THE CHALLENGE OF PALLIATIVE PSYCHOLOGY ACROSS THE LIFESPAN: BETWEEN NEW HEALTH EMERGENCIES AND PARADIGM SHIFTS

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THE CHALLENGE OF PALLIATIVE PSYCHOLOGY ACROSS THE LIFESPAN: BETWEEN NEW HEALTH EMERGENCIES AND PARADIGM SHIFTS

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Editorial: The challenge of palliative psychology across the lifespan: Between new health emergencies and paradigm shifts

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Editorial on the Research Topic
The challenge of palliative psychology across the lifespan: Between new health emergencies and paradigm shifts

Palliative care is an interdisciplinary care service which has been rapidly expanding in health care settings, differentiated between two levels. The first level is administered in hospitals and network care facilities. It concerns interventions to reduce psychophysical pain and distress caused by serious illness, and/or trauma, and the management of suffering caused by disabilities, chronic disease, or other threatening conditions. The second level involves more specifically palliative intervention in end-of-life, hospice and network health care.

Palliative psychology is in a prime position to help patients, their families, social services, and other health professionals coordinate care along the disease continuum to maximize quality of life in critical and end of life situations (Strada, 2017; Testoni et al., 2020). Indeed, palliative health professionals can offer specific competencies, clinical diagnoses of psychological difficulties and perform patient assessments. Palliative health professionals can also deliver empirically-informed counseling, psychotherapy, and assist patients with treatment planning. Further, they deliver staff support services and conduct research on program development and treatment effectiveness. Collaborating with social services, they support patients and families in the managing related difficulties.

However, several barriers exist intervention into palliative care such as the lack of inclusion of these professionals on palliative care teams, gaps in palliative care training, lack of research in the field. Therefore, this Special Topic of Frontiers in Psychology aimed to describe on the one hand the benefits of psychological work in palliative care contexts and welcome empirical findings regarding what psychology can offer in this expanding field. On the other hand, it offers new model and instruments for psychological palliative intervention. All the articles guarantee high-quality theoretical contribution in this field.

The volume is divided into three main sections. The first area is composed by five articles that reflect on death, grief and how to manage the dying and bereavement processes. The first contribution is by Laranjeira et al., titled "*Death Café as a strategy to foster compassionate communities: contributions for death and grief literacy.*" The authors describe the death-positive movement and the most recent manifestation of the death awareness movement, aimed at removing the "death taboo" that characterized the contemporary society. The second article of this first area, by Testoni et al., is inherent to bereavement. The article "*The impact of the COVID-19 pandemic on perinatal loss among Italian couples: A mixed-method study*," describes the dramatic experience of perinatal loss during the COVID-19 pandemic, utilizing a mixed method research design. Results show that participants suffered from important distress symptoms and needed a psychological support. Moriconi and Cantero-García wrote the third article, titled "*Bereavement groups: understanding grief in parents of children with cancer*" considers the dramatic experience of a child's death, which is the most stressful event and the most complex grief that families face. The main aim is to highlight the needs of bereaved parents, in order to increase the specificity and effectiveness of the therapeutic approach to prevent complications in the process of loss-making. In this same line, the fourth study titled "*Explore the bereavement needs of families of children with cancer from the perspective of caregivers: A qualitative study*," by Pakseresht et al., considers psychological, social and spiritual reactions in the parents of children with cancer. Results show that the families of these children need to receive a particular bereavement service. It recommends that members of the health care team be trained in assessing family needs, identifying risks of adverse outcomes, continuing care, and providing resources during bereavement. The fifth study titled "*Caregivers' grief in acquired non-death interpersonal loss (NoDIL): A process based model with implications for theory, research and intervention*" by Yehene et al. proposes a process-based model which addresses cognitive-emotional-behavioral challenges caregivers meet in the face of their difficult reality. These require a revision of the interpersonal schemas and the relationships that takes into account the ongoing interactions with the affected family member. The six contribution is focused on the place of death. In the article "*Levels and determinants of place of-death congruence in palliative patients: a systematic review*" by García-Sanjuán et al. reflects on the agreement between the patient's preferred place of death and their actual place of death. Their systematic literature review shows that treatment-related factors such as physical pain control, marital status, having a non-working relative, age, discussing preferred place of death with a healthcare professional, and caregiver's preference have been associated with higher levels of congruence. In the same thematic line, the article "*Preferred place of death in cancer patients: A systematic review and meta-analysis*" by Fereidouni, Rassouli et al. is specifically focused on the preferred place of death and the factors affecting it for adult patients with cancer. The systematic review and meta-analysis showed that more than half of cancer patients chose home as their preferred place of death. Therefore, guided policies need to ensure that the death of the patients in the preferred place should be considered with priority.

The second area, concerning new instruments for the assessment of psychological palliative care, is composed by five articles. The first one, titled "*Screening for distress in oncological patients: The revised version of the psychological distress inventory (PDI-R)*" by Rossi et al., presents the evaluation of the psychometric properties of the PDI and providing a revised version of the tool, offering a solid factorial structure. Results show that the PDI-R is a reliable measure of psychological distress in different samples of oncological patients, with stronger psychometric properties than the original version. Nir et al., present the second article, titled "*Psychometric properties of the Persian version of palliative outcome scale (POS)*" that examined the psychometric properties of translated version of this instrument, showing that the Persian version is a valid and reliable tool and can be used by the clinician to monitor the consequences of palliative care in Iranian cancer patients. The third article by Fereidouni, Ebadi et al., titled "*Psychometric properties of the Persian version of quality of life during serious illness-family caregiver Version 3 (QOLLTI-F V3) in COVID-19 Patients*" is aimed to indicate the psychometric properties of the Persian version of the Quality of Life in Life-Threatening Illness–Family Carer Version (QOLLTI-F) in patients with COVID-19. Results confirm that this instrument may be utilized in clinical trials and research to enhance the quality of life for family care in Iranian society. The fourth article "*Loneliness in bereavement: Measurement matters*" by Vedder et al., reviews the scientific literature to examine how loneliness after bereavement has been operationalized and measured. Results show major disparities and conclusion is that, in selecting a loneliness measure, health care professionals should come to their own well-informed decision, aided by the information provided in the review. In the fifth article "*The personal wellbeing index in Spanish palliative care professional: A cross-sectional study of wellbeing*," by Pérez-Belmonte et al., aimed to study the Personal Wellbeing Index (PWI) in a sample of Spanish palliative care professionals, as well as to study their levels of well-being and the relationships of well-being with variables such as gender, age, marital status, profession, and professional quality of life, showing that the PWI is adequate to measure personal well-being in Spanish palliative care professionals.

The third section indicates some innovation in the area of interventions. The first article, titled "*How do we talk with people living with dementia about future care: a scoping* *review*" by Visser et al., reviewed the existing research on practical communication aspects related to dementia in ACP conversations, in order to provide practical suggestions for healthcare professionals to improve their communication skills. The second article "*Practical measures for dealing with the struggles of nurses caring for people with amyotrophic lateral sclerosis (ALS) comorbid with cognitive impairment in Japan*," by Ushikubo et al., is aimed to assess the practical measures that nurses had already implemented or wanted to propose regarding care delivery for the ALS targeted. Results underline that guideline and care manual establishment may lead to improved care delivery and to the unification of care deliveries to respond to patients' strong persistency. The third article "*Spirituality during the COVID-19 pandemic: An online creative arts intervention with photocollages for older adults in Italy and Israel*" by Keisari et al. examines how expressions of spirituality were stimulated and reflected in an online creative arts intervention for older adults during COVID-19 lockdowns. The findings illustrate how creative arts intervention guided by the tenets of dignity therapy can contribute to the spiritual care of older adults during periods of social isolation, or to the spiritual support provided in palliative care. The fourth article "*The intervention areas of the psychologist in pediatric palliative care: a retrospective analysis*" by Santini et al. is a retrospective monocentric project consisting of an analysis of characteristics of psychological interventions in a pediatric palliative care (PPC) service. The analysis show how the intervention of the psychologist in PPC does not concern only end-of-life, but a series of topics that are significant for the family to guarantee psycho-social well-being oriented toward the best quality of life. The fifth article "*Developing a Model for the Establishment of Hospice Care Delivery System for Iranian the adult patients with cancer*" by Beiranvand et al. develops a model for establishing hospice care delivery system for the adult patients with cancer. Results describe seven major domains, including the need to provide a variety of settings and services, comprehensive care plan, integration into the health system, specialized manpower, organizing the accountability system, laying the groundwork in the health system, and capacity building in the community. The sixth article "*Explaining caregivers' perceptions of palliative care unmet needs in Iranian Alzheimer's patients: A qualitative study*" by Ashrafizadeh et al. describes the perception of formal and informal caregivers of the unmet needs of Iranian Alzheimer's patients. The findings provide a deep understanding of the unmet needs of Alzheimer's patients in Iran.

Author contributions

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

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.

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Caregivers' Grief in Acquired Non-death Interpersonal Loss (NoDIL): A Process Based Model With Implications for Theory, Research, and Intervention

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The number of family members caring and caregiving for a loved one undergoing physical and mental changes continues to increase dramatically. For many, this ongoing experience not only involves the *"burden of caregiving" but* also the *"burden of grief"* as their loved-one's newfound medical condition can result in the loss of the person they previously knew. Dramatic cognitive, behavioral, and personality changes, often leave caregivers bereft of the significant relationship they shared with the affected person prior to the illness or injury. This results in what we term conditions of acquired "non-death interpersonal loss" (NoDIL). Current approaches to these losses use an amalgam of models drawn from both death and non-death loss. Despite their utility, these frameworks have not adequately addressed the unique processes occurring in the interpersonal sphere where the grieving caregiver needs to reach some *modus vivendi* regarding the triad of "who the person was," "who they are now," and "who they will yet become." In this paper we propose a process-based model which addresses cognitive-emotional-behavioral challenges caregivers meet in the face of their new reality. These require a revision of the interpersonal schemas and the relationships that takes into account the ongoing interactions with the affected family member. The model and its utility to identify adaptive and maladaptive responses to NoDIL is elaborated upon with clinical material obtained from caregivers of people diagnosed with major neuro-cognitive disorder and pediatric traumatic brain injury. The article concludes with implications for theory, research and clinical intervention.

Keywords: caregiving, grief, attachment, continuing bonds matrix, interpersonal loss, ambiguous loss, bereavement

INTRODUCTION

"Bereave," according to the Merriam-Webster dictionary, is defined as "to deprive of something," or "to take away a valued or necessary possession, especially by force" (Merriam-Webster, 2020). Although this definition is rather broad and inclusive in nature, bereavement, mourning and grief are still recognized and understood as responses to the death of a loved one, also referred to as an attachment figure. Such term derives from Bowlby's attachment theory, which stresses the importance of these relationships for physical and psychological survival and well-being (Bowlby, 1969, 1973; Cassidy and Shaver, 2016; Lahousen et al., 2019). Accordingly, grief and mourning are framed as responses to broken attachment bonds (Bowlby, 1980; Jordan, 2020).

