nlm.nih.gov/25186539/
51. Britz JB, McKee M. Charging migrants for health care could compromise public health and increase costs for the NHS. *J Public Health (Oxf).* (2016) 38:384–90. doi: 10.1093/pubmed/fdv043
52. Goossens MCM, Depoorter AM. Contacts between general practitioners and migrants without a residence permit and the use of "urgent" medical care. *Scand J Public Health.* (2011) 39:649–55. doi: 10.1177/1403494811414249
53. Castañeda H. Illegality as risk factor: A survey of unauthorized migrant patients in a Berlin clinic. *Soc Sci Med.* (2009) 68:1552–60. doi: 10.1016/j.socscimed.2009.01.024
54. Jonge A de, Rijnders M, Agyemang C, Stouwe R. van der, Otter J den, Muijsenbergh METCV den, et al. Limited midwifery care for undocumented women in the Netherlands. *J Psychosom Obstet Gynecol.* (2011) 32:182–8. doi: 10.3109/0167482X.2011.589016
55. Kraler A, Reichel D, König A. *Feasibility Study on the Labour Market Trajectories of Regularised Immigrants Within the European Union (REGANE I).* Vienna: International Centre for Migration Policy Development (ICMP) (2014).
56. Medecins Sans Frontieres. *Constructing Crisis at Europe's Borders*. Médecins Sans Frontières (MSF) International (2021).

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: All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

Copyright © 2022 Fakhoury, Burton-Jeangros, Consoli, Duvoisin and Jackson. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

frontiers | Frontiers in Public Health

TYPE Original Research
PUBLISHED 25 July 2022
DOI 10.3389/fpubh.2022.940619

Check for updates

OPEN ACCESS

EDITED BY
Miodraga Stefanovska-Petkovska,
Universidade de Lisboa, Portugal

REVIEWED BY
Masoud Mohammadnezhad,
Fiji National University, Fiji
Clement K. Edet,
Rivers State University, Nigeria

*CORRESPONDENCE
Yihua Xu
xuyihua_6@hotmail.com
Chenxi Liu
liu_chenxi@hust.edu.cn

SPECIALTY SECTION
This article was submitted to
Life-Course Epidemiology and Social
Inequalities in Health,
a section of the journal
Frontiers in Public Health

RECEIVED 10 May 2022
ACCEPTED 06 July 2022
PUBLISHED 25 July 2022

CITATION
Hu H, Xu Y, Shao Y, Liang Y, Wang Q, Luo S, Lu H, Meng H and Liu C (2022) A latent profile analysis of residents' knowledge, attitude, and practice toward common chronic diseases among ethnic minority area in China. *Front. Public Health* 10:940619. doi: 10.3389/fpubh.2022.940619

COPYRIGHT
© 2022 Hu, Xu, Shao, Liang, Wang, Luo, Lu, Meng and Liu. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

A latent profile analysis of residents' knowledge, attitude, and practice toward common chronic diseases among ethnic minority area in China

Huaqin Hu[1], Yihua Xu[1]*, Yingshan Shao[1], Yaxin Liang[1], Qionghua Wang[2], Shunmei Luo[2], Heyun Lu[2], Heng Meng[1] and Chenxi Liu[3]*

[1]School of Public Health, Tongji Medical College, Huazhong University of Science and Technology, Wuhan, China, [2]Lincang Second People's Hospital, Lincang, China, [3]School of Medicine and Health Management, Tongji Medical College, Huazhong University of Science and Technology, Wuhan, China

Background: Health literacy plays an important role in preventing and managing chronic diseases, while low levels of health literacy among ethnic minorities are a major manifestation of health inequities. We believe that before effective health literacy intervention strategies, it is preferable to understand the features of health literacy among ethnic minorities. The present study firstly updated insights on health literacy among ethnic minorities by investigating the knowledge, attitude, and practice (KAP) profile of common chronic diseases in ethnic minority areas, and secondly discussed the KAP profiles in detail to inspire future health education interventions.

Methods: A cross-sectional, health-literacy-sensitive study was conducted in China's typical ethnic minority area. Participants included 801 adult residents who lived in the ethnic minority area. The primary outcome was participant scores on the KAP questionnaire of common chronic diseases, followed by latent profile analysis to identify participants with similar KAP score patterns and determine whether membership in specific groups was associated with demographic or clinical characteristics.

Results: The participants included 496 ethnic minorities (61.9%) and 305 Han Chinese (38.1%). Three-profile solution was determined after the latent profile analysis: incomplete transfer [I.T.] ($n = 215$), better practice [B.P.] ($n = 301$), and average [A.V.] ($n = 285$). IT group (26.84%) was characterized by the highest level of knowledge and attitude toward common chronic diseases and below average level for practice. Participants in B.P. group performed poorly in both knowledge and attitude toward common chronic diseases but had the highest level of practice. A.V. group reflected average knowledge, attitude, and practice toward common chronic diseases among three subgroups. Ethnic minorities were the dominant population in A.V. group (68.8%). Compared with other groups, the A.V. group contained the largest proportions of married participants (84.2%), participants with no formal education (46.7%), and high annual out-of-pocket medical expense (33.3%).

Conclusion: A more specific and nuanced understanding of minority health literacy can enable service providers to provide more effective health education to their recipients, thereby improving health inequities.

KEYWORDS

knowledge-attitude-practice (KAP), chronic disease, ethnic minorites, health literacy, latent profile analysis (LPA)

Introduction

Globally, chronic diseases have become one of the greatest threats to population health (1). Health illiteracy is the inability to understand and use medical information, which can affect access to and use of the health care system, and is a significant contributor to the burden of non-communicable diseases (2, 3). Conversely, as an indicator of an individual's ability to access, understand, assess, and use health information and make health-related decisions to maintain their health (4), health literacy plays an important role in the prevention and management of chronic diseases. Today, the differences in the distribution of health literacy among minority and mainstream populations have attracted the attention of researchers as a major manifestation of so-called health inequities (5).

The evidence supporting the role of health literacy in chronic disease prevention and management is based on health skills research (6). However, health literacy is not easily quantifiable, especially considering that "it (health literacy) is a dynamic construct that emerges from the interaction between patients/citizens and health care systems, organizations, and professionals" (7). Generally speaking, ethnic minority populations are at a low level of health literacy, and it is our concern to improve their health literacy through effective intervention strategies (8–10). Thus, it is a priority to understand the nature of their health literacy.

Despite the centrality of ethnic minorities' health literacy, empirical work on individual differences is nonexistent. At the individual level, knowledge, attitudes, and practice (KAP) toward common chronic diseases are critical components of health literacy (11). KAP theory itself is widely used in chronic disease health promotion. However, it is often used for post-intervention assessment rather than as a reference for pre-intervention design (12–14). The present study addresses this gap by investigating the profile of KAP regarding common chronic diseases among residents of a typical ethnic minority area. We will describe the diversity of these KAP profiles in detail to identify populations with similar KAP patterns and discuss their implications for future health literacy intervention as a work in progress.

Methods

Study design and participants

This study is a pilot study for a health literacy precise intervention study. The intervention study will be conducted in Linxiang District, Yunnan Province, Southwest China, and we divided the 10 townships in the district into ethnic minority areas (two townships) and non-ethnic minority areas (eight townships) using the percentage of ethnic minority population (21.88%) in the district in 2020 as the threshold. We will recruit adult participants aged 18 years or older in ethnic minority areas to implement a health literacy precise intervention based on latent profile analysis. In March 2021, we conducted a cross-sectional survey in Nami Township, one of the two ethnic minority areas. Participants were eligible if they: (1) were permanent residents (continuous cumulative residence in the area for at least 6 months) of Nanmei Township; (2) age 18 years or older; (3) voluntarily participate in the questionnaire and informed consent. Exclusion criteria: (1) age less than 18 years; (2) considered professionally unsuitable for the study (e.g., incapacity).

Ethical consideration

Participants provided written informed consent and received a free physical examination for participation. The local hospital's ethics committee approved the protocol.

Settings

According to the latest national census data, 55 ethnic minorities account for about 8.89% (125.47 million) of China's 1.4 billion people. One of their major settlements is in Yunnan province in rural southwest China. This survey was conducted in Nanmei township, an ethnic minority settlement in Linxiang District, Yunnan Province. 4,906 people lived in Nanmei township at the end of 2020, of which 4,160 (84.8%) were ethnic minorities, and the majority of the ethnic minority population was Lahu, with 3,547 people, accounting for 85.3% of the total ethnic minority population. The area is remote, economically backward, and lacks health resources. The per capita disposable income in 2020 is 10,530 yuan (Yunnan Province: 23,295 yuan), and there are 1.23 (Yunnan Province: 2.5) practicing physicians per 1,000 people. which is far below the average of Yunnan Province.

Sampling

The following formula performed the sample size calculation:

$$N = \frac{Z_{1-\alpha/2}^{2} \times \pi(1-\pi)}{\delta^{2}}$$

(π : expected adequate KAP rate; δ: sampling error)

According to Zhang et al. (15), the adequate KAP rate of chronic diseases among ethnic minority populations in China was 47.9%. In this study, $\pi = 0.48$, $Z_{1-\alpha/2}^{2} = 1.96$, $\delta = 0.1\ \pi = 0.048$, $\alpha = 0.05$. We found that the required sample size was 435. To accommodate the design effect, the calculated sample size was multiplied by 1.8 for correction, alongside the 20% refusal rate, the final sample size of the survey was determined to be 870.

A systematic sampling method was used to select one out of every five households in 1,388 households in Nanmei Town. A total of 290 households were sampled, yielding 872 potential survey respondents, with 801 people actually completing the survey. Uniformly trained investigators will conduct household surveys of households determined at the sampling stage, each investigator equipped with an investigator's handbook for inquiries, and bring an interpreter with them when investigating ethnic minority participants. Han Chinese survey respondents and minority respondents who speak Chinese completed the survey in Chinese, and minority respondents who do not speak Chinese had their questions verbally translated by an interpreter to complete the survey.

Development of the KAP questionnaire for common chronic diseases

We extracted a pool of items on knowledge, attitude, and practice (KAP) related to four common chronic diseases (hypertension, diabetes, obesity, and chronic obstructive pulmonary disease) from previous research (16–19) and then selected appropriate items from the pool to form the KAP questionnaire of common chronic diseases used in this study, and conducted a pre-survey (50 participants included).

Knowledge was measured by statements such as: "Overeating oily food regularly can cause high cholesterol," "Smoking is a factor in causing chronic obstructive pulmonary disease (COPD)," and "Blood pressure has two values: systolic (high) and diastolic (low)," "Diabetes have far reaching effects on various body organs such as the eyes, nerves, feet, and kidneys," and so on. Participants responded with discrete answers (True/False/Uncertain) and got 1 point for a correct answer; otherwise, no point. In addition, some questions were reverse coded, e.g., "Passive smoking does not cause chronic obstructive pulmonary disease."

Attitudes were measured by the following statements. "You are willing to learn about healthy diet," "You believe that systematic rehabilitation can slow the progression of COPD," "You believe that high salt consumption can lead to high blood pressure," "If you are considered a high-risk population for diabetes, you feel you should acquire knowledge about diabetes," and so on. Participants selected "Agree" (1 point), "Not sure" (0 points), or "Disagree" (0 points) from the Likert items. Some statements were reverse coded, e.g., "Patients with COPD should not be receiving long-term oxygen therapy because of the potential for dependence."

Practice was measured by the healthy lifestyle and practice standards proposed in the 66 health literacy items for Chinese residents published by the National Center for Health Education, including smoking (1 point), alcohol consumption (1 point), fruit and vegetable intake (2 points), fried food intake (1 point), sleep (1 point), physical activity (1 point), chronic diseases prevention and control (4 points).

Results from the pre-survey showed that the internal consistency of the subscales was adequate: 0.77 for knowledge (mean score: 17.74 ± 0.2.5 out of a total score of 24), 0.83 for attitude (mean score: 17.44 ± 2.1, out of a total score of 20), and 0.71 for practice (mean score: 5.76 ± 1.5 out of a total score of 11). The content validity of the questionnaire was rated by eight professionals with expertise in chronic disease prevention and control, and the overall content validity index (CVI) of the questionnaire was 0.83.

Other study measures

The participants' age, sex, and nationality were obtained *via* residents' I.D. cards. Other information (education level, marital status, annual income, living with whom, provided by whom, and annual out-of-pocket medical expense) was collected *via* participants' reports. Chronic disease conditions were measured as if participants had been diagnosed with one of the four common chronic diseases or any another one.

Statistical analysis

We firstly standardized each KAP subscale score; each score of the subscale was subtracted from the mean of the subscale scores and then divided by the root mean square of the subscale scores. Then, we performed a latent profile analysis of the standardized score to identify sub-populations of participants presenting distinct KAP profiles toward common chronic diseases with a robust estimator of nonnormality (robust maximum likelihood). We examined solutions with 1 to 6 distinct profiles and replicated each latent profile solution ten times, beginning at random starting values. We considered the best solution to be parsimonious, to have profiles with conceptual meaning, and to have the best fit (as indicated by the lowest Bayesian information criterion and integrated completed likelihood). Bootstrap Likelihood Ratio Test (BLRT) was conducted to observe if an increase in profiles increases fit. Although the Bayesian information criterion and integrated completed likelihood were slightly better for the 5-profile solution and BLRT showed no significant difference between the models of 5- and 6- profile, we chose the 3- profile solution as providing the most conceptually coherent description of participants' KAP features for common chronic diseases. We assigned participants to the latent profile group for which they had the highest membership probability. Descriptive statistics for demographic and clinical characteristics of participants are presented for the full sample and by latent profile groups. To explore potential differences in characteristics across latent profile groups, we performed multinomial logistic regression analyses. All *p*-values represent 2-sided hypothesis tests with a set significance level of 0.05. All analyses were conducted using R 4.1.2 (20).

Results

We enrolled 801 participants after screening 872 participants regarding potential participation (91.85% participation rate). The most common reasons offered for declining to participate included "not interested" [31 (43.1% of nonparticipants)], "physical reasons" [9 (12.5%)], and "don't have enough time" [32 (44.4%)].

The majority of participants in our sample were ethnic minorities (61.9%), and the rest were Han Chinese (38.1%). The mean age of participants was 55 years-old. Thirty-six (4.49%) participants were with education level of high school and above, 110 (13.7%) were with education level of middle school, 347 (43.3%) were education level of primary school, and 308 (38.5%) were without formal education. The annual income of 321 (40.1%) participants was less than the per capita disposable income of Yunnan Province in 2020, 287 (35.8%) participants had an annual income within two times the per capita disposable income of Yunnan Province in 2020, and 193 (24.1%) participants had an annual income more than two times the per capita disposable income of Yunnan Province in 2020. 331 participants (41.9%) relied on others to provide for their living expenses. Two hundred forty-nine (31.1%) participants self-reported hypertension, 63 (7.87%) participants self-reported diabetes, and 483 (60.3%) participants self-reported no chronic diseases. In addition, six participants (0.75%) reported other chronic diseases such as ischemic stroke or rheumatoid arthritis.

The distribution of the KAP score was shown in Figure 1. Participants scored from 1 to 22 on the knowledge section (median: 7 points, IQR: 5–13), from 0 to 19 on the attitude section (median: 6 points, IQR: 3–13), and from 2 to 9 on the practice section (median: 6 points, IQR: 5–7).

Common patterns of participants' KAP scores toward common chronic diseases

Three-profile solution was determined after the latent profile analysis: incomplete transfer [I.T.] ($n = 215$), better practice [B.P.] ($n = 301$), and average [A.V.] ($n = 285$). The propensity of the KAP score defines each profile and reveals its differences. The first profile (26.84%) was characterized by the highest levels of knowledge and attitudes toward common chronic diseases and below-average levels of practice, indicating that the transfer from knowledge and attitudes to practice was incomplete among participants in this subgroup and, therefore, was labeled as "incomplete transfer" (I.T). Participants in this group may be rich in chronic disease-related knowledge and have developed positive attitudes, but lack the ability to engage in appropriate health practices, perhaps with barriers to practice that are not easily overcome or that entail greater costs that offset the benefits of practice. The second profile (37.58%) was characterized by poor performance in terms of knowledge and attitudes toward common chronic diseases, but the highest level of practice, referred to as "better practice" (B.P.). Subjects in this group may not fully understand chronic disease-related knowledge or attitudes due to a low level of education, but are willing (or compelled) to improve their health practices as recommended by their physicians. Lastly, the third profile (35.59%) was reflected average knowledge, attitude, and practice toward common chronic diseases among three subgroups, which was labeled "average" (A.V.). These "average" participants' knowledge, attitudes, and practices toward common chronic diseases fell between the I.T. and B.P. groups, and they may have some level of knowledge and positive attitudes related to chronic diseases, but occasionally encounter obstacles in their practices (Figure 2).

Characteristics of participants in distinct KAP latent profile groups

We examined whether the three KAP latent profile groups were associated with characteristics of participants (Table 1). Overall differences were found in a few characteristics (nation, education level, marriage status, living arrangement, annual family income, self-reported chronic disease, and annual out-of-pocket medical expense). Odds ratios were obtained after being adjusted by multinomial logistic regressions (Table 2).

Participants with higher education level were less likely to be in B.P. (OR = 0.14, $p < 0.01$) or A.V. (OR = 0.84,

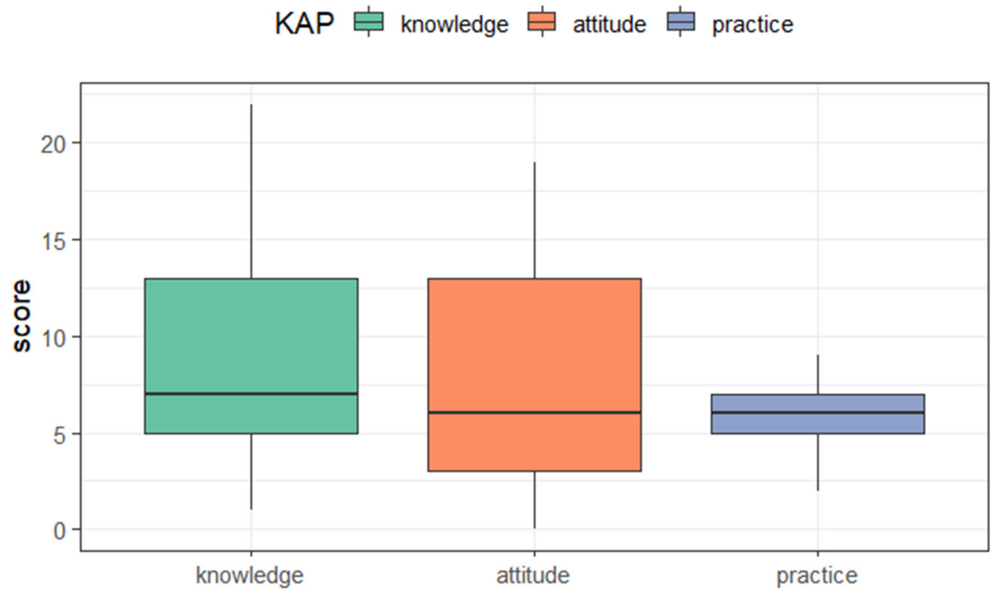


FIGURE 1
The distribution of KAP score.

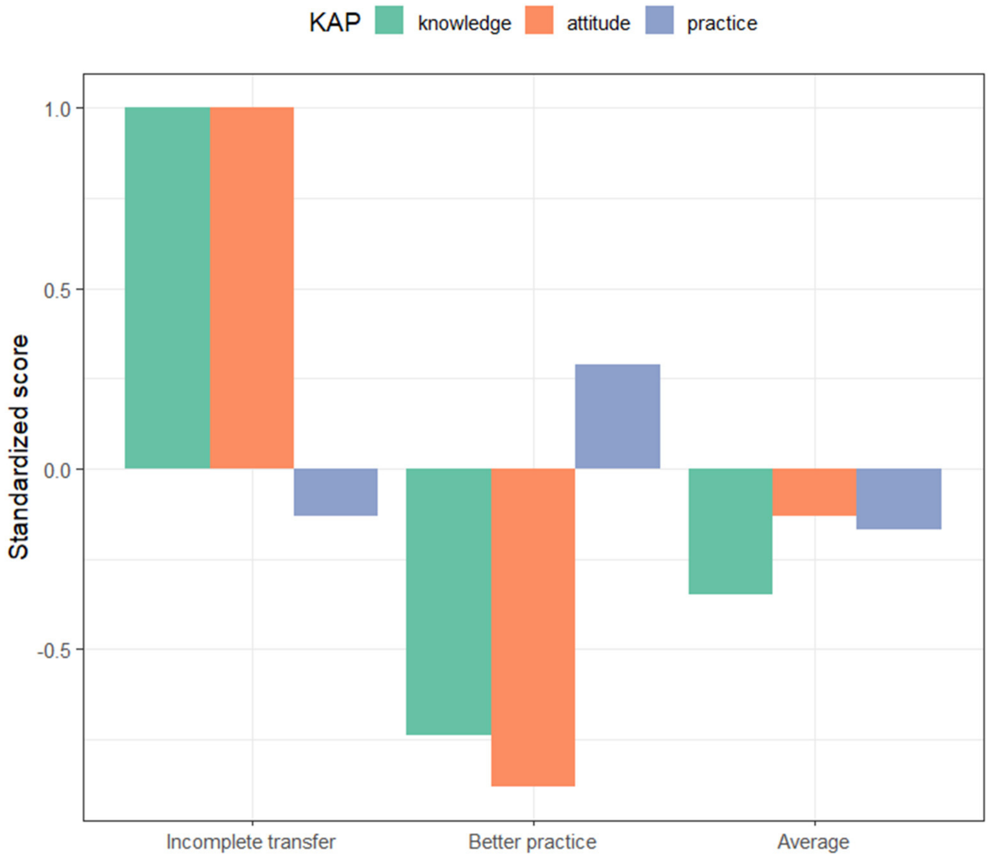


FIGURE 2
Participants' KAP profiles.

TABLE 1 Participant characteristics for full sample and by KAP latent profile groups.

Characteristics	All $N = 801$	Incomplete transfer $N = 215$	Better practice $N = 301$	Average $N = 285$	p-Value
Sex:					0.262
Male	382 (47.7%)	104 (48.4%)	133 (44.2%)	145 (50.9%)	
Female	419 (52.3%)	111 (51.6%)	168 (55.8%)	140 (49.1%)	
Age	55.1 (17.3)	56.8 (17.3)	53.5 (17.3)	55.5 (17.1)	0.095
Nation:					<0.001
Han	305 (38.1%)	129 (60.0%)	87 (28.9%)	89 (31.2%)	
Ethnic minorities	496 (61.9%)	86 (40.0%)	214 (71.1%)	196 (68.8%)	
Education:					<0.001
No formal education	308 (38.5%)	59 (27.4%)	116 (38.5%)	133 (46.7%)	
Primary school	347 (43.3%)	105 (48.8%)	142 (47.2%)	100 (35.1%)	
Middle school	110 (13.7%)	39 (18.1%)	37 (12.3%)	34 (11.9%)	
High school and above	36 (4.49%)	12 (5.58%)	6 (1.99%)	18 (6.32%)	
Marriage:					0.547
Unmarried	39 (4.87%)	10 (4.65%)	18 (5.98%)	11 (3.86%)	
Married	651 (81.3%)	173 (80.5%)	238 (79.1%)	240 (84.2%)	
Divorced	111 (13.9%)	32 (14.9%)	45 (15.0%)	34 (11.9%)	
Living arrangement:					<0.001
Alone	24 (3.00%)	7 (3.26%)	8 (2.66%)	9 (3.16%)	
With spouse	374 (46.7%)	64 (29.8%)	167 (55.5%)	143 (50.2%)	
With children	351 (43.8%)	128 (59.5%)	103 (34.2%)	120 (42.1%)	
With other relatives	52 (6.49%)	16 (7.44%)	23 (7.64%)	13 (4.56%)	
Annual income:					<0.001
Low	321 (40.1%)	68 (31.6%)	152 (50.5%)	101 (35.4%)	
Moderate	287 (35.8%)	81 (37.7%)	95 (31.6%)	111 (38.9%)	
High	193 (24.1%)	66 (30.7%)	54 (17.9%)	73 (25.6%)	
Living expense:					0.638
Self-supplied	470 (58.7%)	132 (61.4%)	174 (57.8%)	164 (57.5%)	
Supplied by others	331 (41.3%)	83 (38.6%)	127 (42.2%)	121 (42.5%)	
Annual out-of-pocket expense:					<0.001
Affordable	651 (81.3%)	196 (91.2%)	278 (92.4%)	177 (62.1%)	
High	125 (15.6%)	9 (4.19%)	21 (6.98%)	95 (33.3%)	
Catastrophic	25 (3.12%)	10 (4.65%)	2 (0.66%)	13 (4.56%)	
Self-reported hypertension	249 (31.1%)	100 (46.5%)	66 (21.9%)	83 (29.1%)	<0.001
Self-reported diabetes	63 (7.87%)	15 (6.98%)	37 (12.3%)	11 (3.86%)	0.001
Self-reported no chronic disease[a]	483 (60.3%)	102 (47.4%)	189 (62.8%)	192 (67.4%)	<0.001

[a] None of the participants reported COPD or obesity.

$p = 0.73$) group than I.T. group. The I.T. group had the largest percentage participants who lived with their children (59.5%), and had high annual income (30.7%), most participants in this group were self-supplied (61.4%), nearly half reported having hypertension (46.5%). Ethnic minorities were more likely to be in B.P. group than I.T (OR = 3.46, $p < 0.01$) or A.V. group (OR = 1.37, $p = 0.15$). This group also had the largest proportions of participants who lived with spouse (55.5%), had low annual income (50.5%) and affordable annual out-of-pocket medical expense (92.4%). Participants who self-reported no chronic disease were more likely to be in the A.V. group than I.T. (OR = 2.68, $p = 0.02$) or B.P. group (OR = 0.27, $p < 0.01$).

TABLE 2 Logistic regression analysis of differences in characteristics across KAP latent profile groups.

	Incomplete transfer vs. Better practice			Incomplete transfer vs. Average			Average vs. Better practice		
	OR	95% CI	*p*-Value	OR	95% CI	*p*-Value	OR	95% CI	*p*-Value
Female sex	1.13	0.74–1.72	0.58	0.87	0.57–1.32	0.51	1.29	0.88–1.91	0.19
Age	0.98	0.96–1.00	0.03	0.99	0.97–1.01	0.19	0.99	0.98–1.01	0.34
Ethnic minorities	3.46	2.23–5.36	<0.01	2.53	1.63–3.93	<0.01	1.37	0.89–2.10	0.15
Education									
No formal education	1 (ref)	–	–	1 (ref)	–	–	1 (ref)	–	–
Primary school	0.84	0.50–1.40	0.50	0.52	0.31–0.87	0.01	1.62	1.02–2.57	0.04
Middle school	0.37	0.17–0.82	0.01	0.51	0.24–1.10	0.09	0.73	0.35–1.50	0.39
High school and above	0.14	0.04–0.49	<0.01	0.84	0.31–2.26	0.73	0.17	0.05–0.53	<0.01
Marriage									
Unmarried	1 (ref)	–	–	1 (ref)	–	–	1 (ref)	–	–
Married	0.34	0.10–1.23	0.10	0.49	0.13–1.92	0.31	0.70	0.19–2.65	0.60
Divorced	0.57	0.15–2.25	0.42	0.40	0.09–1.76	0.23	1.42	0.34–5.88	0.63
Living arrangement									
Alone	1 (ref)	–	–	1 (ref)	–	–	1 (ref)	–	–
With spouse	2.60	0.76–8.97	0.13	2.30	0.62–8.46	0.21	1.13	0.35–3.65	0.83
With children	0.87	0.26–2.91	0.82	1.67	0.47–6.02	0.43	0.52	0.16–1.66	0.27
With other relatives	0.87	0.20–3.76	0.85	0.54	0.11–2.57	0.44	1.61	0.36–7.31	0.53
Annual income									
Low	1 (ref)	–	–	1 (ref)	–	–	1 (ref)	–	–
Moderate	0.50	0.31–0.80	<0.01	0.59	0.36–0.96	0.03	0.85	0.54–1.34	0.49
High	0.37	0.21–0.64	<0.01	0.56	0.32–0.97	0.04	0.65	0.38–1.11	0.12
Living expense									
Self-supplied	1 (ref)	–	–	1 (ref)	–	–	1 (ref)	–	–
Supplied by others	1.26	0.76–2.09	0.36	1.06	0.64–1.77	0.81	1.19	0.75–1.87	0.46
Annual out-of-pocket medical expense									
Affordable	1 (ref)	–	–	1 (ref)	–	–	1 (ref)	–	–
High	1.13	0.47–2.74	0.78	11.84	5.42–25.87	<0.01	0.10	0.05–0.17	<0.01
Catastrophic	0.11	0.02–0.58	0.01	1.88	0.71–4.97	0.20	0.06	0.01–0.29	<0.01
Self-reported hypertension	0.25	0.11–0.53	<0.01	1.10	0.47–2.61	0.82	0.22	0.10–0.49	<0.01
Self-reported diabetes	2.06	0.96–4.43	0.06	0.50	0.19–1.28	0.15	4.16	1.79–9.69	<0.01
Self-reported no chronic diseases	0.73	0.35–1.50	0.39	2.68	1.17–6.15	0.02	0.27	0.13–0.56	<0.01

Also, like the B.P. group, ethnic minorities were the dominant population in the A.V. group (68.8%). Furthermore, the A.V. group contained the largest proportions of married participants (84.2%), participants with no formal education (46.7%), and high annual out-of-pocket medical expense (33.3%).

Discussion

Management services for common chronic diseases have been incorporated into the national basic public health services, including individualized health education services. Through conversations with residents in ethnic minority areas, members of the general practitioner team and social workers generally felt a gulf to be crossed between these residents' health literacy and the available health education services. These analyses of residents' knowledge, attitudes, and practice about common chronic diseases may help understand their health needs and therefore influence how recipients and providers choose appropriate health education services. This choice may be interpreted by providers as relinquishing some of their health management responsibilities but is actually a prioritization of different needs (e.g., the I.T. profile, which requires behavioral management, and the B.P. profile, which lacks knowledge and beliefs). Without an adequate understanding of residents' health literacy and health needs, providers may be ill-prepared to understand residents' choices and help them manage their own health (21).

Research studies on health literacy levels of ethnic minorities have been conducted in Turkey, Sweden, and China (8, 22, 23), and these studies share the understanding that low levels of health literacy are evident in ethnic minorities; however, further individual differences lack relevant research work. This study refined the understanding of the health literacy of residents in ethnic minority areas as reflected in the focus of their KAP scores through the common chronic disease KAP questionnaire and subsequent latent profile analysis. We identified three groups of participants with similar KAP patterns (I.T., B.P., A.V.) and found that certain demographic and clinical characteristics appeared to be associated with group membership. Specifically, the I.T. group included more Han Chinese participants with higher annual incomes, suggesting that people of higher socioeconomic status may be more receptive to health knowledge but less able to translate that knowledge into practice. In contrast, the B.P. group included more minorities, less educated participants, fewer participants who suffered catastrophic medical costs, and more participants who self-reported having diabetes, suggesting that those who are more aware of their health conditions may be more concerned about improving their health practice, even if they are less knowledgeable and correspondingly less at risk for catastrophic medical expenses. In the case of the A.V. group, as the group description implies, participants in this group scored mediocre in all aspects, with demographic characteristics of being more ethnic minority, having lower education levels and lower annual income, and bearing more risk of higher medical expense compared to the I.T. group. There are no significant differences in demographic characteristics of the A.V. group compared to the B.P. group, except for higher education levels and medical expenses. Notably, the A.V. group had the highest self-reported rate of no chronic disease out of the three groups. These findings suggest that some minorities are likely to acquire knowledge from existing health education services, but the translation from health knowledge to health behaviors remains problematic. The differences in participant scores highlight important differences in knowledge, attitudes, and practice concerning common chronic diseases among participants and indicate their essential health needs. The KAP latent profile groups underscore a key point: there is no single correct pattern of health education services for populations with different health needs.

Through previous research (24), we have learned that health inequities exist in health education and health promotion. Ethnic minorities are disadvantaged in many countries regarding accessibility and utilization of health education services (25–27). The question is whether this disadvantage is followed by different unsatisfied health needs, which is confirmed by the results of the present study, in which we selected a typical ethnic minority area as the study site. After analyzing residents' knowledge, attitudes, and practice regarding common chronic diseases, we identified three KAP latent profile groups and the different health needs underlying them. First, there is a clear knowledge-practice gap in the I.T. group, and applying the behavioral economics framework to the field of health education can reveal potential barriers to translating knowledge into practice and entry points for interventions by clinicians and public health professionals (28); furthermore, the development of mobile health technologies, especially the availability of wearable devices and 5G/6G wireless technologies, has made real-time monitoring and telehealth possible (29), facilitating the implementation of behavioral interventions. Second, we argue that the same ethnic minorities still have different health needs; for example, the B.P. group may need more culturally sensitive health knowledge, while the A.V. group has a more substantial need to address the translation of knowledge to practice. These findings may provide novel ideas for future health education work: precise identification of needs followed by corresponding precise interventions (30).

The purpose of this study was not to reaffirm the inadequate health literacy among ethnic minorities; the knowledge, attitudes, and practice about common chronic diseases that we investigated are important components of health literacy but do not fully represent health literacy that is still evolving (31). Instead, the findings reported here present an advance in the study of health literacy among ethnic minorities; the health needs of ethnic minorities should be well understood before we conduct research on health literacy interventions. Some evidence suggests that model-based health literacy assessments or interventions can provide a better picture of these needs (32, 33).

A limitation of this study is that the differences in KAP for chronic diseases among different ethnic minorities are not adequately discussed. This present study focuses on the differences between chronic disease knowledge, attitudes, and practice between Han and ethnic minorities; however, as we mentioned, there are 55 different ethnic minorities in China who may have different levels of acceptance of generalized health education due to their customs, language and script, degree of Hanization (assimilated by Han Chinese), etc., and thus differences between KAP for chronic diseases. However, in our study population, the ethnic minority study population was predominantly Lahu (447 of 496), so this variation in chronic disease KAP among different ethnic minorities was not influential on the overall results. In addition, our search of previous literature revealed some commonalities across ethnic minorities in Yunnan: high illiteracy rates, low awareness of their health concerns, and high tobacco and alcohol consumption (34, 35). Another limitation is that we used self-reported prevalence of chronic diseases. Among the four common chronic diseases surveyed, no one reported prevalence of two chronic diseases (chronic obstructive pulmonary disease and obesity), which is inconsistent with the facts we learned from local CDC and hospitals, and the existence of reporting bias should not be ignored. We will consider this in the intervention design phase of the upcoming health literacy intervention study.

Conclusion

Having a more specific and nuanced understanding of ethnic minority health literacy allows providers to conduct more effective health education with their recipients. Given the differences in health needs, this will naturally lead to different offerings. Providers can do more to encourage recipients to be proactive in managing their own health and to take responsibility for maintaining it. Then, professionals use their specialization to help recipients make health decisions, rather than mandating things or instilling expertise that is difficult to grasp. This form of health education activity builds better relationships between providers and recipients and permits professionals to engage in this work in an empowering way, thereby improving health inequities.