In recent decades, there is a growing recognition that other life-altering events that do not involve death, can also leave family-members bereft of an important relationship, due to dramatic changes to their affected loved one. Those can arise from a number of medical and psychological conditions, typically ongoing in nature and do not allow for closure, hence resulting in what we term as, conditions of *acquired "non-death interpersonal loss"* (NoDIL). Namely, we are referring to that class of loss that describes a change in the relationship driven by the condition where the attachment figure is no longer who he or she had been prior to dramatic change. Instead, the person to whom the griever is connected has dramatically changed in essence -in fact, the person is literally "alive" but effectively "gone." Unlike divorce, for instance, where the other person is basically the "same person" but the relationship has effectively ended, in an acquired NoDIL, the relationship may or may not continue, but it is with an "altered" person, different than the one known previously.

This type of loss is an increasingly common occurrence, constituting a *"silent pandemic"* which greatly impacts modern society. Advances and developments in medicine have led to increased numbers of people surviving what had once been fatal injuries, illnesses, and overall increased life expectancy of elder population (Olshansky, 2018). Such trends resulted in a larger portion of society experiencing pronounced brain impairments due to traumatic brain injury (TBI), stroke, Parkinson's, Alzheimer's disease among other various medical conditions and chronic illnesses. Under these circumstances, many individuals also undergo significant cognitive dysfunction involving intellect, communication and behavioral changes alongside personality alteration (Collings, 2008; Jordan and Linden, 2013; Bodley-Scott and Riley, 2015; Riley, 2016; Galimberti and Scarpini, 2018; Li et al., 2020). These extract a toll from the afflicted and their families as well. The number of family members caring and caregiving for their child/parent/spouse experiencing these conditions is likewise on the rise. In the United States alone, this number has increased by 9.5 million from 2015 to 2020 and is now 53 million (The National Alliance for Caregiving Mission, 2020). Changes in patient-care practices along with the emotional and financial expense of placing a family member in long-term care facilities, oftentimes lead many caregivers to prefer home-care placement as an arrangement to cope with their loved ones' life-situation. Either way, in addition to the stress and burden which caretaking may require, many caregivers also experience and grieve the "interpersonal loss" encountered. The person they once knew has become someone whose personality and behavior may be barely recognizable even as the physical body remains relatively intact. The latter is already known to adversely impact caregivers mental and physical health (Volicer, 2005; Pagani et al., 2014; Tzuang and Gallagher-Thompson, 2014; Saban et al., 2016; Gérain and Zech, 2019; Watson et al., 2019).

Similar to the loss of highly significant relationships following death, which set into motion the grief and mourning processes, recent studies have showed the presence of grief among caregivers in various conditions. These include caregivers of people suffering from major neurocognitive disorder (Dementia) (Chan et al., 2013; Lindauer and Harvath, 2014; Liew et al., 2020; Meichsner et al., 2020; Manevich et al., 2021), Brain Injury (Marwit and Kaye, 2006; Petersen and Sanders, 2015; Yehene et al., 2021), and Disorders of Consciousness (de la Morena and Cruzado, 2013; Yehene et al., 2020), etc. However, despite the significant contribution of these studies, less is known about the mechanisms and processes underlying psychological reactions to such acquired NoDIL and what is the nature of the grief and mourning that follow in their wake.

Accordingly, the present article aims to provide an in-depth analysis of the grief and mourning that often accompany acquired NoDIL. We do this by clarifying and specifying important aspects of the sources, processes, and various outcomes of dealing with these losses. While comparison with interpersonal loss via death contributes to these analyses, we also consider the important differences that exist between these classes of loss, as well as the implications of these differences. Given the millions of caregiving family members and the professional healthcare providers working with them, attention to these issues is highly relevant. In this regard, the paper is geared at both health care professionals and anyone else who finds this topic of relevance including caregivers. At the same time, when considering the field of NoDIL, our aim is to go beyond the familiar concept of burden (Chou, 2000; Volicer, 2005; Aitken et al., 2009; Covelli et al., 2016; Doser and Norup, 2016) and to expand beyond a consideration of well-being and biopsychosocial adaptation (Bleijlevens et al., 2015; Magnani et al., 2020). Specifically, we consider what caregivers grieve and the extent to which they "rebalance" and maintain the relationship with the affected person (Soeterik et al., 2018; Yehene et al., 2019a,b; Lond and Williamson, 2020). In that respect, understanding how caregivers find their way amongst the memories of "who the person was" and to living now with "who they are now" is highly important (see **Figure 1**). Such identification and specification of the psychological processes NoDILs entail has the potential to advance theory and research as well as supportive interventions aimed to promote caregivers' emotional well-being in clinical practice.

FROM DETACHMENT TO CONTINUING BONDS IN BEREAVEMENT DUE TO DEATH

Freud began his 1917 article on Mourning and Melancholia by describing mourning as occurring following the death of a person or the loss of something critically important to the griever. Freud's highly influential article led to decades of theory, research and practice that formulated the processes of grief and mourning as essentially concerned with letting go of the relationship with the deceased or to something highly valued (Freud, 1917). With the benefit of hindsight, the field has moved away from the idea of de-cathexis or the withdrawal of emotional investment in the deceased to one that favors a reworking of the relationship and the continuation of the emotional connection

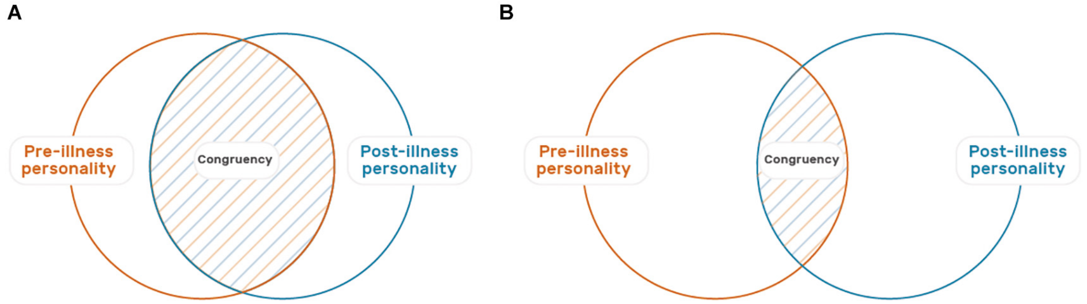

FIGURE 1 | Illustration of the personality changes and relationship challenges associated with Acquired Non-Death Interpersonal Loss (NoDIL). In these two Venn diagrams, the left orange circle represents the personality of the loved one before the illness, and the right blue circle is their current personality. The dotted area in blue and orange is the degree of overlap and continuity between the two. The overlap in diagram **(A)** is greater than that of **(B)**, signifying greater congruence between past and present personality.

to the memories of the deceased (Rubin, 1992; Klass et al., 1996; Klass and Steffen, 2018).

Along with this paradigm-shift in understanding response to death, recent models of such bereavements allude to the "non-linearity" process of grieving and the connection with the deceased throughout life. Unlike the stages-model presented by Kübler-Ross (1969), in these models the reworking of the "continuing bond" with the deceased is a central grief related task. For instance, The Two-Track Model of Bereavement (TTMB) (Rubin, 1981, 1999) addresses not only the bereaved biopsychosocial functioning and coping with the demands of life post loss (Track I), but also the nature of the bond with the deceased and the integration of the "death story" (Track II). In the complex and multi-tiered process of responding to loss, one or the other aspect of functioning and relationship to the deceased can be at the forefront of consciousness, but understanding how loss is being processed requires monitoring both tracks of the bereaved's experience (Rubin et al., 2012, 2020). Another model is the Dual Process Model (DPM) of coping with bereavement which emphasizes how grief experience unfold via an oscillation between focus on the relationship with the deceased (loss-orientation) and alternate focus upon tasks of everyday life and distractions (restoration-orientation) (Stroebe and Schut, 1999, 2010, 2016). Such reworking of the continuing bonds with the deceased is often a focus of the constructivist approach which highlights the importance of meaning making in coping with loss (Neimeyer et al., 2006; Neimeyer, 2016; Smid, 2020).

Shared by these models and by the majority of current approaches to adjustment to loss and bereavement is the understanding that the physical absence of the loved one in death elicits a process of grief and adjustment (Malkinson et al., 2000; Bonanno et al., 2011; Galatzer-Levy et al., 2018). Traversing these processes result in adaptation to their changed life circumstances with the understanding that the continued bond with the deceased fulfills critical adaptive elements for adjustment post loss (Rando, 1984; Klass et al., 1996; Malkinson, 2007; Stroebe et al., 2008; Worden, 2018). Current diagnostic approaches to complications in the bereavement process have particularly targeted the prolonged grief reaction (World Health Organization [WHO], 2018; Boelen et al., 2020; Killikelly and Maercker, 2020; Rubin et al., 2020) with criteria focused on both the yearning for the deceased together with maladaptive behaviors and impairment in the tasks of life. However, such complications in the bereavement processes may be inherently embedded in certain medical conditions involving NoDIL, resulting in prolonged grief reactions (Boerner and Schulz, 2009; Zaksh et al., 2019). Diagnostic criteria do not include or contrast this type of loss with bereavement despite the many elements shared in these two conditions. We will address this point later.

FROM STRESS AND BURDEN TO GRIEF AND MOURNING IN NON-DEATH BEREAVEMENTS

To date, current models of bereavement and loss have not sufficiently provided applicable and clinically relevant conceptualizations concerning the process of managing the relationship with the altered person, where the person has dramatically changed but the relationship continues. In contrast to bereavements due to death, in acquired NoDIL, the physical presence of the "altered" person coexists with the awareness that the essence of the person who had been there before is now "gone." In those many cases where the "lost" person is never to return, the acceptance of the "new" person will continue alongside the memory of the "old" person. The contrast between "old" and "new" has the potential to create significant complications in the grieving process and to make the loss gather significance over time. This is due to the conflicting and often mutually exclusive attachment schema (i.e., schema) regarding who the person is and how to relate to him or her. Thus, to promote psychological adaptation, for the **grieving caregivers**, some manner of processing and re-working of the relationship with their loved one, while he or she is still alive, is a daunting but necessary task.

For many decades, caregivers' psychological reaction to non-death losses was primarily understood through the lens of how their loved one's condition affected their overall well-being (Lim and Zebrack, 2004). Specifically, research has largely focused on the stress and burden characterizing caregivers' daily-life (Lazarus and Folkman, 1984; Pearlin et al., 1990; Zarit and Femia, 2008; Giovannetti et al., 2013; Tan et al., 2019) along with the personal losses resulting from their newfound responsibilities (loss of autonomy, relinquishing of anticipated future plans, etc.) (Pertl et al., 2019). Changes to their belief system regarding the world and the self were also noted ("assumptive world") (Janoff-Bulman, 1992; Parkes, 2001; Harris, 2011). Conversely, despite accumulated evidence regarding grief reaction among caregivers (Petersen and Sanders, 2015; Lond and Williamson, 2020), writings and conceptualizations concerning their loss experience, in particular those related to the interpersonal sphere (Rubin et al., 2019), remained limited in scope over the years.

Almost 20 years ago, Bruce and Schultz (2001) coined the term *"non-finite loss"* to describe an enduring loss, precipitated by a negative life event after which the source of the loss continues to be present. This term was initially used in cases of children with developmental disabilities or chronic diseases (Hobdell, 2004; O'Brien, 2007; Whittingham et al., 2013; Bravo-Benitez et al., 2019). Such loss manifests itself gradually, and is often characterized by a sense of ongoing uncertainty, repeated adjustment and accommodations, with an unforeseen end (Harris, 2011, 2019). Emotional reaction to non-finite loss has often been termed *"chronic sorrow"* (Olshansky, 1962; Coughlin and Sethares, 2017). This term refers to a set of pervasive, profound, enduring grief reactions, that are constantly triggered by painful discrepancies between present reality and what was hoped and imagined for the future. A similar notion was rendered as *"living-loss"* and also stressed the ongoing nature of the loss experience (Roos, 2002). To date, these influential concepts are still used primarily to describe parental experience in cases of child disability or chronic illnesses, and their application to cases of acquired rather than developmental loss, is still scarce. In addition to their limited use, these concepts addressing non-finite loss and chronic sorrow do not consider the relational sphere with the attendant need to consider how the attachment bond with the child is affected by the discrepancies between the "hoped for" and the "lived" developing child which is driving the chronicity of the sorrow.

Another central and influential contribution to the field of non-death loss is the work of Pauline Boss on *"Ambiguous Loss"* (Boss, 1999, 2007, 2010). Boss's framework addresses the relational sphere as when she considers discrepancies between physical and psychological absence. In the model "goodbye without leaving"/"leaving without goodbye" (Boss and Yeats, 2014), the authors refer to cases of physical presence but psychological absence (i.e., coma, dementia, mental illness) or physical absence but psychological presence (i.e., kidnapping, missing in action). According to Boss, such cases of incongruity involve great ambiguity regarding whether the person is still part of the relationship in light of the hope that they will reappear as they were. Being a condition without the finality of death, caregivers' grief remains frozen, non-legitimized and "disenfranchised" by society (Doka, 2002, 2008; Boss and Carnes, 2012). However, while Boss's "goodbye without leaving" concept is relevant for the discussion at hand, it does not sufficiently address the mechanisms or the processes in which the griever is challenged.

In our view, in cases of NoDIL, much of the loss experience is rooted in the discrepancy between "who the person was previously" and "who they are today" and "who they will be in the future." This mix of psychological representations requires a process of "working through" to come to terms with their predicament. Caregivers need to pave a way through an "interpersonal limbo" in which they are not only required to say "goodbye" to what has been the nature of the person and the relationship in the past. They are also required to say "hello" to a dramatically changed person and to manage a relationship that cannot continue unmodified.

While Boss's work largely contributed to the focus on non-death loss, her framework leaves unspecified the complexity of various conditions in which old and new aspects of the person are active in the mind of the family member. Boss refers to who was "lost" and does not sufficiently focus on "what was lost" with regards to that person. Accordingly, she largely emphasizes the management of ambiguous loss within the family alongside strategies for coping (Boss and Yeats, 2014). The mechanisms by which family members balance and re-construct these competing schemas of their altered-other have yet to be outlined. Therefore, clarifying these processes can largely contribute to the progressing field of non-death loss.

At this point, we advance a conceptual framework outlining the mechanism underlying this type of loss and the cognitive, emotional and behavioral aspects it brings about. Such a framework should also take into consideration the impact of the varying nature of the illness and prognosis, helping clinicians to identify nodal points that can elicit maladaptive responses and undue suffering.

DEATH vs. NoDIL—INTRODUCING THE NOTION OF OPEN vs. CLOSED SYSTEM

As mentioned earlier, the heart of the bereavement-due-to-death experience, however, is not the requirement of the griever to manage the demands of life for this crisis as in any other. Rather, it is the reworking of the continuing bonds with the loved one (Rubin et al., 2012, 2020). What had been the connection to a living person with possibilities and experiences of interaction must accommodate a new reality where the connection in the physical sphere has ended but continues to exist in the psychological realm only via memory and imagination. The demands of the reworking of the relationship to the deceased, therefore, become part of a predominantly *closed system*, where no interaction exists in the physical sphere and hence, there is no incoming interpersonal input to be processed. In this respect, one reorients from the external world of no connection to the inner world of memory and affect of an internalized relationship.

Conversely, in non-death losses, the ongoing relationship to the loved one is in its stead, an *open system* where the still living albeit "altered" person continues to be part of the caregiver's reality. Thus, the interaction *continues* in the physical sphere, and that continuation means that the **continuing bond** is not bound by the relationship in the past only, as in the aftermath of death. Rather, it operates as an *open system* in which *new interpersonal input* approaches reality and is being continuously integrated. Thus, the ongoing connection to the altered-loved one is influenced by the nature of the additional interactions that began with the life-altering event and continues onward, often with great uncertainty and ambiguity. Naturally, *new interpersonal input* can vary in quantity and quality per medical condition and its prognosis. Also, it may or may not contradict existing knowledge and schemas about the affected person's personality, the nature of past relationships and the envisioned future prior to the event[1].

Figure 2 illustrates and summarizes this idea on a wide spectrum of prototype conditions from normative healthy relationship, through medical conditions to death.

To summarize, the interaction with the loved one prior to the change has ended and a modified relationship is developing. Therefore, understanding the way that these processes are experienced within the griever, may shed light on the way bereavement and non-death losses unfold over time.

NoDIL—THE CONTINUING BONDS MATRIX MODEL

The following section of the article will be devoted to presenting a process-based model of continuing bonds matrix reconstruction and coping in NoDIL. This model constitutes an integration and expansion of current theoretical, clinical and empirical knowledge as reviewed in this paper, and strives to address issues that have previously remained insufficiently answered. The model described below consists of three main components: ongoing interpersonal input and its quality; 3-dimensional schemas of past, present and future; and cognitive, emotional, and behavioral changes required for adaptive coping. **Figure 3** and accompanying text put forth this process-based model.

First, and based on the above differentiation between closed vs. open systems, NoDIL is an open system that includes an ongoing flow of incoming information from the altered loved one, which is the focus of this experience and its uniqueness. The informational input of this kind leads to branching out in the cognitive-emotional domain. The memory representations of the person as they had been in the past prior to the altering event(s), exist in contrast to how they are today, or are expected to be in the upcoming future. That is, a continuing bonds matrix with and of the loved one that has undergone a personality and functional change due to an acquired medical condition.

Potential disparities between the schemas of the past, present and future may lead to dissonance and an increase in the degree of distress experienced by the individual, and therefore require them to perform a number of parallel and simultaneous processes to minimize the tension caused by these gaps. These processes can be conceptualized by a mechanism of feedback loops of assimilation and schema accommodation at the cognitive, emotional and behavioral levels, in light of the ongoing new information received from the loved one.

Schema updating is done in reference to three dimensions of time. Namely, accommodation of the present schemas in connection to the relationship as it was in the past, and updating the present schemas in light of the future to come. In other words, the personal and functional change that has taken place in the loved one requires the family member to mourn and grieve the loss of the affected person as they were in the past and were imagined to be before the aforementioned change. Accordingly, caregivers are required to integrate and re-construct past representations with present ones. Additionally, the medical prognosis requires emotional and practical preparation for the future to come from the caring and caregiving family member. That is, coordinating expectations and taking preparatory actions regarding the future relationship in the face of the disease. These processes can also be influenced by the quantity and quality of

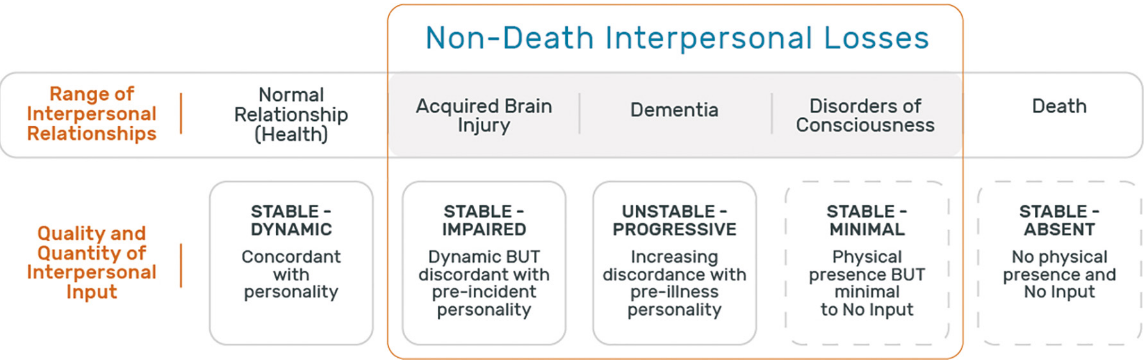

FIGURE 2 | The varying quantity and quality of incoming interpersonal input in the physical sphere in a range of interpersonal relationships. This figure presents a range of interpersonal relationships and the associated incoming information that characterizes each. The top row presents the types of relationships: normal relationship (health), non-death interpersonal losses (acquired brain injury, dementia, and disorders of consciousness as common examples), and finally death. The bottom row describes the quality and quantity of the incoming interpersonal input that characterizes each relationship.

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[1] This distinction (open vs. closed) is a heuristic one since there are many avenues by which new information is actually received in death-loss (e.g., access to social media of the deceased, input from individuals that can occur at any point in time). Nevertheless, in NoDIL this incoming information is continuous and received from the altered-person themselves.