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 Lincang Second People's Hospital. The patients/participants provided their written informed consent to participate in this study.

Author contributions

HH, YX, YS, and YL designed the study. YS and YL cleaned and processed the data. HH wrote the manuscript. YX and CL revised the manuscript. All authors contributed to the subsequent drafts, reviewed, and endorsed the final submission.

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

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

References

1. Raghupathi W, Raghupathi V. An empirical study of chronic diseases in the United States: a visual analytics approach. *Int J Environ Res Public Health.* (2018) 15:431. doi: 10.3390/ijerph15030431

2. Heine M, Lategan F, Erasmus M, Lombaard CM, Mc Carthy N, Olivier J, et al. Health education interventions to promote health literacy in adults with selected non-communicable diseases living in low-to-middle income countries: a systematic review and meta-analysis. *J Eval Clin Pract.* (2021) 27:1417–28. doi: 10.1111/jep.13554

3. The L. The health illiteracy problem in the USA. *Lancet.* (2009) 374:2028. doi: 10.1016/S0140-6736(09)62137-1

4. World Health Organization. *Policy Brief 4: Health Literacy*. Geneva: World Health Organization (2016).

5. Griffin G, Nau SZ, Ali M, Riggs E, Dantas JAR. Seeking health information: a qualitative study of the experiences of women of refugee background from Myanmar in Perth, Western Australia. *Int J Environ Res Public Health.* (2022) 19:3289. doi: 10.3390/ijerph19063289

6. Poureslami I, Nimmon L, Rootman I, Fitzgerald MJ. Health literacy and chronic disease management: drawing from expert knowledge to set an agenda. *Health Promot Int.* (2017) 32:743–54. doi: 10.1093/heapro/daw003

7. Van der Heide I, Poureslami I, Mitic W, Shum J, Rootman I, FitzGerald JM. Health literacy in chronic disease management: a matter of interaction. *J Clin Epidemiol.* (2018) 102:134–8. doi: 10.1016/j.jclinepi.2018.05.010

8. Bergman L, Nilsson U, Dahlberg K, Jaensson M, Wångdahl J. Health literacy and e-health literacy among Arabic-speaking migrants in Sweden: a cross-sectional study. *BMC Public Health.* (2021) 21:2165. doi: 10.1186/s12889-021-12187-5

9. Cheng L, Chen Q, Zhang FY, Wu W, Cui W, Hu X. Functional health literacy among left-behind students in senior high schools in an ethnic minority area: a cross-sectional study. *Medicine.* (2020) 99:e19167. doi: 10.1097/MD.0000000000019167

10. Chui CH, Arat G, Chan K, Wong PWC. Growing old as a member of an ethnic minority in Hong Kong: implications for an inclusive long-term care policy framework. *J Appl Gerontol.* (2020) 39:463–71. doi: 10.1177/0733464819873504

11. Koohpayeh SA, Estebsari F, Lashani F, Homayouni A, Nasiri M. Relationship between health literacy and knowledge, attitude, and performance regarding cancer warning signs. *Iran J Nurs Midwifery Res.* (2021) 26:399–405. doi: 10.4103/ijnmr.IJNMR_359_20

12. Mitchell KE, Johnson-Warrington V, Apps LD, Bankart J, Sewell L, Williams JE, et al. A self-management programme for COPD: a randomised controlled trial. *Eur Respir J.* (2014) 44:1538–47. doi: 10.1183/09031936.00047814

13. Shiha G, Metwally AM, Soliman R, Elbasiony M, Mikhail NNH, Easterbrook P. An educate, test, and treat programme towards elimination of hepatitis C infection in Egypt: a community-based demonstration project. *Lancet Gastroenterol Hepatol.* (2018) 3:778–89. doi: 10.1016/S2468-1253(18)30139-0

14. Pinnock H, Hanley J, McCloughan L, Todd A, Krishan A, Lewis S, et al. Effectiveness of telemonitoring integrated into existing clinical services

on hospital admission for exacerbation of chronic obstructive pulmonary disease: researcher blind, multicentre, randomised controlled trial. *BMJ.* (2013) 347:f6070. doi: 10.1136/bmj.f6070

15. Zhang ZB, Xue ZX, Ma MM Li YH, Luo DM, Song XL, et al. [Knowledge, attitude and practice to chronic diseases and associated influencing factors in Uygur population in Kashgar area of Xinjiang Uygur Autonomous Region]. *Zhonghua Liu Xing Bing Xue Za Zhi.* (2017) 38:715–20. doi: 10.3760/cma.j.issn.0254-6450.2017.06.005

16. Freeman V, Fraser H, Forrester T, Wilks R, Cruickshank J, Rotimi C, et al. A comparative study of hypertension prevalence, awareness, treatment and control rates in St Lucia, Jamaica and Barbados. *J Hypertens.* (1996) 14:495–501. doi: 10.1097/00004872-199604000-00013

17. Xue B, Zhang X, Li T, Gu Y, Wang R, Chen W, et al. Knowledge, attitude, and practice of obesity among university students. *Ann Palliat Med.* (2021) 10:4539–46. doi: 10.21037/apm-21-573

18. White R, Walker P, Roberts S, Kalisky S, White P. Bristol COPD Knowledge Questionnaire (BCKQ): testing what we teach patients about COPD. *Chron Respir Dis.* (2006) 3:123–31. doi: 10.1191/1479972306cd117oa

19. Kheir N, Greer W, Yousif A, Al Geed H, Al Okkah R. Knowledge, attitude and practices of Qatari patients with type 2 diabetes mellitus. *Int J Pharm Pract.* (2011) 19:185–91. doi: 10.1111/j.2042-7174.2011.00118.x

20. R Core Team. *R: A Language and Environment for Statistical Computing.* Vienna: R Foundation for Statistical Computing (2021). Available online at: https://www.R-project.org/

21. Institute of Medicine Committee on Assuring the Health of the Public in the 21st C. *The Future of the Public's Health in the 21st Century*. Washington, DC: National Academies Press (US) Copyright 2003 by the National Academy of Sciences. All rights reserved (2002).

22. Van der Gaag M, van der Heide I, Spreeuwenberg PMM, Brabers AEM, Rademakers JJDJM. Health literacy and primary health care use of ethnic minorities in the Netherlands. *BMC Health Serv Res.* (2017) 17:350. doi: 10.1186/s12913-017-2276-2

23. Wang C, Li H, Li L, Xu D, Kane RL, Meng Q. Health literacy and ethnic disparities in health-related quality of life among rural women: results from a Chinese poor minority area. *Health Qual Life Outcomes.* (2013) 11:153. doi: 10.1186/1477-7525-11-153

24. Deravin L, Francis K, Anderson J. Closing the gap in Indigenous health inequity - Is it making a difference? *Int Nurs Rev.* (2018) 65:477–83. doi: 10.1111/inr.12436

25. Kirksey L, Tucker DL, Taylor E. Jr., White Solaru KT, Modlin CS Jr. Pandemic superimposed on epidemic: Covid-19 disparities in black Americans. *J Natl Med Assoc.* (2021) 113:39–42. doi: 10.1016/j.jnma.2020.07.003

26. Fan Q, DuPont-Reyes MJ, Hossain MM, Chen LS, Lueck J. Ma P. Racial and ethnic differences in major depressive episode, severe role impairment, and mental health service utilization in US adolescents. *J Affect Disord.* (2022) 306:190–9. doi: 10.1016/j.jad.2022.03.015

27. Vandan N, Wong JY, Fong DY. Accessing health care: experiences of South Asian ethnic minority women in Hong Kong. *Nurs Health Sci.* (2019) 21:93–101. doi: 10.1111/nhs.12564

28. Saleska JL, Choi KR, A. behavioral economics perspective on the COVID-19 vaccine amid public mistrust. *Transl Behav Med.* (2021) 11:821–5. doi: 10.1093/tbm/ibaa147

29. Vyas S, Bhargava D, Bhola J, Ujjan JA, Eswaran S, Rahmani AW. Critical retrospection of performance of emerging mobile technologies in health data management. *J Healthc Eng.* (2022) 2022:8903604. doi: 10.1155/2022/8903604

30. Hu H, Liu J, Liang Y, Xu Y. Health literacy of ethnic minority in remote China: precise intervention is needed. *Lancet Reg Health West Pac.* (2021) 17:100322. doi: 10.1016/j.lanwpc.2021.100322

31. Santana S, Brach C, Harris L, Ochiai E, Blakey C, Bevington F, et al. Updating health literacy for healthy people 2030: defining its importance for a new decade in public health. *J Public Health Manag Pract.* (2021) 27:S258–S64. doi: 10.1097/PHH.0000000000001324

32. Handley MA, Harleman E, Gonzalez-Mendez E, Stotland NE, Althavale P, Fisher L, et al. Applying the COM-B model to creation of an IT-enabled health coaching and resource linkage program for low-income Latina moms with recent gestational diabetes: the STAR MAMA program. *Implement Sci.* (2016) 11:73. doi: 10.1186/s13012-016-0426-2

33. Zhang Q, Jia W, Zhang J, Zhan H. Effect of health literacy interventions with the stages of change on Kazakh-Chinese hypertension patients. *Chin J Behav Med Brain Sci.* (2019) 3:259–64. doi: 10.3760/CMA.J.ISSN.1674-6554.2019.03.013

34. Le C, Jun D, Zhankun S, Yichun L, Jie T. Socioeconomic differences in diabetes prevalence, awareness, and treatment in rural southwest China. *Trop Med Int Health.* (2011) 16:1070–6. doi: 10.1111/j.1365-3156.2011.02805.x

35. Wan S, Tang Q, Feng D, Hu Z, Shao W, Chen Y. Epidemiology characteristics of ethnic minority colorectal cancer in Yunnan in Southwestern China. *Transl Cancer Res.* (2020) 9:2692–700. doi: 10.21037/tcr.2020.02.63

frontiers | Frontiers in Public Health

TYPE Original Research
PUBLISHED 23 August 2022
DOI 10.3389/fpubh.2022.919386

Development and validation of cultural competence assessment tool for healthcare professionals, India

Parvathy Balachandran, Vineetha Karuveettil and Chandrashekar Janakiram*

Department of Public Health Dentistry, Amrita School of Dentistry, Amrita Vishwa Vidyapeetham, Kochi, India

OPEN ACCESS

EDITED BY
Srinivas Pachava,
SIBAR Institute of Dental Sciences, India

REVIEWED BY
Suresh Chand Yaddanapalli,
SIBAR Institute of Dental Sciences, India
Nijampatnam P. M. Pavani,
SIBAR Institute of Dental Sciences, India

*CORRESPONDENCE
Chandrashekar Janakiram
sekarcandra@gmail.com

SPECIALTY SECTION
This article was submitted to Family Medicine and Primary Care, a section of the journal Frontiers in Public Health

RECEIVED 13 April 2022
ACCEPTED 27 July 2022
PUBLISHED 23 August 2022

CITATION
Balachandran P, Karuveettil V and Janakiram C (2022) Development and validation of cultural competence assessment tool for healthcare professionals, India. *Front. Public Health* 10:919386. doi: 10.3389/fpubh.2022.919386

COPYRIGHT
© 2022 Balachandran, Karuveettil and Janakiram. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

Culture influences an individual's perception of "health" and "sickness". Therefore, cultural competence assessment of healthcare professionals is very important. Existing assessment scales have limited application in India due to the nation's rich cultural diversity and heterogeneous healthcare streams. This study was undertaken to develop and validate a cultural competence assessment tool for healthcare professionals in India. A cross-sectional study using convenience sampling was conducted following all standard steps among 290 healthcare professionals in India. Item reduction was followed by estimation of validity and reliability. Responses were recorded on a five-point Likert scale, ranging from strongly disagree to strongly agree. The resultant tool, named Cultural Competence Assessment Tool–India (CCT-I) showed an acceptable internal consistency (Cronbach's alpha =0.734). Inter-rater agreement was 81.43%. Face, content, and construct validity were demonstrated. There was no statistically significant difference in cultural competence between the healthcare streams based on years of clinical experience. There was statistically significant difference between streams of healthcare (p-value =0.009) and also between dentistry and Ayurveda groups (p-value = 0.003). This comprehensive tool can be used as the first step toward designing cultural competence training of healthcare manpower and the establishment of culturally sensitive healthcare organizations.

KEYWORDS

cultural competency, cultural diversity, India, health personnel, healthcare system, patient care

Introduction

Health equity is the cornerstone of community-focused health interventions and aims to improve the wellbeing of each individual in the community. However, achieving health equity is a challenging process as it comprises complex interactions among healthcare demand, supply, accessibility, and utilization, which, in turn, depend on the social determinants of health. These are conditions in places where people live, learn, work, and play that affect a wide range of health and quality-of-life risks and outcomes (1) like social norms, education, job, income, and gender roles, which widen the health disparities in the community.

Among the social determinants of health, culture is the most important yet often neglected factor in healthcare. Culture refers to "integrated patterns of human behavior that include language, thoughts, communications, actions, customs, beliefs, values, and institutions of racial, ethnic, religious, or social groups" (2). It is a fundamental characteristic of a community that has a deep-rooted impact on the life of an individual, his/her belief systems, and attitude, including one's perception of "health" and "sickness". Culture determines the presentation and interpretation of disease symptoms, health-seeking behavior, and treatment outcomes. Therefore, a healthcare system that understands and accepts the existence of different cultural groups among healthcare seekers is a huge step toward a successful health service delivery system (3). It also highlights the importance of cultural competence development of healthcare manpower.

Competence implies "having the capacity to function effectively" (4). Cultural competence is "a set of congruent behaviors, attitudes, and policies that come together in a system, agency, or among professionals and enable that system, agency, or those professionals to work effectively in cross-cultural situations" (4). This description of cultural competence gives insights into a culturally competent healthcare system, that is, "one that acknowledges and incorporates—at all levels—the importance of culture, assessment of cross-cultural relations, vigilance toward the dynamics that result from cultural differences, expansion of cultural knowledge, and adaptation of services to meet culturally unique needs" (5).

This dimension of healthcare is highly relevant in the current COVID-19 pandemic, where the literature shows that cultural diversity is an important challenge to equitable distribution of healthcare services and accessibility (6). Efforts to enhance cultural competence in the healthcare system have made a significant impact in the United States, like the Initiative to Eliminate Racial and Ethnic Disparities in Health, which implemented the National Culturally and Linguistically Appropriate Services Standards and delivered culturally appropriate influenza immunization in addition to establishing grants and community networks program centers to reduce cultural disparities in healthcare (7).

Cultural competence enables a healthcare provider to go beyond the pathophysiological knowledge of disease (8). A culturally competent healthcare worker understands patient's perspective of health and illness, has improved healthcare provider–seeker interaction, overcomes language barriers, and increases the quality of care, thereby resulting in a positive treatment outcome (9). Studies have shown that culturally relevant interventions improved health outcomes related to sexually transmitted diseases, type II diabetes (10, 11), and drug addiction (12). Similarly, culturally sensitive healthcare services like use of bilingual community health workers have improved culturally diverse patients" acceptance of cancer screening and health monitoring (13).

"Culture" being a subjective concept is often used synonymously with socioeconomic status, leading to underestimation of the role of culture in a person's life (5). Another challenge, particularly in a vast and diverse nation like India, is the existence of many cultures along with their equally numerous subcultures. Each culture and subculture is unique, and a broad stereotyping of patients by preconceived notions can result in unintentional harms. Scarcity of time and resources, reluctance, or failed efforts in recognizing the cultural impacts on health, and incompetent leadership to highlight the importance of culture are other limitations of cultural competence in healthcare.

Cultural competence development of healthcare professionals is an issue of prime importance. It has gained priority in Western countries; however, it is still an unexplored area in the Indian setting. The first step toward the development of this skill is its assessment. Although there are many cultural competence assessment tools available globally, applicability of such tools varies widely based on the cultural environment of each country. In addition, India has multiple healthcare streams, like allopathy, Ayurveda, homeopathy, Siddha, Unani, and naturopathy, where the perspective of disease, its causes, and treatment approaches are highly heterogeneous. Therefore, to assess the cultural competence of healthcare professionals in India, there is a necessity for an assessment tool that applies to the healthcare system, which is influenced by a multitude of cultures, belief systems, healthcare streams, and social norms. Currently, there is no available assessment tool that is specifically adapted to the Indian setting. The present study was therefore designed to address this significant gap in the literature by developing a cultural competence assessment scale customized for Indian healthcare professionals.

Methods

This cross-sectional study using convenience sampling for selection of participants was undertaken from March 2021 to October 2021. Prior to the conduct of the study, ethical approval was obtained from the Institutional Ethics Committee of Amrita Institute of Medical Sciences, India (ECASM-AIMS-2021-171, date: 23-02-2021). Due to the COVID-19 pandemic, communications related to the study were undertaken through email and online platforms using Google Forms. Description of the study was provided to all stakeholders through email and Google Forms, and informed consent was obtained from them in the same manner.

The proposed cultural competence assessment scale for Indian healthcare professionals was to be developed in the form of a questionnaire with the following features:

- It has the ability to measure cultural competence through participants" responses to the questions.
- Being a novel instrument, it can assess the cultural competence level of healthcare professionals from different healthcare streams in India, a unique feature of the proposed scale as the various healthcare streams often have conflicting theories and approaches. We focused on the patient–caregiver interaction, which is the most important aspect of any treatment, irrespective of the healthcare stream.
- It includes nursing professionals of different healthcare streams as they play a profound role.
- It comprises questions that cover the entire spectrum of cultural competence.
- It is amenable to statistical analysis.
- It has good psychometric properties with satisfactory reliability and validity.