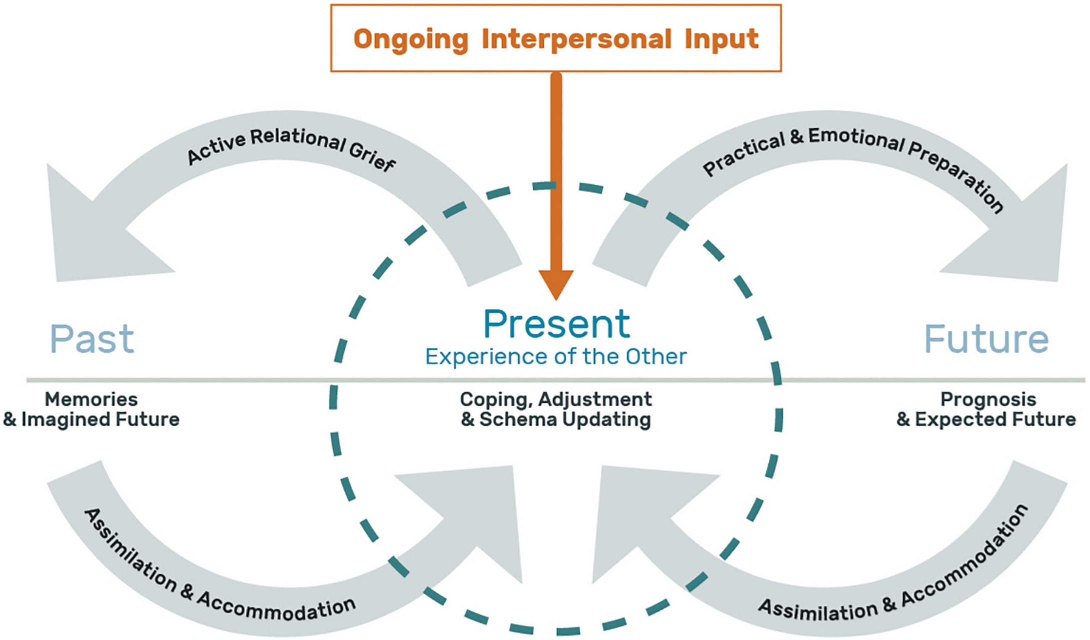

FIGURE 3 | A process-based model of continuing bonds matrix reconstruction and coping in Acquired Non-Death Interpersonal Loss (NoDIL). This figure depicts the NoDIL model where the center circle represents the present experience of the loved one and is placed midway between the time dimensions of past and future. The "past" on the left includes the memories of the person as they were prior to the illness/injury event along with what had been the imagined future of that time. The "present" encompasses the contemporary perception of the significant other along with the tasks of coping, adjustment and schema updating. On the right, the "future" represents the information and experience driven by prognosis and the expected future. Linking the three time dimensions are gray arrows (forming an infinity like symbol) that represent the dynamic interactive processes between them: Active relational grief for the "past" and its assimilation and accommodation in the "present" schema, along with emotional and practical preparation for the "future" and its assimilation and accommodation in the "present." The ongoing interpersonal input in orange, appears within a rectangular frame with an arrow facing downward indicating a continuous stream of incoming information from the loved one that impact the "present" schema. Lastly, the open spaces in the circle's perimeter stress the permeable boundaries of the present time frame.

the incoming information due to the medical condition of the patient, the personal characteristics of the caregiver, as well as the nature of the pre-morbid relationship.

Given the challenges described above, schema boundary permeability is a factor of great importance, so that schemas must be sufficiently flexible, in comparison with rigid boundaries that do not allow for schema updating and may contribute to difficulties in coping. In other words, maladaptive coping is conceptualized as deficiency in adaption and inflexibility of schema modification. Here, the person tries to interpret and force the incoming information to conform the "old" schemas, without being able to modify them sufficiently so as to incorporate this new information into and create a new "hybrid" schema. Moreover, the activation and predominance of the "old" schema, manifest in preoccupation (such as rumination on the relationship that existed before the loss) or avoidance (essentially shutting out information or not integrating it into one or more dimensions of time) serve to disrupt the process of adjustment and thus contribute to the individual's distress.

CLINICAL ILLUSTRATIONS

To illustrate the concepts we have been discussing, we now turn to vignettes of relevant clinical and research material that clarify and illustrate the analyses we have put forward. To minimize variability due to gender (Doka and Matrin, 2010) and familial-role differences in the grief process, we have focused on illustrative adaptive and maladaptive responses for the same familial-role and genders[2].

Ruth's son sustained traumatic brain injury 2 years ago at the midst of his adolescence:

> *"I miss him a lot [...]. To his previous abilities and his stubbornness. I am willing to give everything now so he would return to the way he was and the connection we had. His brothers and sister also remember the way he was in the past and the way he is now and this gap drives you crazy and doesn't let go. It's in front of you. He could do everything he wanted and now he is like a shadow of his former self. The total opposite. He speaks loudly, very repetitive, childish and needy although age wise he is approaching adulthood. He follows me everywhere. He is like a Robot [in his gait and movements]. [...] What happened to us is a terrible thing that no parent can accept! People think that he stayed alive and survived the accident. They don't understand my*

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[2] Vignettes presented in this paper were adapted from two research projects on caregivers' grief in pediatric acquired brain injury and Dementia. For both protocols institution review board (IRB) approval and consent were obtained. Caregiver's verbatim were de-identified by using pseudonymization, and by masking patients' ages, time variables and other personally identifiable information.

> *own inner experience despite the fact that he stayed alive. I fall into despair. Will I ever be able to see him settled in his own place and live independently? There is no single day that goes without this feeling coming up and me trying to push it aside."*

Rachel, married for more than four decades to a husband diagnosed with Alzheimer's disease for the last 5 years:

> *"I am not able to accept and hold onto the fact that the man I knew in the past has faded away in front of my eyes [. . .]. It is so hard to digest that his outer appearance remains unchanged—like just the shell, while the inside is no longer there [. . .]. Little by little his personality changed completely—from a gentleman to a coarse and rude fellow, and I changed from a spouse to a 24/7 caretaker [. . .]. I am constantly remembering the life we had before and our plans for the future before this cursed illness entered our lives. It simply shatters me into fragments [. . .]. Truthfully, it only gets worse every day, and I don't know if I can manage to stand it very much longer. I simply feel exhausted, worn out, angry and hopeless."*

If we utilize the prism of the model described above, the common feature that makes the experience so difficult for both mother and spouse clearly emerges. In both of these cases, the dramatic personality alteration of the loved one results in a steady flow of incoming interpersonal input that continuously clashes and contradicts with the schema of the past person that they both once knew. The different quality and quantity of information generated in their ongoing interactions with their altered loved one (serving also as a daily reminder of the loss), constantly re-activate the schema of the past person as he had been. For both Rachel and Ruth, the inevitable comparison of the "old" person and relationship with the current ones becomes a continuing aspect of their psychological experience. Their constant remembrance and longing for what was in the past and how their future "should" have been, before it was shattered by the medical condition, causes great distress and relational active grieving. Such intensity magnifies their conflicted and troubled experience, making it difficult to integrate the loss into their life-narrative. This also affects how they approach the demands of the present and how they relate to the expected-futures with their loved ones. In other words, both Rachel and Ruth struggle unsuccessfully to assimilate the alternations that have taken place in their loved ones and accommodate existing schema to better fit the "new" states of their loved one. This state of affairs can be conceptualized as relatively rigid and with impenetrable boundaries that hamper schema updating, thus impeding the process of coping and adjusting to the reality of loss.

In the language of psychological schema, the schema of the past son and husband are significantly separated from the current schema of who they now are. Inflexibility of schema modification processes do not allow to reconstruct an integrated perception of the current person and relationship and therefore, past perceptions remain "encapsulated" in the shadow of the loss resulting in an acute [and prolonged] grief reaction. Thus, it is not only the challenge associated with *burden of care* that is the source of their difficulty, but it is also the *burden of grief* that is so painful and unbearable.

We now turn to two additional cases where the process is experienced differently.

Joan's son had sustained traumatic brain injury few years ago, just before entering adolescent:

> *"There are many thoughts about [child's name] before the injury and it really hurts. I also think a lot about his future—what will be with him. Today when I sit with him to do homework I remember how he was in the past and how easy it was. I used to have a brilliant child with unique characteristics and values. He was born with a clear developmental pattern and suddenly it all shatters, it's all gone [. . .]. Nowadays, I need to cope with the acceptance of the situation and with the fact that [child's name] is a different child. The only difference is that at the beginning I didn't want to believe that it's forever and for the long run [. . .]. Things may improve further but won't change in essence. For me as a mother, I always have to go through processes in order to understand who this child is and to 'learn' about him anew. Sometimes I also get to discover his sense of persistence and his 'old' good heart. My husband still struggles and doesn't understand this. He thinks he will return to the way he was before. I really want him to become an independent normative adult, but I don't live in illusions."*

Miriam, married for almost four decades to a husband diagnosed with Alzheimer's disease 5 years ago:

> *"[husband's name] was my anchor and strong support, and now that has reversed. Today most of the time I feel like the mother to a little child rather than a spouse [. . .]. I take very good care of him because I haven't forgotten our past together. He is the biggest love of my life, and I have no doubt that if the situations were reversed, he would do the same for me. We always had this mutual trust and belief and even today, I know that although he does not always remember who I am, he trusts me very much, and I feel secure in my ability to help him, even though it comes at a great personal cost to me [. . .]. True I lost many things over the last few years, but the basic love remains, and at times, on a particular moment in my day, I remember that and it gives me the strength to go on."*

Looking at these two vignettes, the differences from the earlier cases are prominent. Perhaps the most significant of these differences is that along with the incoming interpersonal information, there is a softening of the schema-boundaries between the loved ones as they had been and as they are now. The constantly changing reality they both must face, brings to the forefront encounters with persisting behavioral and functioning changes of the loved one. In a sense, each encounter gradually penetrates, modifies and extends the internal representations of the now compromised loved one. This can be conceptualized as schema boundaries that are flexible enough in a way that allows for updating of existing schema. Assimilation of present reality and capabilities also serve as a basis for accommodation processes. This is evident in both caregivers' recognition that it is them who need to "change" so to better interact with their altered loved one and cope with the situation. Despite ongoing emotional hardships and the time it might take to arrive at such realization, this process eventually enables both the "old" and "new" representations [of the relationship and the person] to be integrated and co-exist within one whole single modified-schema. In this way, the myriad of multiple memories, emotion and attachment are not categorized as belonging to two distinct and discontinuous separate individuals as was demonstrated in the first two cases.

In the material presented heretofore, there are several ways of suggesting this integrated continuity. For Joan, it is reflected in her ability to hold on to past qualities she still identifies in her child along with attending to newfound positive qualities post-injury, as well as in her willingness to learn how to deal with the changes set in motion within him following the accident. In the case of Miriam, it is a product of holding the core of mutual commitment to each other that transcends the personality changes that have overtaken her husband. Collectively, for both Joan and Miriam, the recollection of benevolent characters and the relationship as it once was, also constitutes a resource of emotional refueling for their ability to provide their loved ones with care in the present, despite their own personal losses and costs. Lastly, it also seems that the "illness/accident story" is becoming better integrated into their life-narrative as descriptions are accompanied by movement that carries a *developmental quality*. Together, despite continuous sense of agony, these processes help them approach the future with less acute and intensified emotions and with more realistic expectations accompanied by practical adaptations.

OVERVIEW OF THE VIGNETTES

The four Vignettes presented hereby alongside their analysis underscore a number of points:

- **Open System and Schema Modification:** The incoming new interpersonal information in these "open systems" of ongoing relationships with dramatically changed loved ones, serves as a major determinant of who the current person is perceived to be. Ultimately, however, it is about how the continuing bonds and perceptions are being integrated, namely, how the entire schema of the currently perceived person is modified, that is most important.
- **Subjectivity of the Loss:** Although the degree of change may vary and at times be greater than the sum of the continuation, it is the magnitude of subjectively perceived change or incompatibility of the "old and new" versions of the attachment figure that greatly determine how caregivers will eventually respond and cope. In other words, the subjective elements of the experience may often outweigh the objective characteristics of the affected family member and the magnitude of change involved.
- **Schema Reconstruction and Psychological Outcome:** Within the continuing bonds matrix, schema updating allows for the attachment to the person their loved ones had been, to soften the distress at who they had become. Conversely, in maladaptive process, past representations of the loved one may serve as painful reminders of what has been lost and increases the distress experience within the continuing bond matrix.
- **Islands of Safe-Haven**: Integrated continuing bonds serve as a positive source of connection that enhances family members' ability to better relate to and care for the loved one. In those cases, where the link between the person "who had been" and "the person who is now" is sufficiently flexible as well as able to retain the experiences and memories of the positives in the relationship, a balanced experience can be managed.

CONCLUDING REMARKS

The present paper addresses a lacuna on the field at the intersection of death and non-death losses. To date, cases of non-death losses are understood mainly via grief models addressing losses due to death, together with unique conceptualizations in the field of non-death loss emphasizing their ongoing chronic nature. Despite their utility, these conceptual frameworks have not adequately addressed the processes occurring in the interpersonal sphere between the grieving family member and the significantly changed person who's behavioral and psychological characteristics are dramatically altered from what they had been prior to the accident or illness. In this paper we have proposed a framework that describes and elaborates on the psychological challenges experienced by those who are in close relationships with persons so radically changed. Irrespective of the caregiving burden that may be involved, the psychological bonds and cognitive-emotional representations of the affected individual and the relationship with them require revision and transformation involving grief and adaptation to the new reality.

In our paper, we have stressed the following:

- Interpersonal loss has many manifestations. Our focus on acquired NoDIL addresses those cases where the loved one has undergone major changes that greatly impact the relationship between the affected individual and his or her loved ones. These changes stem from many sources including stroke, physical trauma, dementia, mental illness and any source of dramatic, non-temporary and wide-ranging change in personality and function.
- NoDIL differs significantly from the challenges, grief and mourning associated with the death of close family members. The field of thanatology now stresses the maintenance of the emotional connection to the deceased as a significant aspect of adaptive response to loss by death. The concept of "continuing bonds," however, needs further specification to understand its applicability to the field of non-death losses as we have outlined here.
- In cases of significant deterioration due to NoDIL much of the loss experience is rooted in the discrepancy between "who they previously were" and "who they are today," and "who they are to be in the expected future." Caregivers are faced with the grief over the loss of the person and the relationship known in the past, even as they are challenged to determine what is possible and acceptable to themselves in the present relationship.
- In death, the connection in the physical sphere has ended but continues to exist in the psychological realm only via memory and imagination, thus the relationship becomes part of a predominantly closed system, where no incoming

  interpersonal input continues to arrive. Conversely, in NoDIL, the ongoing relationship to the "altered" loved one continues within an open system in which new interpersonal input is being continuously encountered. This leads to a situation of contrast between the competing experiences of the affected family member as they were in the past and as they are in the present.
- A process-based model of continuing bonds matrix reconstruction and coping in NoDIL was proposed. This model consists of three main components: ongoing interpersonal input and its quality; schemas of past, present and future; and cognitive, emotional and behavioral tasks that a person must perform for adaptive coping.
- Ultimately, the ongoing revision of the attachment bond redefines the representations of the lost loved one, the identity of the caregiver and their joint life story.

The theoretical, clinical, and research implications of these overlapping and simultaneously dichotomous experiences of the relationship with the loved one, have great significance for understanding how acquired NoDIL unfold over time. Future empirical investigations with qualitative and quantitative research methodologies as well as clinical studies are needed to further determine the utility of the model and its contributions. Ultimately, clinical practice and ongoing research focusing on the way in which close family members deal with reconciling the relationship to the person who is with person who had been can expand our understanding and our clinical work with families dealing with this increasingly common life experience.

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

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

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BRIEF RESEARCH REPORT
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The Personal Wellbeing Index in Spanish Palliative Care Professionals: A Cross-Sectional Study of Wellbeing

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Health professionals are especially exposed to stress, with consequences on professionals' health and wellbeing. However, palliative care professionals' wellbeing has been the subject of very little research. The aim of this work is to study the Personal Wellbeing Index (PWI) in a sample of Spanish palliative care professionals, as well as to study their levels of wellbeing and the relationships of wellbeing with variables such as gender, age, marital status, profession, and professional quality of life. A cross-sectional survey of Spanish palliative care professionals was conducted. The Spanish version of the PWI and the Short version of the Professional Quality of Life Scale were used. Here, 296 palliative care professionals attending patients at the end of life participated in the study. They showed medium to high levels of wellbeing. The PWI showed an adequate internal structure [$\chi^2_{(20)} = 116.130$ ($p < 0.001$)]; Comparative Fit Index (CFI) = 0.970; standardized root mean square residual (SRMR) = 0.041; root mean square error of approximation (RMSEA) = 0.140 (0.116, 0.166)] and excellent estimates of reliability [$\alpha = 0.879$ and Composite Reliability Index (CRI) = 0.923]. Wellbeing was higher for married compared to single and showed no relation with age, gender, and profession. Additionally, a structural equation model was estimated, in which a positive relation was found between wellbeing and compassion satisfaction and a negative one with burnout. The PWI is adequate to measure personal wellbeing in Spanish palliative care professionals.

Keywords: palliative care, wellbeing, burnout, health personnel, compassion fatigue, compassion satisfaction

INTRODUCTION

Health professionals are especially exposed to stress (Coetzee and Klopper, 2010). Their work confronts them on a daily basis with human suffering, on many occasions in environments of high management requirements (Galiana et al., 2017). Specifically, palliative care professionals are involved in managing situations for which a series of etiological factors have been described that contribute to the development of mental exhaustion, such as excessive care pressure, shift work, great responsibility in decision-making, little recognition by the organization, or continuous contact with illness, pain, and death, among others (Arranz et al., 2008). Consequences of such factors include insomnia, irritability, and alcohol and drug use (Shirom, 2011).

These stressful, emotionally demanding circumstances provide a favorable context to suffer psychosocial risks such as work stress, burnout, or compassion fatigue (Sansó et al., 2015), which would in turn affect their health, individual and group satisfaction, and wellbeing (Griffin and Clarke, 2011). Indeed, these psychosocial risks could also have, together with consequences on professionals' health and wellbeing, a negative impact on the productivity of the organization and the quality of care (Pipe et al., 2009; Salyers et al., 2017).

In this line, recent studies have pointed to the relation between professional quality of life and healthcare professionals' wellbeing. Professional quality of life, understood as "the quality one feels in relation to their work as a helper" (Stamm, 2010, p. 8), is defined by three components: burnout, compassion fatigue, and compassion satisfaction. Burnout is a state of mental and physical exhaustion especially relevant to human service workers (Freudenberger, 1974). Healthcare professionals are vulnerable to burnout because their work context is characterized by high-risk decisions, interacting with people, and expectations of compassion and sensitivity. However, burnout alone does not explain professionals' emotional problems from working with individuals who are suffering (Stamm, 2012; Figley, 2013). In this context, compassion fatigue, defined as the negative outcome of working with traumatized people (Bride et al., 2007), has received increasing attention in recent years. Compassion fatigue focuses specifically on the chronic worry and tension produced by continued exposure to traumatized individuals (Figley, 2013). Research on compassion fatigue has also defined its opposite or converse counteract, compassion satisfaction. Compassion satisfaction takes place when exposure to traumatic events produces gratification (Hooper et al., 2010) from the joy that comes from helping others (Stamm, 2012).

Professional quality of life has been deeply studied in the palliative care context. It has been linked to protective factors, such as self-care (Alkema et al., 2008; Aycock and Boyle, 2009; Neville and Cole, 2013), empathy (Hansen et al., 2018), awareness (Cole, 1997; Novack et al., 1997; Epstein, 1999; Hutchinson, 2011; Thieleman and Cacciatore, 2014; Thompson et al., 2014), or competence and attitudes toward death (Holland and Neimeyer, 2005). For instance, Sansó et al. (2015) tested a mapping model with variables involved in palliative care professionals' quality of life. Protective variables found in the study were self-care, awareness, and coping with death competence. The practice of self-care has been found to be important for coping with occupational stressors in general healthcare professionals (Neville and Cole, 2013; Sorenson et al., 2016) and seems to be even more important in the palliative care context, as professionals frequently face high stress and an emotionally charged environment, with prominent spiritual and existential issues. In the same way, increasing levels of self-awareness also affect levels of professional quality of life (for example, see Gustin and Wagner, 2013; Sansó et al., 2019). For example, Sansó et al. (2019) found that palliative care professionals' compassion fatigue and burnout decreased after an intervention based on mindfulness and compassion.

Regarding the relationship between wellbeing and professional quality of life, in the work of Sansó et al. (2020), the authors found that professional quality of life explained almost 60% of variance of nurses' wellbeing. As pointed out in this study, a detriment to the quality of professional life of healthcare personnel can have important consequences on their personal wellbeing (Koh et al., 2015; Sansó et al., 2020). In this sense, health professionals can develop optimal states of wellbeing as long as they have work and personal resources to turn work demands into a source of learning and professional and personal growth that give meaning to the work done (Donoso et al., 2015). It is worth highlighting the importance of studying wellbeing in healthcare personnel, as we know that low levels of wellbeing are related to poorer patient safety (Hall et al., 2016).

Traditionally, wellbeing has been conceptualized from two different perspectives (Sancho et al., 2020). On the one hand, the hedonic tradition conceptualizes a happy person as one who experienced greater positive than negative emotions; that is, wellbeing is equivalent to maximizing personal pleasures through the satisfaction of certain needs (Henderson and Knight, 2012). On the other hand, wellbeing is considered eudaemonic when complex vital goals are achieved with a high degree of personal significance (Henderson and Knight, 2012). Specifically, Ryff (1989) defined psychological or eudaemonic wellbeing as a broad construct considered the main indicator of positive functioning, composed of six well-differentiated dimensions: self-acceptance, positive relationships with others, mastery of the environment, autonomy, purpose in life, and personal growth. Despite the fact that many studies have considered wellbeing from a unilateral approach (either hedonic or eudaemonic), research recognizes that both perspectives are different (Diener et al., 2009) and even complement each other (Ryan and Deci, 2001). Seligman (2002) presented an integrated approach to psychological wellbeing in his first theory, *Authentic Happiness Theory*, which has been currently reformulated into the theory of wellbeing PERMA (Positive emotion, Engagement, Relationships, Meaning, and Accomplishments) (Seligman, 2011). In his first theory, Seligman proposed three ways to achieve satisfaction with life: positive emotions, strengths, and positive groups or institutions (Seligman, 2002). In his latest reformulation, Seligman focuses in flourishing and proposes two other complementary elements: personal ties and achievement (Seligman, 2011). This new theory is an interesting proposal in terms of welfare. However, it has not been fully developed and does not yet have solid empirical proof.