Only healthcare professionals belonging to medical, dental, Ayurveda, nursing, and homeopathy streams, with at least 3 years of clinical experience after their graduation, were included in the study. The nursing group also included dental assistants, Ayurveda and homeopathic nurses, and those with auxiliary nurse midwife (ANM) and general nursing and midwifery (GNM) qualifications. Eligible healthcare professionals who were unwilling to participate were excluded from the study. Healthcare graduates who had changed their careers to other streams, like bioinformatics and insurance sectors, were also excluded from the study.

Questionnaire development was performed in three phases:

- Phase of item development involving the identification of domains, item generation, and content validation;
- Phase of scale development involving pretesting the developed questionnaire using cognitive interviews and exploratory factor analysis;
- Phase of scale evaluation consisting of tests of reliability and validity.

Domains were identified and developed by literature review. The distinction between the domains was ensured by expert validation. A pilot version of the questionnaire with 159 items was developed using a deductive approach through extensive literature review. After the removal of overlapping and redundant items, it had seven domains and 43 items in total. For validation of domains and items, two Google Forms were designed: one for domains and the other for items.

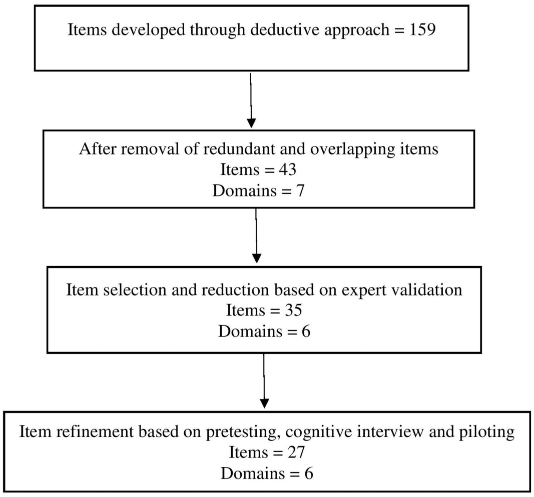


FIGURE 1
Item generation and item reduction.

The degree of relevance of each domain and item to measure the cultural competence of healthcare professionals was recorded by a three-point Likert scale and four-point Likert scale, respectively. Google Forms also comprised the participant information sheet and certificate of consent. These Google Forms were administered to an expert committee of five members. Based on their responses, kappa scores of agreements were calculated. The resultant questionnaire comprised six domains and 35 items.

Cognitive interviews were carried out among 10 participants to ensure the quality and accuracy of the questionnaire and to identify sources of response errors before administering it to final users. We used the "think aloud" approach for five participants and the "verbal probing approach" for five participants. The tool was then piloted on a sample of 30 participants. Internal consistency of the cultural competence questionnaire was determined using Cronbach's alpha. Test–retest reliability was assessed among the 30 participants after 2 weeks by using the intraclass correlation coefficient. Based on the test–retest reliability score, eight items were eliminated. The resultant tool had six domains and 27 items (Figure 1).

The sample size for scale development is based on the rule of thumb of at least 10 participants for each scale item, with a 10:1 respondent-to-item ratio (14). Therefore, the minimum sample size of the pilot study was estimated to be 270, with at least 54 participants from each healthcare stream to ensure uniform participation. The content validity index (CVI) is the most commonly used content validity approach. In estimating the scale-level-CVI, two methods were employed: the average-CVI

(S-CVI/Ave) and universal agreement (UA) among experts (S-CVI/UA).

The reliability of inter-rater agreement was estimated by using Fleiss kappa. Known group validity was assessed by comparing scores among different healthcare streams. Based on the responses, construct validity was assessed using exploratory factor analysis (EFA). This step was carried out to reduce the number of items and to ensure that the developed tool successfully measured the cultural competence of healthcare professionals. Based on the EFA, one item was removed. Following this, the floor and ceiling effects were determined.

The final tool to assess the cultural competence of healthcare professionals in India was named Cultural Competence assessment Tool–India (CCT-I). It had six domains and 26 items (Figure 2). Participants' responses were recorded on a five-point Likert scale with the following scoring criteria: 1= strongly disagree, 2= disagree, 3= neutral, 4= agree, and 5=strongly disagree. To minimize social desirability bias, six items were negative worded, and hence, their scores were inversed during analysis. Standardization of scores was based on percentile rank of scores. Statistical analysis was performed using IBM SPSS Statistics for Windows, version 23 (IBM Corp., Armonk, N.Y., USA).

Results

Sample description

A total of 290 participants completed the survey, with a response rate of 86.05%. The mean age of the participants was 35.09 ± 9.85 years. A maximum number of the participants were from nursing ($n = 67$), followed by dentistry ($n = 58$), homeopathy ($n = 56$), Ayurveda ($n = 55$), and medicine ($n = 54$). It was found that the majority of participants had <5 years of clinical experience (35.9%) (Table 1). The Shapiro–Wilk test was used to test normality as the sample size was more than 50. Since the data were skewed, median and interquartile range were estimated.

For the domain of cultural awareness, the median and interquartile range is 25 (22–27). The median and interquartile range of the cultural sensitivity domain is reported to be 12 (11–14), 14 (13–16) for the cultural knowledge domain, 10 (9–11) for the cultural skills domain, 14 (13–15) for the cultural behavior domain, and 16 (14–16) for cultural desire domain. The overall median and interquartile range of the cultural competence scale is 91 (84–96) (Supplementary Table 1).

Floor and ceiling effect calculation

The floor and ceiling effects for the cultural competence scale were satisfactory. The overall floor effect was 11.38%, and the ceiling effect was 12.41%. The floor effect for each domain ranged from 7.58 to 14.14%, and the ceiling effect ranged from 5.52 to 24.48%. When considering individual domains, except for cultural knowledge, which had a ceiling effect of 24.8%, other domains maintained the required scores (Supplementary Table 2).

Reliability estimates

Reliability analyses were performed on the 35-item questionnaire in phase I (Supplementary Table 3) and the 27-item questionnaire in phase II. Test–retest reliability after item reduction was measured for 27 items. The overall intraclass correlation coefficient was estimated as 0.767, indicating satisfactory stability (Supplementary Table 4). Item-wise test–retest reliability was also performed, and the intraclass correlation coefficient ranged between 0.566 and 0.822. Internal consistency of the 35-item questionnaire was estimated using Cronbach's alpha (Supplementary Table 5), and eight items were removed. Overall internal consistency using Cronbach's alpha for the 27-item questionnaire was estimated to be 0.700, which was acceptable (Supplementary Table 6). After removal of one item with a lower eigenvalue, overall internal consistency improved to 0.734, which was acceptable (Supplementary Table 7).

Validity estimates

The face validity and content validity of the proposed tool were determined by assessing its content during the expert reviews (Supplementary Tables 8, 9) and peer reviews in the phase of item development. After the removal of one domain (domain 7) and two items, for domains, S-CVI/Ave was 0.92 and S-CVI/UA was 0.5. Fleiss kappa for domains yielded a fair score of 0.67, and the overall agreement was 83.30% (Supplementary Table 10). For items, the average-CVI (S-CVI/Ave) was estimated to be 0.87 and S-CVI/UA was 0.58. Fleiss kappa was found to be 0.25, and the score-combined kappa was 0.72. Agreement among the raters was 81.43% (Supplementary Table 11).

Construct validity was established by using exploratory factor analysis (EFA) (Supplementary Table 12) with principal component analysis (PCA) and varimax rotation in the phase of scale evaluation. Initial factor extraction with PCA yielded 49.35% as cumulative percentage variance explained by the tool (Supplementary Table 13). Minimum loading for an item with a factor is expected to be 0.35 (15). Then, one item was found to have a factor loading <0.35 and was removed from the tool; six items showed a cross-loading effect. However, these items were relevant to the tool in the assessment and were therefore retained. Factor analysis was performed for the remaining 26

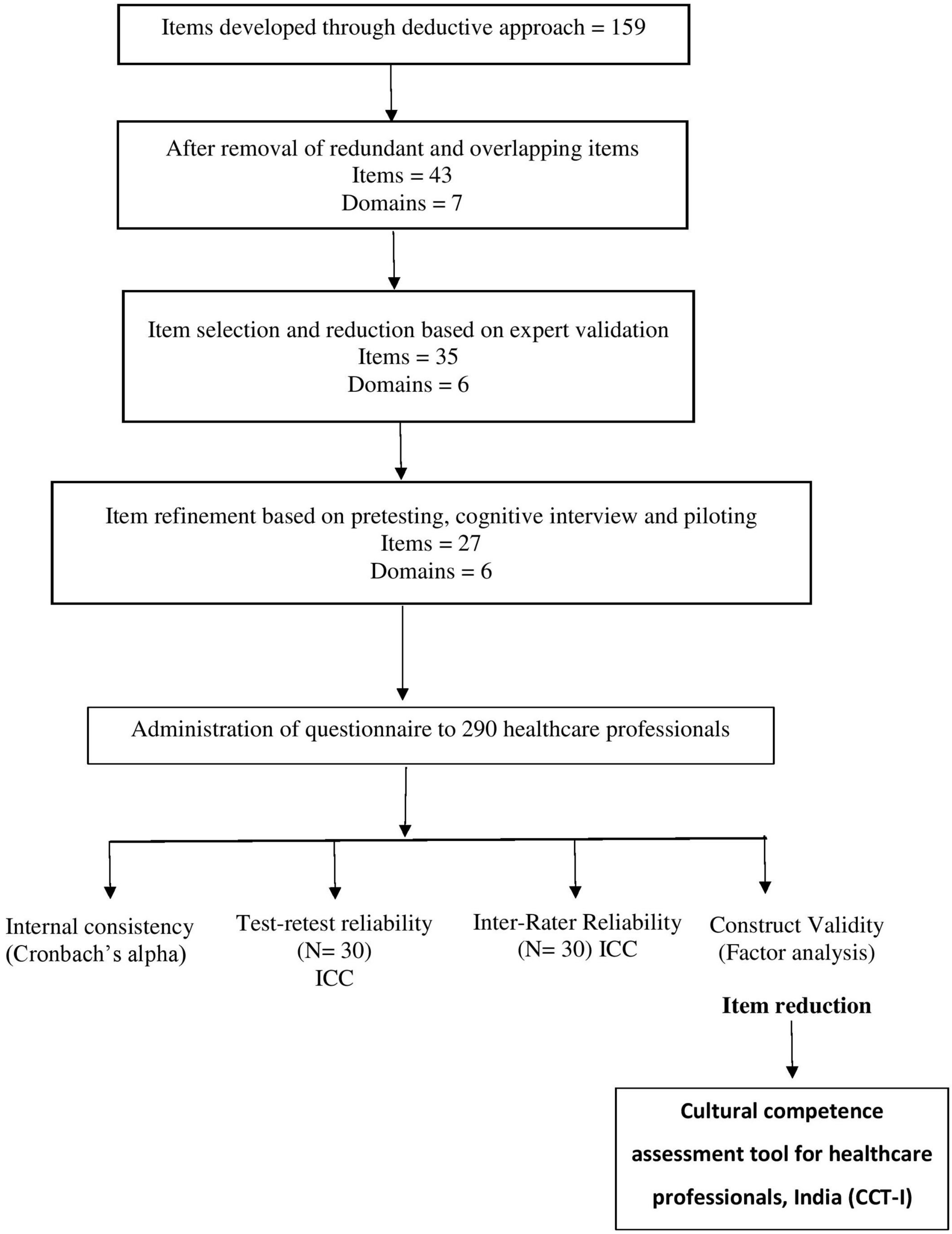


FIGURE 2
Flow chart showing the steps in the development of the new tool.

items. The cumulative percentage variance explained by the tool improved to 50.36% (Supplementary Table 14).

The final factor analysis resulted in six domains, with a total of 26 items being established. The absolute loading value gives the relationship between the item and the concept of cultural competence. Only one item showed a low loading value, and six items showed a cross-loading effect. Since the items were relevant for the assessment of cultural competence, these items were retained in the tool.

TABLE 1 Characteristics of the study population.

Parameters	Result
Age in years	35.09 ± 9.85 years
Healthcare system	
Medicine	54 (18.6%)
Dentistry	58 (20%)
Ayurveda	55 (19%)
Homeopathy	56 (19.3%)
Nursing	67 (23.1%)
Years of clinical experience	
3– 5 years	104 (35.9%)
5– 10 years	81 (27.9%)
10–15 years	48 (16.6%)
15–20 years	12 (4.1%)
20–25 years	16 (5.5%)
More than 25 years	29 (10.0%)

Known group validity was estimated using the Kruskal–Wallis test for years of experience and stream of healthcare. There was no statistically significant difference between the groups for years of experience (Table 2A). However, there was statistically significant difference between the groups in relation to the stream of healthcare (p-value = 0.009). Mean and standard deviation was highest for the Ayurveda group (93.38 ± 8.39), followed by homeopathy, medicine, nursing, and dentistry, respectively (Table 2B). The difference in cultural competence between dentistry and Ayurveda groups was found to be statistically significant (p-value = 0.003) (Table 3).

Standardization of scores

Cultural competence scores of the range 26 to 84 were considered as low (0 to 24th percentile). Scores from 85 to 96 were considered as average cultural competence (25th to 75th percentile), and scores from 97 to 130 were considered as high cultural competence (76th to 100th percentile).

Table 2A Known group validity. Known group validity based on years of experience: Kruskal–Wallis test.

Years of experience	Mean ± SD	Median	IQR			Chi-Square	df	Sig.
			Q1	Q2	Q3			
<5 years	88.63 ± 8.74	88	82	88	95	6.96	5	0.223
5–10 years	90.96 ± 7.75	92	84.5	92	96			
10–15 years	92.17 ± 7.13	92.5	88.25	92.5	97.75			
15–20 years	92.00 ± 7.75	90	86	90	99			
20–25 years	91.88 ± 12.76	92	85	92	96.75			
More than 25 years	89.97 ± 7.78	91	82.5	91	95.50			

p-value ≤0.05 is considered statistically significant.
IQR, interquartile range.

Table 2B Known group validity based on healthcare streams.

Stream	Mean ± SD	Median	IQR			Chi-Square	df	Sig.
			Q1	Q2	Q3			
Medicine	90.15 ± 7.53	91.5	85	91.5	95	13.61	4	0.009*
Dentistry	87.28 ± 7.70	86.5	81	86.5	94			
Ayurveda	93.38 ± 8.39	92	88	92	99			
Homeopathy	91.23 ± 8.19	91	84.25	91	96.75			
Nursing	89.82 ± 9.07	90	83	90	96			

*p-value ≤ 0.05.

Table 3 Known group validity based on healthcare streams.

Stream	Median 1	Median 2	Sig.
Dentistry-medicine	86.5	91.5	0.632
Dentistry-nursing	86.5	90	0.939
Dentistry-homeopathy	86.5	91	0.158
Dentistry-ayurveda	86.5	92	0.003*
Medicine-nursing	91.5	90	1.000
Medicine-homeopathy	91.5	91	1.000
Medicine-ayurveda	91.5	92	0.926
Nursing-homeopathy	90	91	1.000
Nursing-ayurveda	90	92	0.403
Homeopathy-ayurveda	91	92	1.000

*p-value $\leq$ 0.05.

Discussion

This article reports on the field test of the CCT-I as part of the instrument development process. Existing cultural competence assessment scales had limited applications in the Indian setting due to the country's diversity of cultures and healing systems. Moreover, existing scales varied widely in their interpretation of "culture," "cultural competence," and therefore the assessments (15). Most of these tools were group-specific, focusing only on nurses, pharmacists, and dentists, thereby curtailing their application in the comprehensive assessment of cultural competence at the organization or national level (16). On this account, there was a compelling need to develop and validate a novel assessment tool that is focused on assessing the wide spectrum of cross-cultural competence, irrespective of the healthcare stream.

The present study was performed in accordance with the standard steps of scale development and validation (14) and the COnsensus-based Standards for the selection of health status Measurement INstruments (COSMIN) (17). The proposed scale was developed as a self-reported assessment tool since it is more appropriate for expressing one's attitude, beliefs, and behaviors. Although there are drawbacks like social desirability bias, response bias, and lack of opportunity to clarify the respondent's doubts, the current tool was designed as a self-reporting questionnaire because respondents were accustomed to the issues in question and the information they give in self-report questionnaires tends to be more accurate (18).

Domains and items constituting the CCT-I scale were derived by literature review and consensus. Domains were validated by a committee of six experts and items by a committee of five experts. For a scale to be considered as having excellent content validity, all its items should have an I-CVI score of 1 (19). As reported, the I-CVI was acceptable after elimination of two items; four items which showed low I-CVI scores of 0.6 were revised. The remaining items showed scores ranging from 0.80 to 1. Such modifications were made in similar studies like the development of an instrument to measure patient-centered communication (20). In this study, items that scored below 0.7 were eliminated and that showed scores between 0.7 and 0.79 were revised. This shows that all items in the tool were conceptually relevant and appropriate to assess the cultural competence of healthcare professionals in India.

In this study, S-CVI/UA was 0.43 for domains and 0.47 for items. S-CVI/Ave for domains was 0.86 for domains and 0.67 for items. After the removal of one domain (domain 7) and two items (items 10 and 37), S-CVI/UA for domains was found to be 0.5 for domains and 0.58 for items. S-CVI/Ave improved to 0.92 for domains and 0.87 for items. S-CVI/Ave $>$0.90 denotes excellent score for scale-level content validity (19). According to Lynn's criteria for item acceptability, excellent content validity is characterized by I-CVIs of 0.78 or higher (21). Similarly, the S-CVI/UA value of 0.8, as per the conservative requirement of 100% agreement at the item level for at least 80% of items (22–24), and S-CVI/Ave value of 0.9 or higher (25), denote excellent content validity. These benchmarks show that the CCT-I scale has satisfactory content validity.