These theories have resulted in the development of various instruments to measure wellbeing, such as the Satisfaction With Life Scale (Diener et al., 1985), Ryff's Psychological WellBeing scales (Ryff, 1989), or the Flourishing Scale (Diener et al., 2010). These scales, however, do not take into account relevant facets of wellbeing, such as living standards, health, or safety. The Personal Wellbeing Index (PWI) responds to these limitations. The PWI is derived from the Comprehensive Quality of Life Scale (ComQol), which was originally developed by Cummins (1997). The PWI is composed of eight domains of wellbeing, including standard of living, personal health, achieving in life, personal relationships, personal safety, community connectedness, future security, and spirituality–religion (International Wellbeing Group, 2006). Its psychometric properties have been studied in several countries and populations, including Chilean young adults (Alfaro et al., 2013), vulnerable users of the public health system in Santiago de Chile (Oyanedel et al., 2015), Australian patients with end-stage kidney disease (Weinberg et al., 2016), Dutch adults (Van Beuningen and De Jonge, 2011), and Portuguese patients with chronic kidney disorder (Mota et al., 2016). Despite this evidence, and up to date, it has not been used to assess wellbeing in palliative care professionals. In fact, palliative care professionals' wellbeing has been the subject of very little research. Although it is true that the literature on their quality of professional life is increasing (i.e., Sansó et al., 2015; Frey et al., 2018; O'Mahony et al., 2018), very little is known about their wellbeing.

The objective of this work is to study the psychometric properties PWI in a sample of Spanish palliative care professionals, as well as to study their levels of wellbeing and the relationships of wellbeing with variables such as gender, age, marital status, profession, and professional quality of life.

MATERIALS AND METHODS

Study Design

A cross-sectional survey of Spanish palliative care professionals was conducted to assess professionals' wellbeing and other variables related to work conditions. This cross-sectional study has been reported using the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) Statement (von Elm et al., 2008).

Setting and Participants

The study was conducted during January–February 2020. Professionals were encouraged to participate through the list of members of the Spanish Society for Palliative Care (SECPAL). For inclusion, the participant had to be a healthcare professional (physician, nurse, psychologist, nursing assistant, social worker, or other), who currently cared for patients at the end of their lives, albeit not necessarily in palliative care settings. Those professionals not working in the moment of the survey were excluded in order to address potential sources of bias.

We took as a starting point for sample size determination the Monte Carlo data simulation study carried out by Wolf et al. (2013). According to their results, and expecting the minimum value for standardized factorial loadings (0.50), with an estimated power of 80% or greater, $\alpha = 0.05$, and bias in parameter or standard error estimates not exceeding 5%, the minimum sample size for a CFA model of one-factor with eight indicators would be $N = 70$. However, a minimum sample size of $N = 200$ was established, following Kline's (2015) recommendation for structural equation models.

Measures

The present study included two main outcomes:

- The Spanish version of the PWI. For the translation of the scale, we used the backward and forward translation process. First, the scale was translated into Spanish by a professional native; it was then translated back into English by another native professional, and no differences were found. The resulting Spanish version of the scale can be consulted in **Table 1**.
- The Short version of the Professional Quality of Life Scale (Galiana et al., 2020). It comprises three subscales: compassion satisfaction, compassion fatigue, and burnout. Each dimension is represented in the scale by three items and scored by the use of a 5-point Likert scale. Reliability estimates in this study ranged from 0.821 to 0.843.

Data Analysis

First, descriptive statistics for the items of the scale, including means, standard deviations, and minimum and maximum scores, were calculated.

Second, for the study of the internal structure, a confirmatory factor analysis was hypothesized, estimated, and tested, in which a factor of wellbeing explained the eight items of the PWI. To assess the model fit, we used the chi-square statistic, the Comparative Fit Index (CFI), the standardized root mean square residual (SRMR), and the root mean square error of approximation (RMSEA). Cutoff criteria to determine good fit were CFI above 0.90 (better over 0.95) and SRMR or RMSEA below 0.08 (better under 0.05) (Hu and Bentler, 1999). However, RMSEA has shown poor performance in structural models with few degrees of freedom (Kenny et al., 2015).

We employed weighted least square mean and variance-corrected (WLSMV) as the estimation method, according to the

TABLE 1 | English and Spanish versions of the Personal Wellbeing Index.

English version of the Personal Wellbeing Index (International Wellbeing Group, 2006)	Spanish version of the Personal Wellbeing Index
How satisfied are you with. . .?	¿Cómo de satisfecho estás con. . .?
1. your standard of living? [Standard of Living]	1. tu nivel de vida? [Nivel de Vida]
2. your health? [Personal Health]	2. tu salud? [Salud Personal]
3. what you are achieving in life? [Achieving in Life]	3. lo que has conseguido en tu vida? [Éxitos en la Vida]
4. your personal relationships? [Personal Relationships]	4. tus relaciones personales? [Relaciones Personales]
5. how safe you feel? [Personal Safety]	5. tu seguridad? [Seguridad Personal]
6. feeling part of your community? [Community-Connectedness]	6. tu sentimiento de formar parte de una comunidad? [Conexión con la Comunidad]
7. your future security? [Future Security]	7. tu seguridad futura? [Seguridad Futura]
8. your spirituality or religion? [Spirituality–Religion]	8. tu espiritualidad o religión? [Espiritualidad–Religión]

ordinal nature of the data and its non-normality (Flora and Curran, 2004; Brown, 2006; Muthén and Muthén, 2017).

Third, we studied the reliability of the scale, with both internal consistency estimates for the items (homogeneity, alpha if item deleted, and inter-item correlations) and the scale [Cronbach's alpha and Composite Reliability Index (CRI)].

Fourth, the relations of wellbeing with age, gender, marital status, and profession were analyzed. We calculated Pearson correlation between PWI and age; *t*-tests for independent samples to study the relation between PWI and gender; and analyses of variance (ANOVA) to study the relation between PWI and marital status and profession. For the ANOVAs, marital status was recoded, and widows/widowers were eliminated ($n = 2$). Profession was also recoded, and nursing assistants ($n = 13$) and social workers ($n = 17$) were recoded into "other professions" because of its small sample size.

Finally, personal wellbeing was related to professional quality using a full structural equation model. Specifically, compassion satisfaction, burnout, and compassion fatigue were hypothesized to predict personal wellbeing. Both the dimensions of professional quality of life and personal wellbeing were modeled as latent factors, and consequently, free of measurement error. In order to assess model fit, the fit criteria mentioned above were used.

For the statistical analyses, SPSS version 24 (IBM Corp, 2016) and MPLUS version 8.4 (Muthén and Muthén, 2017) were used.

Ethical Considerations

The study was approved by the Ethics Research Committee at the University of the Balearic Islands (82CER18). People participated voluntarily and anonymously. The study complied with the ethical principles for research in health sciences established in the Declaration of Helsinki (World Medical Association, 2013). The participants signed an informed consent document to authorize the collection and processing of their information, and they were able to withdraw their consent at any time and without any consequences.

RESULTS

A total of 303 palliative care professionals completed the survey. Out of them, 296 attended patients at the end of life. Mean age was 43.9 years old ($SD = 10.15$). Here, 77.40% ($n = 229$) were women, 22.3% ($n = 66$) were men; 0.3% ($n = 1$) was missing. Concerning their marital status, 64.9% ($n = 192$) were married or living as a couple, 24.0% ($n = 71$) were single, 9.8% ($n = 29$) were divorced, and 0.7% ($n = 2$) were widowed; 0.7% ($n = 2$) were missing. Finally, regarding profession, 43.6% ($n = 129$) were nurses, 31.4% ($n = 93$) were clinicians, 8.4% ($n = 25$) were psychologists, 5.7% ($n = 17$) were social workers, 4.4% ($n = 13$) were nursing assistants, and 5.1% ($n = 15$) had other professions; 1.4% ($n = 4$) were missing.

TABLE 2 | Item description: Mean, standard deviation, minimum and maximum scores, factorial loadings, and reliability estimates.

Item number	Mean	SD	Min.	Max.	λ	r_{it}	$\alpha_{i.i.d.}$
1	4.06	0.66	1.00	5.00	0.762	0.607	0.868
2	3.93	0.74	1.00	5.00	0.702	0.592	0.869
3	4.14	0.69	1.00	5.00	0.851	0.719	0.858
4	4.09	0.81	1.00	5.00	0.792	0.693	0.859
5	3.90	0.80	1.00	5.00	0.921	0.774	0.850
6	4.00	0.81	1.00	5.00	0.790	0.653	0.863
7	3.68	0.78	1.00	5.00	0.807	0.672	0.861
8	3.77	0.88	1.00	5.00	0.544	0.473	0.885

SD, standard deviation; Min., minimum score; Max., maximum score; λ*, factor loading;* r_{it}*, correlation item-total;* $\alpha_{i.i.d.}$*, alpha if item deleted.*

Items' Descriptive Statistics

PWI items showed medium to high levels in the eight domains of personal wellbeing, with means ranging from 3.68 (item 7, Future Security) to 4.14 (item 3, Achieving in Life; **Table 2**).

Confirmatory Factor Analysis

The CFA showed an adequate fit, except for the RMSEA: $\chi^2_{(20)} = 116.130$ ($p < 0.001$); CFI = 0.970; SRMR = 0.041; RMSEA = 0.140 (0.116, 0.166). Based on the results of Kenny et al. (2015), the overall fit was considered good. Factor loadings were adequate, ranging from 0.544 (item 8) to 0.921 (item 5). Details can be consulted in **Table 2**.

Reliability

Evidence of reliability of the scale was excellent: Cronbach's alpha was 0.879, and CRI was 0.923. Item 8 (Spirituality–Religion) was the one with the lower homogeneity and reliability, with a correlation with the total score of 0.473 and increasing up to 0.885 the alpha of the scale if the item was deleted (**Table 2**).