Chance agreement is an issue of concern in validation by assessors (26); hence, kappa statistics was also computed. The combined kappa value for the developed tool was 0.72, thereby indicating a good score (20). Overall percentage agreement among the expert judges improved to 83.30% for domains and 81.43% for items. This is in accordance with recommendations that an agreement of 80% or higher is considered ideal for tool development (20).

The next step in tool development was pretesting the tool *via* cognitive interviews to ensure that the target population clearly understands the domains and items (14). Think aloud is a method of cognitive interview in which the participants are given the opportunity to verbalize their thought process as they answer the items. Verbal probing is the alternative procedure of cognitive interview, where the interviewer probes the interviewee with additional questions to elicit further information on the items of the tool (27). Since both are unique in their approach and technique, we conducted cognitive interviews for five participants using the think aloud process and another five participants using the verbal probing method.

Typographical and grammatical errors were identified through cognitive interviews. It was recommended to revise certain items to make the assessment tool suited for the Indian scenario. A change in the rating of the Likert scale was suggested by three interviewees due to the difficulty in differentiating between strongly agree and agree and likewise between strongly disagree and disagree. However, we did not change the five-point Likert scale as it was befitting our questionnaire. Another suggestion was to rephrase some of the items to reduce possible social desirability bias. To resolve this issue, some of the items were negative worded, and hence, their scores were inversed during analysis. There was an overall consensus on the length of the questionnaire and time taken to answer.

Another component of content validation is the identification of floor and ceiling (F/C) effects. The floor effect implies that the items are hard to understand, while the ceiling effect means that items are easy to understand. In some studies (28, 29), 5 or 10% is considered the benchmark for the F/C effect. However, in the majority of studies, a score of ≥15% is considered to have a significant F/C effect. We also followed the 15% criterion in this study. The domain corresponding to cultural knowledge showed a ceiling effect of 24.48%. However, the overall F/C effect score of the developed tool was 11.38 and 12.41%, respectively, which was acceptable.

We used test–retest reliability and Cronbach's alpha to determine the reliability of the tool. For the test–retest reliability (coefficient of stability) approach, the assessment tool was administered to 30 participants as Google Forms contained 35 items. The suggested gap between the test and retest is 2 weeks (30), which was followed in this study. The intraclass correlation coefficient was used to determine the reliability of the scale. Items that showed values closer to 0 indicated low reliability (14). Internal consistency of the developed tool was assessed by Cronbach's alpha. An alpha coefficient of 0.70 is an acceptable threshold for reliability (14). A benchmark of 0.70 for Cronbach's alpha was used in studies associated with the development of similar scales like the cultural capacity scale and validation of its Arabic version (31). Based on the intraclass correlation coefficient and Cronbach's alpha, eight items were removed. The resultant questionnaire had 27 items.

Factor analysis required a sample size of at least 10 participants for each scale item (14), with a 10:1 respondent-to-item ratio, resulting in 270 samples. To achieve equal response rate from the five streams of healthcare, we obtained a minimum of 54 samples from each group through convenience sampling. This ensured the applicability of the tool to the various healthcare streams in India. An assessment tool should be a parsimonious representation of the entire spectrum of the concept of interest. Our efforts were to develop a cultural competence assessment questionnaire for healthcare professionals in India with items that were unique to the domain represented, thereby minimizing overlaps. This constitutes the property of construct validity, which was carried out by using exploratory factor analysis (EFA) with principal component analysis (PCA) and varimax rotation.

EFA involves deciding on a factoring method, choosing a rotation procedure, and interpreting the results. The number of factors that are retained during the process of EFA is decided by eigenvalues of each factor (32). According to the Kaiser–Guttman rule, all factors for which the eigenvalue is >1.0 should be retained (33). In the developed tool, the eigenvalue was >1. This is in congruent with previous studies involving EFA (34, 35).

The number of items that distinctly measured a particular domain was estimated through factor loading using PCA. Initial factor extraction with PCA yielded 49.35% as cumulative percentage variance explained by the tool. PCA was followed by varimax rotation, which is the most common orthogonal rotation method (36). Factor loading was used on 27 items. The factor loading matrix that showed a higher value implied a strong relation between the factor and the item (37), and a value of 0.35 is assumed to be the minimum loading value (38). After removal of the item with a factor loading value <0.35, factor analysis yielded a cumulative variance percentage of 50.36%.

Cultural competence of healthcare professionals implies their ability to successfully interact with and treat patients from diverse cultural backgrounds. Our validity of the hypothesis was that the Indian healthcare environment was unique due to multiple healthcare streams with often conflicting principles and practices. Thus, the difference of cultural competence with respect to healthcare streams and years of clinical experience was tested for their significance as a further step to ensure known group validity. The difference in cultural competence based on years of clinical experience, as estimated by the Kruskal–Wallis test, was not statistically significant. This finding is similar to the result of a study among nurses in Bangkok, where it was found that nursing experience did not have a significant correlation with cultural competence (39). Another study conducted among registered nurses and psychiatric unit healthcare workers also showed that experience alone does not have a significant effect on the cultural competence level (40).

There was statistically significant difference in cultural competence based on the healthcare stream. Currently, there are no available studies comparing the cultural competence of healthcare workers from different healthcare streams. Multiple assessment tools, priority of transcultural nursing in curriculum, and various cultural models in nursing prove that cultural competence is given an important role in nursing than in other healthcare professions (40–49). Analysis of the various healthcare streams yielded a statistically significant difference in cultural competence between dentistry and Ayurveda.

Strengths and limitations

The Cultural Competence Assessment Tool–India (CCT-I) is a novel attempt specifically focused on the Indian healthcare environment, where culture plays a deep-rooted effect on health. Since cultural competence development is a dynamic process, we have covered its various aspects, making this a comprehensive assessment tool that is applicable to different healthcare systems being practiced in India. Currently, India is witnessing a paradigm shift to patient-centered healthcare, which is a harbinger for the establishment of a culturally sensitive healthcare system and culturally sensitive health workers. The first step toward this is the assessment of the existing cultural competence level of organizations and its manpower for which the developed tool is appropriate. A major strength of this tool is the broad coverage of the concept of "cultural competence". Existing assessment tools confine mostly to two or three domains, while our tool covers six domains, thereby helping in a comprehensive assessment.

However, our study has a few limitations. The main limitation of the developed questionnaire is the subjective nature of the concept of "culture." Consequently, we did not undertake focus group discussion for domain and item preparation as it will be impractical to achieve a saturation of viewpoints on this vast topic. Moreover, the COVID-19 pandemic was a barrier to our communications throughout this study. Another drawback was the delicate distinction between the various aspects of the spectrum of cultural competence as depicted by the domains of the tool. In addition, the response rate of validators and participants for cognitive interview was low. Although precautions were taken to overcome social desirability bias, the sensitive nature of the topic may limit its elimination.

Policy implication

The past decade has witnessed tremendous improvement in the healthcare system owing to advancements in technology and research. However, the disease burden in the country remains unchanged, particularly among the underprivileged and underrepresented communities. This dilemma in the Indian health system highlights the urgency to identify and resolve barriers to the "health for All" concept. Multiple factors like poverty, ignorance, healthcare accessibility, social norms, and gender roles challenge our health system. An insight into these multifarious barriers shows that the majority of these factors are based on the cultural beliefs of the people. Culture plays a crucial role in the lifestyle and practices of an individual in India. However, this vital determinant is overlooked at the organizational and policymaking levels. This neglect eventually cripples the system because the benefits of medical advancements will be channeled solely to the "elite" group.

The alarming contribution of India to the global burden of disease emphasizes the critical need of integrating cultural competence training into the healthcare curriculum. Cultural competence orientation of healthcare trainees from the time they start their clinical postings is an effective strategy in enhancing healthcare accessibility and utilization, thereby downsizing the "cultural gap" existing in the Indian healthcare setting. The Cultural Competence Assessment Tool–India (CCT-I) scale that is developed through this study is an important landmark in such a scenario. This is because the assessment of healthcare workforce, irrespective of their healthcare streams and trainings, will be a harbinger of reforms in the healthcare setting like development and implementation of cultural competence enhancement programs, incorporation of such training programs in the healthcare undergraduate courses, and establishing patient-centered, culturally competent healthcare facilities. This orientation of healthcare is being witnessed in Western countries where accrediting boards and the higher education system have started making cultural competence training a mandatory exercise (45, 50–52).

Studies have shown that cultural competence of healthcare personnel bears positive outcomes in treatment and communications (41, 50, 51, 53–55). It helps mitigate the longstanding mistrust of communities in treatments and overcome the social ostracism commonly seen in diseases like leprosy, skin diseases, depression, and epilepsy. Strengthening cultural competence of healthcare professionals helps patients communicate better regarding their concerns, expectations, and fears, thereby enabling the care providers to incorporate their decisions in treatment. To achieve this patient-centered approach, it is essential for the policymakers to understand the importance of cultural competence in healthcare. Quantifying it based on a tool like CCT-I, which focuses on the Indian context, is an ideal step to capture the attention of policymaking circles for this purpose.

Research implication

The concept of cultural competence is a less ventured domain in India. Although there are multiple studies on culture and its implications on a person's life, there is scarcity of the literature in the context of healthcare. India is a land of many cultures and subcultures, and each of these has manifold beliefs and practices related to health and healing. Moreover, some indigenous healing systems in India are firmly based on cultural beliefs. Often, these multiple healing systems and patient beliefs are conflicting, thereby delaying treatments resulting in morbidity and mortality.

Therefore, the scope of research on the various culturally rooted health practices is vast. Moreover, studies on the prevalence of culture-bound syndromes and cultural practices that influence health are inadequate in India. This study also warrants future cultural competence assessment studies in the Indian setting using the CCT-I scale along with qualitative approaches like patient simulation for improved knowledge in this domain. The developed CCT-I is a steppingstone to the identification of the cultural impact in healthcare, which subsequently leads to widening the research prospects in this field.

Conclusion

This study resulted in the development of a novel cultural competence assessment tool specifically designed for Indian healthcare professionals. The tool, named Cultural Competence assessment Tool–India (CCT-I), consists of six domains and 26 items. This comprehensive tool can be used to assess the cultural competence level of healthcare professionals as the first step toward designing cultural competence training for healthcare manpower and the establishment of culturally sensitive healthcare organizations in India.

Data availability statement

The datasets presented in this study can be found in online repositories. The names of the repository/repositories and accession number(s) can be found in the article/Supplementary materials.

Ethics statement

The studies involving human participants were reviewed and approved by Institutional Ethics Committee, Amrita Institute of Medical Science. The patients/participants provided their written informed consent to participate in this study.

Author contributions

PB contributed to design of study, data collection, data interpretation, scale development, manuscript writing, manuscript revision, and final approval of the version to be published. VK contributed to design of study, statistical analysis, data interpretation, scale development, manuscript revision, and final approval of the version to be published. CJ contributed to conceptualization, design of study, data interpretation, supervision, scale development, manuscript revision, and final approval of the version to be published. All authors contributed to the article and approved the submitted version.

Acknowledgments

The authors wish to thank the validators and participants of 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

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2022.919386/full#supplementary-material

References

1. Centers for Disease Control and Prevention (CDC). *Social Determinants of Health: Know What Affects Health.* (2021). Available online at: https://www.cdc.gov/socialdeterminants/index.htm

2. Anderson LM, Scrimshaw SC, Fullilove MT, Fielding JE, Normand J. Task force on community preventive services. Culturally competent healthcare systems a systematic review. *Am J Prev Med.* (2003) 24:68–79. doi: 10.1016/S0749-3797(02)00657-8

3. Bjarnason D, Mick J, Thompson JA, Cloyd E. Perspectives on transcultural care. *Nurs Clin North Am.* (2009) 44:495–503. doi: 10.1016/j.cnur.2009.07.009

4. Cross TL, Bazron BJ, Dennis KW, Isaacs MR. *Towards a Culturally Competent System of Care: A Monograph on Effective Services for Minority Children Who Are Severely Emotionally Disturbed [Internet]. CASSP Technical Assistance Center, Georgetown University Child Development Center, 3800 Reservoir Rd.* (1989). p. 90 Available online at: https://eric.ed.gov/?id=ED330171 [cited 2021 Nov 12].

5. Betancourt JR, Green AR, Carrillo JE, Ananeh-Firempong O. Defining cultural competence: a practical framework for addressing racial/ethnic disparities in health and health care. *Public Health Rep.* (2003) 118:293–302. doi: 10.1016/S0033-3549(04)50253-4

6. Chowkwanyun M, Reed AL. Racial health disparities and COVID-19 — caution and Context. *N Engl J Med.* (2020) 383:201–3. doi: 10.1056/NEJMp2012910

7. ASPE. *HHS Action Plan to Reduce Racial and Ethnic Health Disparities: Implementation Progress Report 2011-2014.* (2015). Available online at: https://aspe.hhs.gov/reports/hhs-action-plan-reduce-racial-ethnic-health-disparities-implementation-progress-report-2011-2014-0 [cited Mar 2, 2022].

8. Carrillo JE, Green AR, Betancourt JR. Cross-cultural primary care: a patient-based approach. *Ann Intern Med.* (1999) 130:829–34. doi: 10.7326/0003-4819-130-10-199905180-00017

9. Kelly F, Papadopoulos I. Enhancing the cultural competence of healthcare professionals through an online course. *Divers Equal Health Care.* (2009) 6:77–84.

10. Sumlin LL, Garcia AA. Effects of food-related interventions for African American women with type 2 diabetes. *Diabetes Educ.* (2012) 38:236–49. doi: 10.1177/0145721711422412

11. Whittemore R. Culturally competent interventions for Hispanic adults with type 2 diabetes: a systematic review. *J Transcult Nurs.* (2007) 18:157–66. doi: 10.1177/1043659606298615

12. Kehoe KA, Melkus GD, Newlin K. *Culture Within the Context of Care: An Integrative Review [Internet]. Database of Abstracts of Reviews of Effects (DARE): Quality-assessed Reviews. Centre for Reviews and Dissemination (UK).* (2003). Available online at: https://www.ncbi.nlm.nih.gov/books/NBK70227/ [cited Mar 2, 2022].

13. Henderson S, Kendall E, See L. The effectiveness of culturally appropriate interventions to manage or prevent chronic disease in culturally and linguistically diverse communities: a systematic literature review. *Health Soc Care Comm.* (2011) 19:225–49. doi: 10.1111/j.1365-2524.2010.00972.x

14. Boateng GO, Neilands TB, Frongillo EA, Melgar-Quiñonez HR, Young SL. Best practices for developing and validating scales for health, social, and behavioral research: a primer. *Front Public Health.* (2018) 6:1–18. doi: 10.3389/fpubh.2018.00149

15. Muraleedhar Seetha S, Thomas V, Sivaram R, Sreedharan S, Nayar BR. Caries Risk Assessment and Referral Tool (CRA-RT)—a novel risk scoring system for early childhood caries in community settings. *Community Dent Oral Epidemiol.* (2020) 48:379–86. doi: 10.1111/cdoe.12542

16. Lin C-J, Lee C-K, Huang M-C. Cultural competence of healthcare providers: a systematic review of assessment instruments. *J Nurs Res.* (2017) 25:174–86. doi: 10.1097/JNR.0000000000000153

17. Schim SM, Doorenbos AZ, Miller J, Benkert R. Development of a cultural competence assessment instrument. *J Nurs Meas.* (2003) 11:29–40. doi: 10.1891/jnum.11.1.29.52062

18. Rosenkoetter U, Tate RL. Assessing features of psychometric assessment instruments: a comparison of the COSMIN checklist with other critical appraisal tools. *Brain Impair.* (2018) 19:103–18. doi: 10.1017/BrImp.2017.29

19. Demetriou C, Ozer BU, Essau CA. Self-report questionnaires. In: *The Encyclopedia of Clinical Psychology.* Hoboken, NJ: John Wiley & Sons, Ltd (2015). [cited 2022 Jan 15]. p. 1–6. Available online at: https://onlinelibrary.wiley.com/doi/abs/10.1002/9781118625392.wbecp507 doi: 10.1002/9781118625392.wbecp507

20. Polit DF, Beck CT. The content validity index: are you sure you know what's being reported? Critique and recommendations. *Res Nurs Health.* (2006) 29:489–97. doi: 10.1002/nur.20147

21. Zamanzadeh V, Ghahramanian A, Rassouli M, Abbaszadeh A, Alavi-Majd H, Nikanfar A-R. Design and implementation content validity study: development of an instrument for measuring patient-centered communication. *J Caring Sci.* (2015) 4:165–78. doi: 10.15171/jcs.2015.017

22. Lynn MR. Determination and quantification of content validity. *Nurs Res.* (1986) 35:382–5. doi: 10.1097/00006199-198611000-00017

23. Davis LL. Instrument review: getting the most from a panel of experts. *Appl Nurs Res.* (1992) 5:194–7. doi: 10.1016/S0897-1897(05)80008-4

24. Grant JS, Davis LL. Selection and use of content experts for instrument development. *Res Nurs Health.* (1997) 20:269–74.

25. Polit DF, Beck CT, Owen SV. Is the CVI an acceptable indicator of content validity? Appraisal and recommendations. *Res Nurs Health.* (2007) 30:459–67. doi: 10.1002/nur.20199

26. Shi J, Mo X, Sun Z. Content validity index in scale development. *Zhong Nan Da Xue Xue Bao Yi Xue Ban.* (2012) 37:152–5. doi: 10.3969/j.issn.1672-7347.2012.02.007

27. Wynd CA, Schmidt B, Schaefer MA. Two quantitative approaches for estimating content validity. *West J Nurs Res.* (2003) 25:508–18. doi: 10.1177/0193945903252998

28. Haeger H, Lambert AD, Kinzie J, Gieser J. Using cognitive interviews to improve survey instruments. In: *Presented at the 2012 Association for Institutional Research Annual Forum in New Orleans, LA. Louisiana: Association for Institutional Research Annual Forum.* (2012). Available from: https://scholarworks.iu.edu/dspace/handle/2022/24334 [cited 2022 Jan 8].

29. Hung M, Hon SD, Franklin JD, Kendall RW, Lawrence BD, Neese A, et al. Psychometric properties of the PROMIS physical function item bank in patients with spinal disorders. *Spine.* (2014) 39:158–63. doi: 10.1097/BRS.0000000000000097

30. Hung M, Voss MW, Bounsanga J, Gu Y, Granger EK, Tashjian RZ. Psychometrics of the patient-reported outcomes measurement information system physical function instrument administered by computerized adaptive testing and the disabilities of arm, shoulder and hand in the orthopedic elbow patient population. *J Shoulder Elbow Surg.* (2018) 27:515–22. doi: 10.1016/j.jse.2017.10.015

31. Terwee CB, Bot SDM, de Boer MR, van der Windt DAWM, Knol DL, Dekker J, et al. Quality criteria were proposed for measurement properties of health status questionnaires. *J Clin Epidemiol.* (2007) 60:34–42. doi: 10.1016/j.jclinepi.2006.03.012

32. Cruz JP, Colet PC, Bashtawi MA, Mesde JH, Cruz CP. Psychometric evaluation of the Cultural Capacity Scale Arabic version for nursing students. *Contemp Nurse.* (2017) 53:13–22. doi: 10.1080/10376178.2016.1255153

33. Larsen R, Warne RT. Estimating confidence intervals for eigenvalues in exploratory factor analysis. *Behav Res Methods.* (2010) 42:871–6. doi: 10.3758/BRM.42.3.871

34. Kaiser HF. On cliff's formula, the Kaiser-Guttman Rule, and the number of factors - Henry F. Kaiser, 1992. *Percept Mot skills.* (1992) 74:595–8. doi: 10.2466/pms.1992.74.2.595

35. Ruscio J, Roche B. Determining the number of factors to retain in an exploratory factor analysis using comparison data of known factorial structure. *Psychol Assess.* (2012) 24:282–92. doi: 10.1037/a0025697

36. Worthington RL, Whittaker TA. Scale development research: a content analysis and recommendations for best practices. *Couns Psychol.* (2006) 34:806–38. doi: 10.1177/0011000006288127

37. Costello A, Osborne J. Best practices in exploratory factor analysis: four recommendations for getting the most from your analysis. *Practi Assess Res Evaluation.* (2005) 10:1–9.

38. Norman GR, Streiner DL Lewiston. *Biostatistics : The Bare Essentials*, 3rd ed. New York, NY: Hamilton (2008). Available online at: https://www.worldcat.org/title/biostatistics-the-bare-essentials/oclc/656783039 [cited 2022 Jan 18].

39. Bunjitpimol P, Somrongthong R, Kumar R. Factors affecting nursing cultural competency in private hospitals at Bangkok, Thailand. *Int J Healthcare.* (2016) 2:5–11. doi: 10.5430/ijh.v2n1p5

40. Alpers L-M, Hanssen I. Caring for ethnic minority patients: a mixed method study of nurses" self-assessment of cultural competency. *Nurse Educ Today.* (2014) 34:999–1004. doi: 10.1016/j.nedt.2013.12.004

41. Beamon CJ, Devisetty V, Forcina Hill JM, Huang W, Shumate JA, A. *Guide to Incorporating Cultural Competency into Health Professionals" Education and Training. The National Health Law Program.* Medical School, University of Medicine and Dentistry of New Jersey, Newark, New Jersey (2006).

42. Campinha-Bacote J, A. model and instrument for addressing cultural competence in health care. *J Nurs Educ.* (1999) 38:203–7. doi: 10.3928/0148-4834-19990501-06

43. Flinkman M, Leino-Kilpi H, Numminen O, Jeon Y, Kuokkanen L, Meretoja R. Nurse Competence Scale: a systematic and psychometric review. *J Adv Nurs.* (2017) 73:1035–50. doi: 10.1111/jan.13183

44. Hagman LW. Cultural self-efficacy of licensed registered nurses in New Mexico. *J Cult Divers.* (2006) 13:105–12.

45. Holyfield LJ, Miller BH, A tool for assessing cultural competence training in dental education. *J Dent Educ.* (2013) 77:990–7. doi: 10.1002/j.0022-0337.2013.77.8.tb05567.x

46. Jeffreys MR. Development and psychometric evaluation of the transcultural self-efficacy tool: a synthesis of findings. *J Transcult Nurs.* (2000) 11:127–36. doi: 10.1177/104365960001100207

47. Lin MH, Chang TH, Lee YH, Wang PY, Lin LH, Hsu HC. Developing and validating the nursing cultural competence scale in Taiwan. *PLoS ONE.* (2019) 14:174–86. doi: 10.1371/journal.pone.0220944

48. Sargent SE, Sedlak CA, Martsolf DS. Cultural competence among nursing students and faculty. *Nurse Educ Today.* (2005) 25:214–21. doi: 10.1016/j.nedt.2004.12.005

49. Zarzycka D, Chrzan-Rodak A, Bak J, Niedorys-Karczmarczyk B, Slusarska B. Nurse Cultural Competence-cultural adaptation and validation of the Polish version of the nurse cultural competence scale and preliminary research results. *PLoS ONE.* (2020) 15:e0240884. doi: 10.1371/journal.pone.0240884

50. Coffman MJ, Shellman J, Bernal H. An integrative review of American nurses" perceived cultural self-efficacy. *J Nurs Scholarsh.* (2004) 36:180–5. doi: 10.1111/j.1547-5069.2004.04032.x

51. Duffy ME. A critique of cultural education in nursing. *J Adv Nurs.* (2001) 36:487–95. doi: 10.1046/j.1365-2648.2001.02000.x

52. Kripalani S, Bussey-Jones J, Katz MG, Genao I. A prescription for cultural competence in medical education. *J Gen Intern Med.* (2006) 21:1116–20. doi: 10.1111/j.1525-1497.2006.00557.x

53. Cai D-Y. A concept analysis of cultural competence. *Int J Nurs Sci.* (2016) 3:268–73. doi: 10.1016/j.ijnss.2016.08.002

54. Gibbs H, Like RC, South Paul JE. *Building Diverse and Inclusive Communities in Academic Health Centers. 2008 ELAM Forum on Emerging Issues.* (2008). Available online at: https://www.academia.edu/525087/Building_Diverse_and_Inclusive_Communities_in_Academic_Health_Centers [cited 2021 Nov 16].

55. Jackson CS, Gracia JN. Addressing health and health-care disparities: the role of a diverse workforce and the social determinants of health. *Public Health Rep.* (2014) 129:57–61. doi: 10.1177/00333549141291S211

frontiers | Frontiers in Public Health

TYPE Original Research
PUBLISHED 06 September 2022
DOI 10.3389/fpubh.2022.954035

Check for updates

OPEN ACCESS

EDITED BY
Miodraga Stefanovska-Petkovska,
Universidade de Lisboa, Portugal

REVIEWED BY
Sarvarubini Nainee,
Tunku Abdul Rahman
University, Malaysia
Mack Shelley,
Iowa State University, United States

*CORRESPONDENCE
Zheng Zang
zangzheng@suda.edu.cn

SPECIALTY SECTION
This article was submitted to
Life-Course Epidemiology and Social
Inequalities in Health,
a section of the journal
Frontiers in Public Health

RECEIVED 26 May 2022
ACCEPTED 02 August 2022
PUBLISHED 06 September 2022

CITATION
Zang Z (2022) The care types choice in filial culture: A cross-sectional study of disabled elderly in China.
Front. Public Health 10:954035.
doi: 10.3389/fpubh.2022.954035

COPYRIGHT
© 2022 Zang. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

The care types choice in filial culture: A cross-sectional study of disabled elderly in China

Zheng Zang [1,2]*

[1]School of Marxism, Soochow University, Suzhou, China, [2]Soochow University Base, Jiangsu Research Centre for Socialist Theory System With Chinese Characteristics, Suzhou, China

For the past few decades, studies of care types choice have been restricted to the scope of individual characteristics and health status. Meanwhile, the historiography of the research largely ignores the role of filial culture within China. This study sets out to examine the influence of the factors in the cultural context of filial piety on the choice of care types for older people with disability in China. According to the characteristics of filial culture, the factors influencing the choice of care type for the older people in China are summarized as family endowment and support. The study concludes that gender, residence, living alone or not, family income, real estate, pension and community service have momentous effects on the choice of care type of older people with disability; informal care has a substitutive effect on formal care. The research was based on cross-sectional data of CLHLS 2018 and utilized binary logistic regression analysis to compare the factors influencing the choice of old disabled people between formal and informal care. The study implies that in the context of filial culture, the older people's choice of care types is affected by family endowment and community service supply for the older people in China. In the background of filial culture, the government should give informal care official support such as cash and services, so as to change its attribute of private domain of it and enhance the quality of long-term care.

KEYWORDS

quality of care, older people with disability, informal care, formal care, aging, long-term care, influencing factor

Introduction

In the study of quality of care, the choice of care types, as an important factor in long-term care, has attracted an increasing attention. There is increasing evidence which suggests that formal care has a higher quality of care than informal one (1–6). This is not only reflected in the assessment of physical and functional health of the care recipient, but also means that the psychological stress of them is reduced due to the reduced mental and life burden of their family members (7–10). Regrettably, in the past, the research on choice of care types in long-term care in Chinese academia focused more on individual characteristics and health status (11–13). And the historiography of the research largely ignores the role of filial culture within China (14, 15).

Background and problem statement

Under the dual background of aging and filial culture, balancing formal and informal care is one of the effective ways to optimize the long-term care system and meet the caring needs of older people with disabilities (16–18). With the extension of life expectancy and the decline of human fecundity, the proportion of the aging population in China is growing rapidly, which makes China face the challenge of aging population (19). The extension of life expectancy is often accompanied by the decline of self-care ability. In other words, aging and disability occur at the same time (20). Another challenge of the aging population is the huge economic burden of long-term care on families and society (21). Care is one of the core concepts of social policy (22). The World Health Organization (WHO) proposes that long-term care is a system of activities carried out by informal caregivers (family, friends and/or neighbors) and/or professionals (health, social and others), so as to ensure that people who lack full self-care ability can maintain the highest possible quality of life according to their personal priorities and enjoy the greatest possible independence, autonomy, participation, personal enrichment and human dignity (23). In this article, formal care includes social services and nanny care (24, 25). Informal caregivers include spouse, children, grandchildren, daughter-in-law, son-in-law, other relatives, friends and neighbors (26, 27). There is also a care type which is a combination of formal and informal care (28). However, many researchers believe that this is not the mainstream type of care, so it will not be discussed in this article (29, 30).

Current research on formal and informal care is mainly focused on the research in Europe, America, Japan and Korea (6, 17, 25, 26). Fewer researchers in developing countries conducted research on this topic due to young demographic structure and the low pressure of aging. In the last decade or so, due to the rapid development of the aging population in China, Chinese researchers have been conducting research on formal and informal caregiving, drawing on research findings from developed countries (12, 18, 20). There has been much academic debate about the relationship between formal and informal care. Some studies from developed countries have suggested there might be three different relationships between them, which are complementary, alternative and parallel (31–34). According to the view of complementary one, there is a supplementary relationship between formal and informal care, that is, the more family care the older people with disability receive, the more social support they will receive (31). From the perspective of alternative relationship, the increase of informal care will produce a crowding out effect on formal care, which not only reduces the probability of older people with disability entering the nursing home or delays entry (32), but also reduces the use timing and probability of formal care services (33). In the view of parallel relationship, informal and formal care have an impact on the choice of care mode for older people with disability at the same time, rather than mutual influence. After the disabled use formal care, the demand for informal care will decline, but still maintain at a fixed level (34).

Theoretical framework

Traditionally, China has been dominated by informal care in the form of family care. Formal long-term care in China started late, but there has been a long practice of 'quasi' long-term care services based on institutionalized care of older people (20). In contrast to institutional long-term care, formal home-based and community-based long-term care in China has started to develop in the last decade or so (18). With regard to the long-term care insurance system that accompanies long-term care services, there has been a marked acceleration in the pace of government-led long-term care insurance in China's mainland over the last 5 years (21). At present, the formal care service system for older people with disability in China has not been established, and the existing care service supply cannot effectively meet the caring needs of older people with disability, resulting in the limited substitution of formal care for informal one (13, 15). At the same time, the service contents of formal and informal care are quite different, so it is difficult to realize the perfect replacement of informal care (18). With the influence of traditional culture, especially filial piety culture, informal care is still the main way of care in China (14, 20). However, with the empty nest and fewer children, the supply of informal care represented by family care cannot meet the caring needs of the existing older people with disabilities (21). Some researchers believe that informal care for the older people, as an important part of long-term care, is not free, and may exceed the economic expenditure of formal one (35, 36). Therefore, more and more researchers realize that the evaluation of informal care can help to formulate long-term care policies for its sustainable development (37). At present, the related research mainly includes: the cost of disease care of older people (38), caregiver's economic burden and opportunity cost (39), caregiver's emotional and health cost (40, 41), the trend of informal care cost (42, 43), assessment tools of older people (44), care management (45), the impact of informal care on the health of the older people (46), the comprehensive assessment of the older people (47), the impact of informal care on the health of the older people (43), etc. Informal care faces heavy burden and opportunity cost. More importantly, with the change of traditional concepts and the full opening of the pension service market, formal care services have developed rapidly and become another choice for older people with disability. This article uses CLHLS 2018 (The Chinese Longitudinal Healthy Longevity Survey in 2018) data to analyze the choice of care mode and its influencing factors for the older people with disability in China. Previous studies using CLHLS on older people's care type choices have focused on factors such as gender, education, income, widowhood, ethnic minority status, health status, number of children, and the availability of health insurance and pensions (48–51). However, the information and data collected in the CLHLS on the number of sons, housing status, community services, neighborhood relations and other information of older people in relation to the cultural context of filial piety is often overlooked. Traditional filial culture believes that filial piety is a naturally occurring affection (52). The essence of filial piety, in Mencius' view, is to provide for one's old age (53). As a son or daughter, he or she is obligated to take care of his or her parents, which includes not only taking care of their daily lives, but also comforting their hearts. Whether the children were paying respect and whether they were providing care in times of illness or elderly were most important in determining a sense of filial discrepancy in the parent (54). This feature is reflected in the proverb "Raise children to prevent aging, accumulate grain to prevent hunger." The culture of filial piety has had a great influence not only in traditional Chinese society, but also in East Asian countries such as Japan and Korea. In traditional East Asian societies, the problem of aging is basically solved within the family (55). In a previous interview with the head of the Japan Welfare Council, when it came to what the biggest challenge of developing a formal long-term care model in Japan was, it was believed that Japan had been traditionally influenced by the Confucian filial culture for a long time making the concept of family-based informal care prevalent. In the midst of rapid aging in Japan, to shift the focus of long-term care from family to society, the need to transform and break through the concept of filial culture was the most difficult and long-term task at that time. In light of this, the article argues that it may be more useful and beneficial in Chinese society to consider cultural factors of filial piety as an influencing factor in the choice of type of care for the older people. The number of activities of daily living (ADL) that cannot be completed is used to measure the disability degree of the older people, and emphasize the functional orientation between informal and formal care among groups with different disability degrees, so as to provide reference for promoting healthy aging.

In this article, we investigate the significant influence of each independent variable on the dependent variable, and test the research hypothesis. The selection of independent variables, in addition to the usual choice of individual characteristics and health status, embodies the characteristics of Chinese filial piety culture mainly in two aspects, namely "raising children for aging" and "living and working in peace and contentment". "Raising male children for aging" can be reflected in the family endowment represented by the number of sons, family income level, whether to own housing. "Living and working in peace and contentment" can be reflected in whether the community can provide services for the older people. Based on the ideas described above, the following hypotheses were proposed:

Hypothesis 1 (H1). *Different care types have different care effects.*

Hypothesis 2 (H2). *Disabled individuals with different individual characteristics have different preferences for different care types.*

Hypothesis 3 (H3). *Older people with disability with different physical and mental health status have different preferences for care types.*

Hypothesis 4 (H4). *Older people with disability with different family endowments have different preferences for care types.*

Hypothesis 5 (H5). *Whether the community provides services for the older people has an impact on the choice of care types for older people with disability.*

Materials and methods

The research was based on cross-sectional data of CLHLS 2018 to compare the influencing factors of older people with disability between formal care and informal care.

Data sources

The data used in this article is from Chinese Longitudinal Healthy Longevity Survey (CLHLS) of Peking University Center for aging health and family research in 2018. The data survey is a follow-up survey of the older people organized by the Research Center for Healthy Aging and Development of Peking University and the National Development Research Institute of China, covering 23 provinces and autonomous regions in China. The respondents are the older people aged 65 and above and the adult family members aged 35–64. The questionnaire is divided into two types: the surviving respondents' questionnaire and the family members of the deceased older people questionnaire. The survey contents of the surviving respondents' questionnaire include the basic situation of the older people and their families, socio-economic background and family structure, economic source and status, self-evaluation of health quality of life, cognitive function, personality and psychological characteristics, daily activities, life-style, life care, disease treatment and medical expenses. The survey contents of the family members of the deceased older people include the time and the cause of death in addition to all the survey content of the surviving one. After the baseline survey in 1998, the survey was conducted in 2000, 2002, 2005, 2008–2009, 2011–2012, 2014 and 2017–2018. The latest follow-up survey data (2017–2018) used in this article interviewed 15,874 older people aged 65 and above, and collected the information of 2,226 older people who died during 2014–2018. CLHLS included a large number of disabled and elderly population samples, and the disability degree of the older people was measured by the Activities of Daily Living Scale (ADLs) and the Instrumental Activities of Daily Living Scale

(IADLs), which is helpful to compare the disability degree of the older people in addition. At the same time, CLHLS data is highly representative and reliable.