Univariate Differences in Wellbeing

The correlation with age was $r = 0.057$, 95% CI (−0.059, 0.171) ($p = 0.332$), indicating no relation between age and palliative care professionals' wellbeing. Concerning the relation between gender and wellbeing, no statistical differences were found between men and women: $t_{(293)} = -0.383$; $p = 0.702$; Cohen's d = −0.054, 95% CI (−0.327, 0.220). There were statistically significant differences in the wellbeing of professionals depending on their marital status: $F_{(2,289)} = 4.835$; $p = 0.009$; $\eta^2 = 0.032$, 95% CI (0.002, 0.078). *Post hoc* pairwise comparisons pointed to higher values of wellbeing for married when compared to single: $t_{(293)} = -2.908$; $p = 0.011$; Cohen's d = −0.426, 95% CI (−0.701, −0.151). Regarding profession, no statistically significant differences were found in physicians, nurses, psychologists, and other professionals' levels of wellbeing [$F_{(3,\ 288)} = 1.141$; $p = .333$; $\eta^2 = 0.012$; 95% CI (0.000, 0.038); see **Table 3**].

Relation of Wellbeing and Professional Quality of Life

Personal wellbeing was related to professional quality of life using the short version of the ProQOL (Galiana et al., 2020).

TABLE 3 | Descriptive statistics for personal wellbeing in the different groups.

Variable	Groups	Personal wellbeing score	
		Mean	SD
Gender	Women (*n* = 229)	3.93	0.60
	Men (*n* = 66)	3.96	0.49
Marital status	Single (*n* = 71)	3.81	0.57
	Married/living as a couple (*n* = 192)	4.02	0.46
	Divorced (*n* = 29)	3.85	0.72
Profession	Doctor (*n* = 93)	3.88	0.61
	Nurse (*n* = 129)	4.01	0.49
	Psychologist (*n* = 25)	3.93	0.41
	Others (*n* = 45)	3.93	0.48

Specifically, we hypothesized, estimated, and tested a structural equation model, in which the dimensions of compassion satisfaction, burnout, and compassion fatigue predicted personal wellbeing.

The model showed an adequate fit: $\chi^2_{(113)} = 297.372$ ($p < 0.001$); CFI = 0.944; SRMR = 0.055; RMSEA = 0.075 (0.065, 0.085). Considering analytical fit, as displayed in **Figure 1**, compassion satisfaction positively predicted personal wellbeing, whereas burnout was a negative predictor. Compassion fatigue did not show a statistically significant relation with personal wellbeing. Overall, 32.6% of personal wellbeing variance was explained ($R^2 = 0.326$; $p < 0.001$).

DISCUSSION

The aim of this study was to offer evidence of the psychometric properties of the PWI in a sample of Spanish palliative care professionals, as well as to study their levels of wellbeing and the relationships of wellbeing with variables such as gender, age, marital status, profession, and professional quality of life.

In regard to wellbeing levels, professionals showed medium–high levels of wellbeing, with higher levels in the domains of achieving in life, personal relationships, standard of living, and community connectedness and slightly lower levels in future security, spirituality–religion, personal safety, and personal health. However, in all domains, means exceeded the medium point of the scale. This is somehow counterintuitive. A systematic review carried out in healthcare professionals' wellbeing showed that over half of the articles measuring wellbeing pointed to poor wellbeing, as measured using a variety of definitions (depression, anxiety, job stress, mental health, or distress; Hall et al., 2016). These differences could be due to the definition and measurement of wellbeing, given that this research included a specific measure of wellbeing, the PWI, whereas in the study of Hall et al. (2016), wellbeing measures were far more diverse. Differences observed in the present study with respect to previous research could also be explained by the fact that this project has been carried out exclusively on palliative care professionals instead of healthcare personnel in general.

Results regarding the factor structure offered clear evidence of a single dimension of wellbeing, as in previous research (Van Beuningen and De Jonge, 2011; Weinberg et al., 2016). Domains with higher factor loadings, and therefore, with higher relevance in the personal wellbeing conceptualization for this population, included personal safety, achieving in life, and future security. These results are similar to the ones found by Van Beuningen and De Jonge (2011), who found higher factor loadings for the dimensions of living standard, achieving in life, community, and future security. They were also similar to the results of Weinberg et al. (2016), who found, in a sample of people with end-stage kidney disease, higher factor loadings for future security, personal safety, and standard of living. According to our results, we could suggest that, for the general factor of wellbeing, issues arising as most important for the conceptualization of palliative care professionals' wellbeing are also those that have been found in different populations, such as general population or patients at end of life.

Concerning reliability results, the scale showed adequate evidence. In fact, Cronbach's alpha value was very similar to the one found by Van Beuningen and De Jonge (2011). With respect to items' reliability evidence, item eight (spirituality–religion) showed some reliability problems, which also occurred in the study of Van Beuningen and De Jonge (2011). However, several studies have highlighted the importance of spirituality and/or religion for personal wellbeing (Holland and Neimeyer, 2005; Alkema et al., 2008), and the global scale showed appropriate psychometric properties when the item was included, thus we retained item 8 in the Spanish version of the PWI.

In regard to the relation of wellbeing with age, it was not statistically significant. Although Van Beuningen and De Jonge (2011) found that young people score higher on wellbeing than old people, these results were reported in a sample with higher age dispersion (from 18 years old to 65 and older), whereas in our sample, most participants were middle-aged adults. Therefore, we can argue that age does not work differently for the general population and palliative care professionals, as there is not such variability of age in our study.

Concerning gender, no differences in wellbeing levels were found between men and women. This result is also counterintuitive, as previous research has shown poor levels of health and quality of life for women. For instance, Ta'an et al. (2020) found higher levels of depression, anxiety, stress, and occupational stress for women in a sample of Jordanian healthcare professionals. However, in specific studies with the PWI (Van Beuningen and De Jonge, 2011), no differences in wellbeing have been found, as in our study. In fact, even in the literature on professional quality of life, results regarding gender differences are not clear. For example, Okoli et al. (2020) have recently found the predictive power of gender on compassion satisfaction (which was higher for women) but no differences in burnout or compassion fatigue. Previously, Sprang et al. (2007) had pointed higher levels of compassion fatigue in women but no differences in compassion satisfaction or burnout. Future studies that delve into the relationship between gender and quality of professional life and wellbeing in health professionals, and specifically in palliative care professionals, will be welcome.

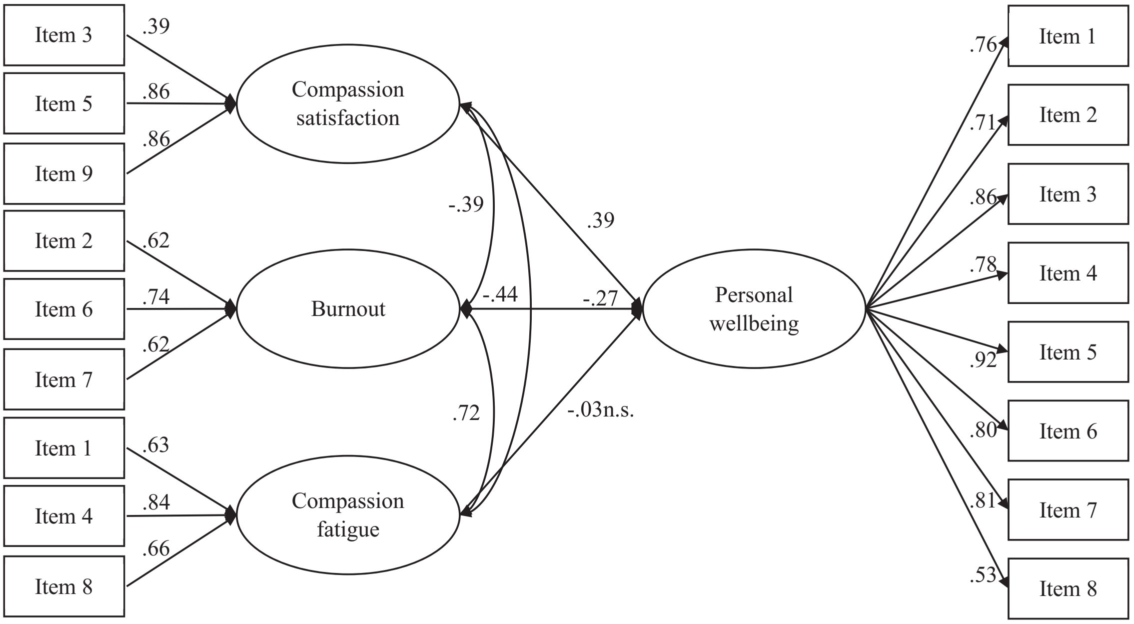

FIGURE 1 | Standardized model results of the structural equation model. All the factor loadings, effects, and correlations were statistically significant ($p < 0.001$), except for the one marked with n.s. ($p > 0.050$). For the sake of clarity, errors are not shown.

Regarding marital status, statistically significant differences were found, in this case, favoring married when compared to single. This is in line with previous studies using either the PWI (Van Beuningen and De Jonge, 2011) or other instruments (Yu et al., 2020). Being married, then, seems to function as a protector of wellbeing, beyond instruments and populations, including the palliative care context.

The last sociodemographic variable studied was profession. Our results indicated no difference in wellbeing levels across professions. Previous studies have pointed different results. For instance, in the work of Ta'an et al. (2020), physicians showed higher levels of depression, anxiety, stress, and occupational stress compared to nurses. However, in the study of Bettinsoli et al. (2020), nurses reported somewhat worse mental health compared to physicians and other hospital staff, although differences were small. Specifically, in the palliative care context, and regarding professional quality of life, clinicians have displayed higher levels of compassion satisfaction, whereas nurses have shown higher levels of compassion fatigue, and no differences in burnout have been found (Slocum-Gori et al., 2013). Again, results on the relationship between wellbeing, professional quality of life, and professional affiliation are not clear, and further evidence would be welcomed.

Last but not least, results regarding the relationship between wellbeing and professional quality of life conformed to previous literature (Koh et al., 2015; Sansó et al., 2020). Professional quality of life arose as an important variable in healthcare professionals' wellbeing, with higher levels on compassion satisfaction and lower levels on burnout predicting higher levels of wellbeing. Palliative care professionals' wellbeing, beyond its relationships with variables such as medical errors, sick leaves, and absenteeism (Pipe et al., 2009), or better quality of care (Salyers et al., 2017), should be one of the tasks of healthcare systems. The fact that professional quality of life explained almost one third of wellbeing of palliative care professionals makes us emphasize the importance of the quality of the helpers' work, not only for patients, but also for their personal wellbeing.

Limitations of this work include, first, that modest sample size could limit the representativeness and generalizability of our findings. Second, we did not study the content or face validity of the PWI items. Although this work was previously done (Van Beuningen and De Jonge, 2011), no specific analyses were conducted to test this in palliative care professionals. Other limitations include, for example, not having evaluated the years of professional experience of the participants. Additionally, it must be borne in mind that the survey was undertaken before coronavirus disease (COVID) pandemics, and consequently further research in current circumstances may be needed to generalize our results.