In this study, the older people who need long-term care were selected according to the six indicators of ADL (bathing, dressing, eating, going to the toilet, controlling defecation and walking indoors) and the time needed to be cared for by others. According to international practice, an older person, who is partially or totally unable to care for himself/herself on at least one of the six indicators or who requires the care of another person for more than 90 days, is considered to be in need of long-term care (7, 9, 12). Through screening from CLHLS 2018, 3510 eligible people were selected as the research sample.

Variable selection

Dependent variable

There are two dependent variables: one is the choice of daily care for the older people with caring needs, including formal and informal care. Formal care includes social services and nanny care (24, 25). Informal caregivers include spouse, children, grandchildren, daughter-in-law, son-in-law, other relatives, friends and neighbors (26, 27). In the initial processing of data, formal care was assigned to 1, while informal care was assigned to 0. The second is the effect of care, including fully meeting the needs of care and not fully meeting the needs of care. The question is measured as the questionnaire "whether the help you get in the six daily activities of e1–e6 can meet your needs" (where e1–e6 stands for the six indicators of ADL) to measure, as the evaluation of control nursing effect. Through the "data conversion" processing, the answer is "fully satisfied" is assigned to 1; the answers are "basically satisfied" and "not satisfied" as "not fully satisfied," are assigned to 0.

Independent variable

According to previous studies (48, 50, 51), and considering the availability of specific data, this article selects a total of 20 independent variables, including individual characteristics, physical and mental health status, family endowment, community services for the older people. The main variables and their assignments are shown in Table 1. The reliability and validity of the collected data were tested using the Cronbach Alpha coefficient and the KMO and Bartlett tests. The reliability of the variables was analyzed using SPSS. The reliability of the variables was 0.829, which was reliable and passed the reliability test, while the KMO coefficient was 0.708, which had good validity and allowed for factor analysis.

Characteristics of the sample

At present, according to the results of China's seventh census in 2020, the average age of the Chinese population is 38.8 years old (56). Of the Chinese population, 51.24% are male; 48.76% are female (57). In China, 18.70% of the population is aged 60 and over, of which 13.50% is aged 65 and over (58).

Through the analysis of the data, the sample number of older people with disability in CLHLS data in 2018 is 3,510, and the estimated overall disability rate is 22.11%. From the internal structure of the older people with disability, the average age is 95.24 years old. The proportion of the older people is relatively large. What's more, the older they are, the more disabled they are. The proportion of older people with disability in Chinese women is 35.1% higher than that in men, which is 67.55%. The proportion of older people with disability in rural areas is 69.72%, which is 39.44% higher than that in cities and towns. Informal care provided by family members and neighbors is the main care mode for the older people with disability in China, accounting for 85.48%. The proportion of formal social long-term care is relatively small, only 14.52%. In China, 86.20% of older women with disability choose informal care. In rural areas, 93.38% of the older people with disability choose informal care. There are significant gender and urban-rural differences in the long-term care choices of older people with disability in China.

From the perspective of individual characteristics through the analysis of the sample, the average length of education of the older people with disability in China is about 3 years, which is basically equal to the level of primary school. The older people with disability living with their spouses accounts for 30.28%, and the older people with disability living alone accounts for 16.53%. 52.35% of disabled old people enjoy medical insurance, but 94.50% of disabled old people have to pay for care by themselves or their families. From the evaluation of their health status, the vast majority of disabled old people's health status is poor. Because formal care is largely based on trust in others, this article takes trust in others as an independent variable and finds that most older people with disability have higher trust in others.

From the perspective of the background characteristics of filial piety culture through the analysis of the sample, "raising male children for old age" and "living and working in peace and contentment" are two important contents of filial piety culture. In terms of family endowment, the older people with disability in China have an average of 2.8 sons. In addition, 83.62% of the disabled old people own their houses. However, only 26.91% of the older people enjoy retirement insurance benefits. Most old people think their family income is average level. In terms of community services for the older people, only 13.79% of the communities provide daily care services, 17.57% provide spiritual comfort services, and 11.59% provide daily shopping services. 22.59% of the communities will organize community and recreational activities; 21.47% of the communities provide legal aid services. Generally, the three services for the older

TABLE 1 Description of variables.

Variable category	Variable	Variable value	Mean	Standard deviation
Dependent variable	Types of care	Formal care = 1; informal care = 0	0.1452	
	Caring effect	Fully satisfied = 1; not fully satisfied = 0	0.4867	
Independent variable	**Individual characteristics**			
	Validated age	Year (continuous numerical variable)	95.24	8.411
	Gender	Male = 1; female = 0	0.3245	
	Years of schooling	Year (continuous numerical variable)	2.98	9.548
	Cohabitation with spouse	Living with spouse = 1; living without spouse = 0	0.1480	
	Current residence	City and town = 1; Rural = 0	0.3028	
	Do you live alone	Non living alone (living with family members or pension institutions) = 1; living alone = 0	0.8347	
	Medical expenses	Non family expenditure = 1; self or family expenditure = 0	0.5235	
	Care expenses	Non family expenditure = 1; self or family expenditure = 0	0.0550	
	Physical and mental health			
	Physical health	Level 1–5 (not healthy, not very healthy, middle, relatively healthy, very healthy)	2.3789	1.63555
	Trust in others	Level 1–5 (very distrusting, not very trusting, middle, relatively trusting, very trusting)	3.8621	1.18447
	Family endowment			
	Number of male children ever born	Number (continuous numerical variable)	2.80	6.993
	Income level	Level 1–5 (very poor, relatively poor, middle, relatively rich, very rich)	3.0555	0.68208
	Do you own a house	Yes = 1; no = 0	0.8362	
	Do you enjoy retirement benefits	Yes = 1; no = 0	0.2691	
	Community services for the older people			
	Daily care services	There are =1; there is no =0.	0.1379	
	On–site medical treatment and medicine delivery service	There are =1; there is no =0.	0.3676	
	Spiritual consolation service	There are =1; there is no =0.	0.1757	
	Daily shopping service	There are =1; there is no =0.	0.1159	
	Organizing social and recreational activities	There are =1; there is no =0.	0.2259	
	Legal aid services	There are =1; there is no =0.	0.2147	
	Provide health knowledge	There are =1; there is no =0.	0.4283	
	Handling neighborhood disputes	There are =1; there is no =0.	0.3115	

TABLE 2 Care effects on older people with disability of different characteristics.

Variable	Proportion of different satisfaction		χ^2	*P*
	Fully satisfied (%)	Not fully satisfied (%)		
Types of care			0.542	0.461
Formal care	48.59	51.51		
Informal care	50.42	49.58		
Validated age			74.168	0.012
Gender			2.010	0.156
Male	50.48	49.52		
Female	47.81	52.19		
Years of schooling			72.344	0.000
Cohabitation with spouse			1.687	0.194
Living with spouse	45.88	54.12		
Living without spouse	49.18	50.82		
Current residence			57.719	0.000
City and town	58.64	41.36		
Rural	44.20	55.80		
Do you live alone			3.325	0.068
Non living alone (living with family members or pension institutions)	49.44	50.56		
Living alone	45.11	54.89		
Medical expenses			21.016	0.000
Non family expenditure	52.53	47.47		
Self or family expenditure	44.23	55.77		
Care expenses			0.321	0.571
Non family expenditure	45.45	54.55		
Self or family expenditure	47.80	52.20		
Physical health			142.006	0.000
Very healthy	68.60	31.40		
Relatively healthy	61.05	38.95		
Middle	50.22	49.78		
Not very healthy	41.68	58.32		
Not healthy	21.69	78.31		
Trust in others			33.648	0.000
Very distrusting	65.18	34.81		
Not very trusting	51.19	48.81		
Middle	41.21	58.79		
Relatively trusting	51.28	48.72		
Very trusting	60.20	39.80		
Number of male children ever born			5.830	0.884
Income level			199.544	0.000
Very rich	71.11	28.89		
Relatively rich	69.90	30.10		
Middle	47.46	52.54		
Relatively poor	25.62	74.38		
Very poor	25.35	74.65		
Does it own a house			0.045	0.832
Yes	48.59	51.41		
No	49.08	50.92		
Do you enjoy retirement benefits			39.428	0.000
Yes	58.12	41.88		
No	45.50	54.50		

people provided by the community were health knowledge publicity (42.83%), visiting doctors and drug delivery (36.76%), and mediation of neighborhood disputes (31.15%).

Model construction

In this article, when describing the effect of care style for older people with disability, we use the interactive analysis method for the Chi-square test. Because the choice of daily care for older people with disability is a dependent variable, which belongs to a binary variable, the factors influencing the choice of care mode: individual characteristics, health status, family endowment, community service supply for the older people, as control variables, are included in the model for analysis. Therefore, the Binary Logistic Model was used for regression analysis. The Binary Logistic Regression Model was constructed as follows:

$$logit\,(P) = ln\frac{P}{1-P} = a + \sum_{i=1}^{n} \beta_i X_i \qquad (1)$$

In this model: P is the probability that the daily care of the older people is formal care. 1–P is the probability of informal care. X_i denotes the i_{th} influencing factor. β_i is the partial regression coefficient of the ith influencing factor. α is a constant term.

Results

Influencing factors of care satisfaction of older people with disability

According to Table 2, and the Chi-square test, there is no significant difference in formal and informal care satisfaction. Informal care has an alternative effect on formal care. Meanwhile, there are likely significant differences in the satisfaction degree of the older people with different individual characteristics. Among them, age, years of education, living in urban or rural areas, medical insurance, physical health and trust in others all significantly affect the degree of care satisfaction. In terms of family endowments, the number of sons and whether or not to own housing has no significant effect on care satisfaction. However, family income level and whether or not to enjoy retirement benefits significantly likely affect care satisfaction.

Multiple factors influencing the arrangement of care for the older people

According to the above model, a binary logistic regression analysis using SPSS was conducted to identify the factors influencing the care arrangements for the older people.

TABLE 3 Logistic regression analysis of factors influencing the choice of care types for older people with disability.

Variable	B	Sig.	Exp (B)
Individual characteristics			
Validated age	0.008	0.543	1.008
Gender	−0.477	0.025	0.620
Years of schooling	0.056	0.009	1.058
Cohabitation with spouse	−0.265	0.423	0.767
Current residence	1.588	0.000	4.892
Do you live alone	−2.089	0.000	0.124
Medical expenses	−0.145	0.490	0.865
Care expenses	0.059	0.896	1.061
Physical and mental health			
Physical health	−0.116	0.244	0.890
Trust in others	0.058	0.454	1.060
Family endowment			
Number of male children ever born	0.008	0.515	1.008
Income level	0.477	0.002	1.612
Do you own a house	−1.686	0.000	0.185
Do you enjoy retirement benefits	1.018	0.000	2.768
Community services for the older people			
Daily care services	0.767	0.006	2.154
On-site medical treatment and medicine delivery service	0.095	0.682	1.100
Spiritual consolation service	0.551	0.048	1.734
Daily shopping service	0.529	0.131	1.698
Organizing social and recreational activities	−0.467	0.156	0.627
Legal aid services	0.320	0.342	1.377
Providing health knowledge	−0.379	0.151	0.685
Handling neighborhood disputes	−1.033	0.002	0.356
Constant	−2.422	0.082	0.089
Chi-square	488.932		
-2LL	743.316		
Cox and Snell R^2	0.306		
Nagelkerke R^2	0.508		

According to the model fitting information generated by Binary Logistic Regression, $\chi^2 = 488.932$, significance level (Sig = 0.000) < 0.01, $R^2 = 0.508$, which indicates that the model has good fitting degree and good explanatory ability, as shown in Table 3.

The influence of individual characteristics on the choice of care types for older people with disability

From Table 3, we can see that gender, education years, residence and whether living alone have a likely significant impact on the choice of care type of older people with disability. The model showed that there is likely no difference in the choice of care type among age, living with spouse or not, payment method of medical expenses and payment method of care expenses. Female older people with disability tend to choose formal care, which is 1.6 times that of male older people with disability. In addition, the educated older people are more likely to choose formal care, which may be less influenced by traditional filial piety culture such as "raising children to guard against old age." For the choice of care type, older people with disability living in rural and urban areas are different. The older people with disability living in urban areas are 4.89 times more likely to choose formal care than those living in rural areas. Coupled with the influence of traditional filial piety, most rural older people will mainly focus on informal care. Urban older people are better off and can afford to pay for nannies or aged care facilities, while traditional filial influence is less influential, so they are more likely to choose formal care. Compared with the older people who are not living alone, the older people who live alone are more likely to choose formal care, and the probability is eight times of that of the older people who are not living alone.

The influence of family endowment on the choice of care types for older people with disability

It can be seen from Table 3 that family income, whether they own their housing and whether they enjoy retirement benefits have a likely significant impact on their choice of care types. The older people with disability with higher incomes are more likely to choose formal care. The older people with disability without their own housing are more likely to choose formal care, which is 5.4 times of those with real estate. This may be due to the influence of the traditional filial piety culture of "hate to leave a place where one has lived for a long time " and "living and working in peace and contentment." The older people with real estate prefer to receive informal care at home. The older people with pension benefits are more likely to receive formal care, which is 2.77 times of those without pension benefits. The number of sons has no significant effect on the choice of care type of older people with disability, indicating that the traditional concept of "raising male children for old age" is likely weakening in China.

The impact of community-based services on the choice of care types for older people with disability

It can be seen from Table 3 that "visiting doctors and delivering medicine," "daily shopping service," "organizing social and recreational activities," "providing legal aid activities" and "providing health knowledge" provided by the community have no likely significant impact on the choice of care types for the older people with disability. The "living care service" "spiritual comfort" and "solving neighborhood disputes" provided by the community have a likely significant impact on the choice of care types for older people with disability. If the community provides "living care service" and "spiritual comfort service," the older people with disability are more likely to choose formal care. In communities providing "neighborhood disputes" services, older people with disability are more likely to choose informal care.

Discussion and conclusions

Based on the latest 2018 CLHLS data, this article explores the factors that affect the choice of older people with disabilities between formal and informal care. At present, the main ways of caring for the older people in China are informal care based on family and formal care services provided by relying on social resources such as community and pension institutions (20). The study found that: (1) There are differences in the satisfaction degree of the older people with different characteristics. The accessibility of care resources is an important factor affecting the satisfaction of the older people. Family member care is usually the primary choice for the older people. (2) There is no significant correlation between the type of care and the degree of care satisfaction. Informal care has an alternative effect on formal care. (3) Male, rural, non-living alone, low-income, owing real estate, no pension older people tend to choose informal care; female, urban, living alone, high-income, pension, comprehensive community services for the older people tend to choose formal care.

Based on the above conclusion, this article argues that informal care can replace formal one, and that informal care itself is less costly and can reduce the burden on social pensions (59). Therefore, informal care should be encouraged by, giving cash subsidies, proper vacations and social endowment insurance to some caregivers to encourage them to continue to provide informal care (5). These measures could give formal support to informal care, thereby changing the private domain attributes of in-formal care at present. This is a disguised respect for the right to informal care services (60).

Informal care is still the main care model for older people with disability in China nowadays. The results of this study show that 85.48% of older people with disability choose informal care through the analysis of the sample, although the number of male children does not significantly affect the choice of care type for the older people. However, through the analysis of the sample, older people with disability in China now have an average of 2.8 sons, so they still have a broad and realistic basis for playing a role in family care service for older peoples. The choice of informal care for older people with disability may be due to the lack of necessary formal care services (61). However, from the perspective of economy, emotional needs and cultural inheritance, families are still important places for China's older people to provide for aging (18). Although the changes in social and economic development and family structure have weakened the function of family support for thousands of years, social pension services can not completely replace family support, family is the link to maintaining the emotional needs of the older people, and is also the first choice for the older people after their disability (62).

Traditional filial piety culture still has a strong influence in China. Thus, by encouraging the older people to return to the familiar community, with the help of various services and facilities provided by the community, the operation cost of public finance can be reduced and the emotional needs and spiritual dependence of the older people can be satisfied. Influenced by the Chinese culture of filial piety, increasing long-term care services for the older people at the community level and establishing a large number of community-based, fully functional embedded micro-aged care institutions should better meet the needs of the older people at different times and in different self-care situations, allowing them to enjoy continuous professional and personalized services in a familiar environment (63).

Finally, although family care can replace professional care, the long-term care of the older people must depend on the social pension service for older people with disability without children and widowed. In addition, some families will choose formal care because of various reasons (64). Therefore, it is necessary to further improve the long-term care system and to increase the input to formal care institutions. In addition, speeding up the full implementation of the long-term care insurance system can effectively reduce the cost of formal care and enable more older people to have the ability to choose professional care (65).

In the case of China, the traditional culture of filial piety regards long-term care for the older people as one of the main obligations of the family (52). However, as China faces the peak of its aging population, the increase in the number of only-child families since the implementation of family planning policies, the widespread employment of women and the diversification of family living patterns, it may be difficult to sustain a family approach to old age based on the traditional concept of filial piety (20). In this sense, the traditional concept of filial piety is also facing a possible transformation. Long-term care for older people has gradually evolved from a responsibility of traditional family to a practical social issue. What is the current state of demand for long-term care among older people in the context of China's aging population, and how to make the long-term care system work and develop in a healthy way, are questions that the article does not address but that may merit further research. At the same time, although this article analyzes the influencing factors of care types choice in the context of filial culture in China's mainland, it is lacked an analysis about the persistence and challenges to filial piety and care types choice of older people in Hong Kong, which is strongly influenced by the intersection of Chinese and Western cultures (66). In addition, this article is not able to analyze how the cultural meaning and social practice of filial care for aging parents have been transformed in Chinese immigrant families in the Western context (67).

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.

Author contributions

The author confirms being the sole contributor of this work and has approved it for publication.

Funding

This work was supported by the Chinese Ministry of Education of Humanities and Social Science Project (Grant Number 19YJCZH235; PI: ZZ), the Borui Special Project of Soochow University on the Creation of a Strong Sense of the Chinese National Community (Grant Number 21BRRT002; PI: ZZ), and the Major Projects of China's Ministry of Education's Key Research Center for Humanities and Social Sciences (Grant Number 17JJD720007).

Conflict of interest

The author declares 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

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

References

1. Chappell N, Blandford A. Informal and formal care: exploring the complementarity. *Ageing Soc.* (1991) 11:299–317. doi: 10.1017/S0144686X00004189

2. Hickenbottom SL, Fendrick AM, Kutcher JS, Kabeto MU, Katz SJ, Langa KMA, et al. National study of the quantity and cost of informal caregiving for the elderly with stroke. *Neurology.* (2002) 58:1754–9. doi: 10.1212/WNL.58.12.1754

3. Bolin K, Lindgren B, Lundborg P. Informal and formal care among single-living elderly in Europe. *Health Econ.* (2008) 17:393–409. doi: 10.1002/hec.1275

4. Wimo A, Wimo A, Sjolund BM, Jönsson L, Skoldunger A, Winblad B, et al. Trends in informal care in Sweden: is it decreasing? Cross-sectional studies with the RUD instrument. *Alzheimer and Dementia.* (2009) 5:410. doi: 10.1016/j.jalz.2009.04.1001

5. Nordberg G, von Strauss E, Kåreholt I, Johansson L, Wimo A. The amount of informal and formal care among non-demented and demented elderly persons-results from a Swedish population-based study. *Int J Geriatr Psychiatry.* (2010) 20:862–71. doi: 10.1002/gps.1371

6. Rodríguez M. Use of informal and formal care among community dwelling dependent elderly in Spain. *Eur J Public Health.* (2014) 24:668–73. doi: 10.1093/eurpub/ckt088

7. Kiyak HA, Teri L, Borson S. Physical and functional health assessment in normal aging and in Alzheimer's disease: self-reports vs family reports. *Gerontologist.* (1994) 34:324–30. doi: 10.1093/geront/34.3.324

8. Bond MJ, Clark MS, Davies S. The quality of life of spouse dementia caregivers: changes associated with yielding to formal care and widowhood. *Soc Sci Med.* (2003) 57:2385–95. doi: 10.1016/S0277-9536(03)00133-3

9. Bravell ME, Berg S, Bo M. Health, functional capacity, formal care, and survival in the oldest old: a longitudinal study. *Arch Gerontol Geriatr.* (2008) 46:1–14. doi: 10.1016/j.archger.2007.02.003

10. Vossius C, Rongve A, Testad I, Wimo A, Aarsland D. The use and costs of formal care in newly diagnosed dementia: a three-year prospective follow-up study. *Am J Geriatric Psychiatry.* (2014) 22:381–8. doi: 10.1016/j.jagp.2012.08.014

11. Tsai CH, Lai TM. Predicting the need of formal care in Taiwan: analysis of a national random sample. *Arch Gerontol Geriatr.* (2011) 53:298–302. doi: 10.1016/j.archger.2010.12.003

12. Ku L, Liu LF, Wen MJ. Trends and determinants of informal and formal caregiving in the community for disabled elderly people in Taiwan. *Arch Gerontol Geriatr.* (2013) 56:370–6. doi: 10.1016/j.archger.2012.11.005

13. Research Team of Harmonious Community (2006). Analysis of the living condition of the aged and their demands for home-based care in the Chenghua District of Chengdu. *J Chengdu Univ Technol (Social Sciences).* (2006) 21–5. doi: 10.3969/j.issn.1672-0539.2006.04.006

14. De-Hui LI, Xiao-Lu LU, Wang L. SWOT analysis on internet + community family medical model-taking "care for life" project as an example. *J Jiangxi Univ Trad Chin Med.* (2017) 29:84–88.

15. Guo JP, Cui YJ, Yin Y. SWOT analysis of rural home care for the aged-taking Liuhe district of Nanjing as an example. *Hubei Agric Sci.* (2018) 57:168–72,76. doi: 10.14088/j.cnki.issn0439-8114.2018.23.038

16. Swane CE. *The Relationship Between Informal and Formal Care.* Japan: Springer (1999).

17. Ham S, HongBaeg E. The relationship between formal care and informal care: focusing on home care service for elderly. *Korean J Soc Welf.* (2017) 69:203–25. doi: 10.20970/kasw.2017.69.4.008

18. Liu H. Formal and informal care: complementary or substitutes in care for elderly people? Empirical evidence from China. *SAGE Open.* (2021) 11:215824402110164. doi: 10.1177/21582440211016413

19. Li S, Jiang T, Song Z, Han Z. Economic impacts of policy adopted by china for its aging population. *Singapore Econ Rev.* (2020) 67:1517–43. doi: 10.1142/S0217590820500186

20. Liu M, Ma QP. *Population Aging and Prospect of China's Elderly Care and Its Related Industries.* (2021). Hershey, PA: IGI Global. doi: 10.4018/978-1-7998-4126-5.ch005

21. Jiang Y, Zhao T, Zheng H. Population aging and its effects on the gap of urban public health insurance in China. *China Econ Rev.* (2021) 68:101646. doi: 10.1016/j.chieco.2021.101646

22. Williams F. In and beyond new labour: towards a new political ethics of care. *Critic Social Policy.* (2001) 21:467–93. doi: 10.1177/026101830102100405

23. Norton EC. Long-term care. In Culyer AJ, Newhouse JP, editor. *Handbook of Health Economics.* New York, NY: North-Holland (2000).

24. Ward-Griffin C, Marshall VW. Reconceptualizing the relationship between "public" and "private" eldercare. *J Aging Stud.* (2003). doi: 10.1016/S0890-4065(03)00004-5

25. Viitanen TK. *Informal and Formal Care in Europe.* IZA Discussion Papers (2007). doi: 10.2139/ssrn.970484

26. Swinkels JC, Suanet B, Deeg DJ, van Groenou MIB. Trends in the informal and formal home-care use of older adults in the Netherlands between 1992 and 2012. *Ageing Soc.* (2016) 36:1870–90. doi: 10.1017/S0144686X1500077X

27. Kalwij A, Pasini G, Wu M. Home care for the elderly: the role of relatives, friends and neighbors. *Rev Econ Househ.* (2012) 12:379–404. doi: 10.1007/s11150-012-9159-4

28. McNamee P, Gregson BA, Buck D, Bamford CH, Bond J, Wright K, et al. Costs of formal care for frail older people in England: the resource implications of study. *Soc Sci Med.* (1999) 48:331–41. doi: 10.1016/S0277-9536(98)00351-7

29. Almberg B, Jansson W, Grafström M, Winblad B. Differences between and within genders in caregiving strain: a comparison between caregivers of demented and non-caregivers of non-demented elderly people. *J Adv Nurs.* (2010) 28:849–58. doi: 10.1046/j.1365-2648.1998.00711.x

30. Houser A, Gibson MJ, Redfoot DL. *Trends in Family Caregiving and Paid Home Care for Older People with Disabilities in the Community: Data from the National Long-Term Care Survey.* Washington, DC: Aarp Public Policy Institute (2010).

31. Langa KM, Chernew ME, Kabeto MU, Katz SJ. The explosion in paid home care in the 1990s: who received the additional services? *Med Care.* (2001) 2:147–57. doi: 10.1097/00005650-200102000-00005

32. Van Houtven CH, Norton C. Informal care and health care use of older adults. *J Health Econ.* (2004) 6:1159–80. doi: 10.1016/j.jhealeco.2004.04.008

33. Bonsang E. Does informal care from children to their elderly parents substitute for formal care in Europe. *J Health Econ.* (2009) 1:143–54. doi: 10.1016/j.jhealeco.2008.09.002

34. Li LW. Longitudinal changes in the amount of informal care among publicly paid home care recipients. *Gerontologist.* (2005) 4:465–73. doi: 10.1093/geront/45.4.465

35. Oliva-Moreno J, Peña-Longobardo LM, García-Mochón L, del Río Lozano M, Mosquera Metcalfe I, García-Calvente MDM, et al. The economic value of time of informal care and its determinants (the CUIDARSE study). *PLoS ONE.* (2019) 14:e0217016. doi: 10.1371/journal.pone.0217016

36. Mosca I, Van Der Wees PJ, Mot ES, Wammes JJ, Jeurissen PP. Sustainability of long-term care: puzzling tasks ahead for policy-makers. *Int J Health Policy Manag.* (2016) 6:195–205. doi: 10.15171/ijhpm.2016.109

37. Krol M, Papenburg J, Van Exel J. Does including informal care in economic evaluations matter? A systematic review of inclusion and impact of informal care in cost-effectiveness studies. *Pharm Econ.* (2015) 33:123–35. doi: 10.1007/s40273-014-0218-y

38. Zrubka Z. Measurement and health economic evaluation of informal care. *Orv Hetil.* (2017) 158:1363–72. doi: 10.1556/650.2017.30841

39. Rowen D, Dixon S, Hernández-Alava M, Mukuria C. Estimating informal care inputs associated with EQ-5D for use in economic evaluation. *Eur J Health Econ.* (2016) 17:733–44. doi: 10.1007/s10198-015-0718-5

40. Jansson WM, Grafström, Winblad B. Daughters and sons as caregivers for their demented and non-demented elderly parents. A part of a population-based study carried out in Sweden. *Scand J Soc Med.* (1997) 25:289. doi: 10.1177/140349489702500412

41. Swinkels JC, Tilburg TGV, Groenou MBV. Why do spouses provide personal care? A study among care-receiving Dutch community welling older adults. *Health Soc Care Commun.* (2021) 30:e953–61. doi: 10.1111/hsc.13497

42. Van den Berg B, Brouwer W, van Exel J, Koopmanschap M, van den Bos GA, Rutten F, et al. Economic valuation of informal care: lessons from the application of the opportunity costs and proxy good methods. *Soc Sci Med.* (2006) 62:835–45. doi: 10.1016/j.socscimed.2005.06.046

43. Abtan R, Rotondi NK, Macpherson A, Rotondi MA. The effect of informal caregiver support on utilization of acute health services among home care clients: a prospective observational study. *BMC Health Serv Res.* (2018) 18:1–7. doi: 10.1186/s12913-018-2880-9

44. Ross L, Neergaard MA, Petersen MA, Groenvold M. Measuring the quality of end-of-life care: development, testing, and cultural validation of the Danish version of views of informal carers' evaluation of services-short form. *Palliat Med.* (2018) 32:804–14. doi: 10.1177/0269216317740274

45. Hirschman KB, Hodgson NA. Evidence-based interventions for transitions in care for individuals living with dementia. *Gerontologist.* (2018) 58:129–40. doi: 10.1093/geront/gnx152

46. Sinn CLJ, Betini RSD, Wright J, Eckler L, Chang BW, Hogeveen S, et al. Adverse events in home care: identifying and responding with inter RAI scales and clinical assessment protocols. *Can J Aging-Revue.* (2018) 37:60–9. doi: 10.1017/S0714980817000538

47. Denewet N, Breucker D, Luce S, Kennes S, Higuet BS, Pepersack T. Comprehensive geriatric assessment and comorbidities predict survival in geriatric oncology. *Acta Clin Belgica.* (2016). 71:206–13. doi: 10.1080/17843286.2016.1153816

48. Lin WJ, Wang H, Zhou ZP. Care selection of older adults in china: informal or formal care-empirical study based on the data of CLHLS and CHARLS. *J Shanghai Univ Fin Econ.* (2014) 16:1–62.

49. Li Z. Determinants of living arrangements among the Chinese Elderly: new evidence from the CLHLS 2011 wave. *Chin Stud.* (2015) 4:32–43. doi: 10.4236/chnstd.2015.41006

50. Cheng X. Life Quality of elderly and living arrangements: empirical study based on CLHLS data. *Social Secur Stud.* (2016) 2016:31–7. doi: 10.3969/j.issn.1674-4802.2016.01.004

51. Lin W. The relationship between formal and informal care among Chinese older adults: based on the 2014 CLHLS dataset. *BMC Health Serv Res.* (2019) 19:1–8. doi: 10.1186/s12913-019-4160-8

52. Yu TD, Dimond MF. Filial piety. *J Gerontol Nurs.* (1998) 24:13–8. doi: 10.3928/0098-9134-19980301-05

53. Lau DC. *Mencius*. Shatin: Chinese University Press (2003).

54. Cheng ST, Chan AC. Filial piety and psychological well-being in well older Chinese. *J Gerontol B Psychol Sci Soc Sci.* (2006) 61:P262–9. doi: 10.1093/geronb/61.5.P262

55. Sung K-t. Measures and dimensions of filial piety in Korea. *Gerontologist.* (1995) 35:240–7. doi: 10.1093/geront/35.2.240

56. *Press Conference on Key Data Results of the Seventh National Population Census.* Available online at: http://www.stats.gov.cn/ztjc/zdtjgz/zgrkpc/dqcrkpc/ggl/202105/t20210519_1817702.html (accessed August 2, 2022).

57. *Seventh National Population Census Bulletin (No. 4).* Available online at: http://www.stats.gov.cn/ztjc/zdtjgz/zgrkpc/dqcrkpc/ggl/202105/t20210519_1817697.html (accessed August 2, 2022).

58. *Seventh National Population Census Bulletin (No. 5).* Available online at: http://www.stats.gov.cn/ztjc/zdtjgz/zgrkpc/dqcrkpc/ggl/202105/t20210519_1817698.html (accessed August 2, 2022).

59. Verbakel E, Metzelthin SF, Kempen G. Caregiving to older adults: determinants of informal caregivers' subjective well-being and formal and informal support as alleviating conditions. *J Gerontol B Psychol Soc.* (2016) 2016:gbw047. doi: 10.1093/geronb/gbw047

60. Pfau-feeinger B, Geissler B. *Care and Social Integration in European Societies.* 4th ed. Polity Press: Cambridge (2005). doi: 10.46692/9781847421340

61. Holly A, Lufkin TA, Norton EC, Van Houtven C. *Informal Care And Medical Care Utilization in Europe and the United States.* Rochester: Social Science Electronic Publishing (2007).

62. Hazif-Thomas C, Tritschler-LeMa MH, Thomas P. Family caregivers of demented elderly people and access to medical care: who gets worn out, why and what for? *Open J Psychiatry.* (2013) 3:251–5. doi: 10.4236/ojpsych.2013.32023

63. Mundt J, Lusch RF. Informal and formal care for the elderly: decision determinants and their implications. *Health Marketing Q.* (1997) 14:53. doi: 10.1300/J026v14n03_05

64. Hopp FP. Patterns and predictors of formal and informal care among elderly persons living in board and care homes. *Gerontologist.* (1999) 39:167–76. doi: 10.1093/geront/39.2.167

65. Litwin H, Attias-Donfut C. The inter-relationship between formal and informal care: a study in France and Israel. *Ageing Soc.* (2009) 29:71–91. doi: 10.1017/S0144686X08007666

66. Ng ACY, Phillips DR, Lee WKM. Persistence and challenges to filial piety and informal support of older persons in a modern Chinese society: a case study in Tuen Mu, Hong Kong. *J Aging Stud.* (2002) 16:132–53. doi: 10.1016/S0890-4065(02)00040-3

67. Lan PC. Subcontracting filial piety elder care in ethnic Chinese immigrant families in California. *J Fam Issues.* (2002) 23:812–35. doi: 10.1177/019251302236596

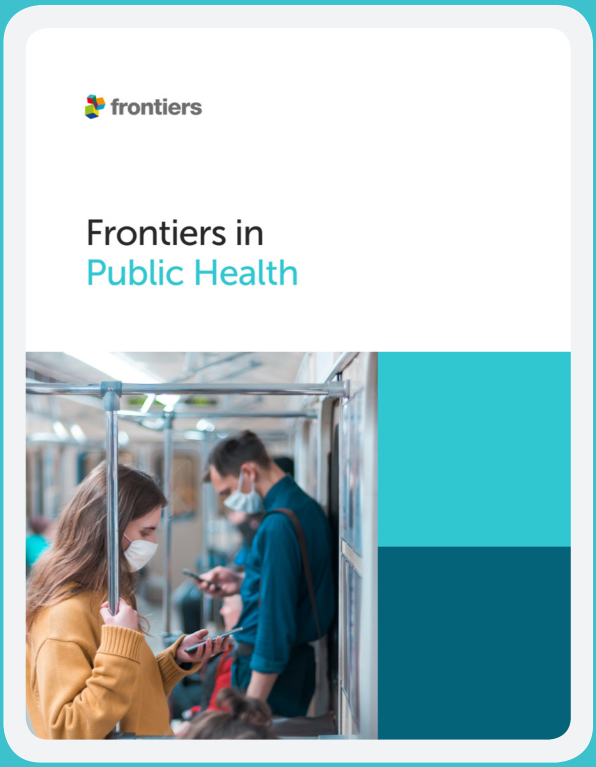
Frontiers in
Public Health

Explores and addresses today's fast-moving healthcare challenges

One of the most cited journals in its field, which promotes discussion around inter-sectoral public health challenges spanning health promotion to climate change, transportation, environmental change and even species diversity.

Discover the latest Research Topics

See more →



frontiers

Frontiers in
Public Health

Frontiers

Avenue du Tribunal-Fédéral 34
1005 Lausanne, Switzerland
frontiersin.org

Contact us

+41 (0)21 510 17 00
frontiersin.org/about/contact