In conclusion, this work points out the adequateness of the PWI to measure personal wellbeing in Spanish healthcare professionals, specifically in palliative care workers. The instrument showed adequate internal structure, with one factor representing the eight items of personal wellbeing, excellent evidence of reliability, and the expected relations with other variables such as professional quality of life. Palliative care professionals with higher levels of wellbeing were married and showed higher levels of compassion fatigue and lower levels of burnout, but there was not a statistically significant association between the level of wellbeing and variables such as age, gender, or profession.

Implications of this work are clear, as it opens a new area of study. It is important to take care not only of patients' needs but also of professionals' ones. We can no longer afford to ignore the great importance of ensuring adequate wellbeing in healthcare professionals, especially when we know the direct effect on patient safety, among other effects. It is well known that most palliative care patients are facing the end of their own life, so it is a moral imperative not to add more avoidable suffering. Therefore, the study of their wellbeing should be on the agenda for the forthcoming research, and the PWI could be a good instrument to do that.

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 Ethics Research Committee at the University of the Balearic Islands. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

LG, GV-B, and NS designed the questionnaire and were involved in the data collection. LG and IF carried out the analysis. SP-B, LG, and IF interpretation of the data was discussed among. SP-B, LG, and NS drafted the manuscript. All authors were involved in the critical revision of the manuscript and approved the final version of the manuscript.

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Explaining Caregivers' Perceptions of Palliative Care Unmet Needs in Iranian Alzheimer's Patients: A Qualitative Study

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Introduction: The needs of Alzheimer's patients are very complex and diverse and many of them are considered unmet needs. Understanding and responding to the unmet and complex needs of Alzheimer's patients can affect the quality of care. Therefore, the present study aimed to explain the perception of formal and informal caregivers of the unmet needs of Iranian Alzheimer's patients.

Methods: The present qualitative study employed a Directed Content Analysis approach and was conducted in Iran in 2020. This research was done through in-depth and semi-structured interviews with 19 qualified caregivers enrolled (11 informal caregivers and 8 formal caregivers) with the mean age of 46.05 ± 10.98 years in a purposive sampling method. Interviews continued until data saturation. Data analysis was performed simultaneously with data collection. After recording and transcribing, the data were analyzed using the Elo and Kyngas method based on the National Consensus Project framework (NCP). Data management was done with MAXQDA software. In order to achieve the accuracy and validity of the study, the criteria presented by Lincoln and Guba Credibility, Dependability, Confirmability, and Transformability were considered and used.

Results: The mean age of participants was 46.05 ± 10.98. Participants in this study acknowledged that Alzheimer's patients need comprehensive needs management with a holistic approach to increase quality of life. This theme based on the NCP framework consists of seven main categories including "Structure and processes of care," "Physical aspects of care," "Psychological and psychiatric aspects of care," "Social aspects of care," "Cultural aspects of care," "Care of the patient nearing the end of life," and "Ethical and legal aspects of care" with 16 subcategories.

Conclusion: The findings of this study provide a deep understanding of the unmet needs of Alzheimer's patients in Iran. Identifying the unmet needs of patients can pave the way for the treatment team to provide effective solutions to meet the needs and empower caregivers to provide comprehensive care for patients.

Keywords: unmet need, palliative care, Alzheimers' disease, caregiver, Iran

INTRODUCTION

The growth of elderly population in the world has resulted in a considerable increase in the incidence of disabling disorders and cognitive problems (Farhadi et al., 2018). One of the most prevalent disorders accompanied by serious and progressive disability during old ages is Alzheimer's disease, which is the most common type of dementia (Craik and Salthouse, 2011; Ministry of Health, 2016). This disease is recognized by a variety of problems, such as amnesia, aphasia, apraxia, agnosia, and executive function disorder (American-Psychiatric-Association, 2013). The number of patients with Alzheimer's disease and other types of dementia in the world increased by nearly 117% from 1990 to 2016. Accordingly, the number of these patients increased from 20.2 million in 1990 to 40.8 million in 2016 (Alzheimer's-Association, 2018), and the prevalence of this disorder has been reported to be 3.6% in the Middle East (Adlimoghaddam et al., 2018). In Iran, it has been estimated that 700,000 elderly individuals suffer from Alzheimer's disease and that every 11.5 min, one person develops the disease (Salehi, 2011; Assocciation-Alzheimers-Iran, 2018).

Patients with Alzheimer's disease have various needs, such a way that their needs in the primary stage of the disease are quite different from those at the third and fourth (end) stages. Considering the incurable nature of Alzheimer's disease, it requires a long-term care approach in form of palliative care (Connor and Sepulveda Bermedo, 2020). Palliative care aims at meeting the needs of patients with incurable diseases, controlling pain, supporting patients' physical, mental, spiritual, and social needs, and improving patients' and their families' quality of life (Smith et al., 2012). Moreover, palliative care approach is compatible with the goals of person-centered dementia care. However, studies have revealed weak care standards among many patients with advanced Alzheimer's disease. In addition, no high-quality evidence is available for supporting palliative care approaches among these patients (Sampson, 2010).

Perceiving and meeting the needs of patients with Alzheimer's disease can directly affect care provision. Nonetheless, numerous studies have indicated that a large number of these patients' needs have remained unmet or have been met inappropriately (Cohen-Mansfield et al., 2015; Black et al., 2019; Mazurek et al., 2019; Sandman and Hofmann, 2019). Unmet needs could lead to a lower quality of life, higher levels of depression, worsening of neuropsychiatric symptoms, and increased disruptive behaviors (Hancock et al., 2006; Cohen-Mansfield et al., 2015). According to one study the major needs of elderly patients with Alzheimer's disease were the need for being heard, increase of knowledge level, and promotion of health (Bossen et al., 2009). Nevertheless, another research demonstrated that caring for patients with Alzheimer's disease was mainly focused on the physical dimension. In other words, these patients were provided with daily care services similar to other patients, while other dimensions of their needs were neglected by caregivers (Yektatalab et al., 2013). Hence, professional health care and family caregivers (formal and informal caregivers) are recommended to understand the needs of patients with Alzheimer's disease to be able to better find strategies to meet their needs.

Since perceptions and behaviors are shaped by culture, these needs and strategies for meeting them can be different depending on individual, cultural, and social differences (Zamanzadeh et al., 2014). In such countries as Iran, due to the existence of strong relationships among family members, patients with Alzheimer's disease are mainly cared by their own family members at home, also there is no codified plan to provide palliative care services, care is provided only in a limited number of centers, and patients do not receive the full range of essential care and training such as nursing care, physician consultation, and other care related to social, psychological, physical, or spiritual needs. Providing care, pain relief, and management of other symptoms in untreated patients by family members puts a lot of pressure on them (Rassouli and Sajjadi, 2016). Thus, the pressure imposed on patients and their family caregivers can result in a familial crisis. In this context, effective participation of formal caregivers in planning for a comprehensive care is of utmost importance, because they play a critical role in providing an appropriate environment as well as high-quality care for these patients (Yektatalab et al., 2013). Care dynamic in Alzheimer's disease requires taking individual, familial, caregiver, and social factors into account. Therefore, identification of formal and informal caregivers' viewpoints regarding patients' needs provides the ground for accurate evaluation of care services. Qualitative research is the best method for investigation of human phenomena and assessment of various perspectives, because human, social, cultural, and relational dimensions and values cannot be completely explored through quantitative approaches (LoBiondo-Wood and Haber, 2014). Moreover, qualitative studies can be used for in-depth description of unknown or less known phenomena from the perspective of individuals experiencing them in various cultures (Elo and Kyngäs, 2008). To the best of our knowledge, the available evidence has not well-explained the needs of patients with Alzheimer's disease. Hence, the present study aims to determine the perceptions of unmet palliative care needs among caregivers of patients suffering from Alzheimer's disease.

MATERIALS AND METHODS

Study Design

This study was conducted using a directed content analysis approach in order to determine the perceptions of unmet palliative care needs among the caregivers of patients with Alzheimer's disease. This qualitative research aimed at development of a theoretical framework, because the existing knowledge regarding the intended phenomenon was not comprehensive and required further explanation (Elo and Kyngäs, 2008).

Study Setting and Participants

The study participants included 11 informal caregivers of patients with Alzheimer's disease as well as eight formal caregivers, including physicians and nurses with the mean age of 46.05 ± 10.98 years. Eighty-four percent of the participants were female and the other 16% were male (**Table 1**). The participants were selected via purposeful sampling by referring to hospitals, nursing homes, and active branches of Alzheimer's Association in some provinces.

The inclusion criteria for the family caregivers were being able to speak Persian, aging at least 18 years, being the patient's first-degree relative, having been responsible for taking care of the patient for at least 6 months, not suffering from psychiatric disorders, and being willing to express one's experiences regarding taking care of a patient with Alzheimer's disease. Healthcare providers with at least 1 year of experience in provision of healthcare services for elderly individuals with Alzheimer's disease were also invited to take part in the research. The study was conducted in the participants' workplaces in order to investigate the intended phenomenon in the natural setting.

Data Collection Procedure

The study data were collected using in-depth, semi-structured interviews from April 2020 to January 2021. Each interview lasted for 20–45 min. The interviews were continued until data saturation when the new data did not develop or modify the theory, change the existing categories, or provide suggestions for creating a new category (Gustavsson, 2007). After all, 22 interviews were conducted with 19 participants. The interviews with family caregivers were begun with the following question: "can you describe a normal day of taking care of your patient." The interviews with formal caregivers were also begun with the following questions: "what services are usually demanded by patients and their families" and "which needs of patients and their families have not been met by service providers." The main framework of interview questions is based on a conceptual framework such as different dimensions of care as well as end-of-life care. Then, probing questions, such as "can you explain more" and "can you provide an example," were used for further investigations.

Research Framework

Due to the variety of symptoms and the long and complicated process of Alzheimer's disease, management of this disease can impose huge expenditures on families (Harris, 2007). Hence, these patients require palliative care (van der Steen et al., 2014). This type of care was initially used for patients with cancer and HIV/AIDS. Afterwards, dementia, Alzheimer's disease, and other cognitive disorders were also added to the groups requiring palliative care (Batiste and Connor, 2017).

The conceptual framework of the present study was based on the National Consensus Project (NCP). In this project, palliative care has been defined as the activities focused on management of pain and other symptoms, evaluating and meeting caregivers' needs, and coordination of care, leading to physical, functional, mental, practical, and spiritual outcomes of a serious disease. Accordingly, palliative care is a personal and family-based attitude toward care, helps relieve people with serious diseases who suffer from the related stress and symptoms, and promotes the quality of life of patients and their families. The NCP has considered eight care dimensions for patients with restrictive and incurable disorders and has identified palliative care guidelines in these dimensions. These eight dimensions include the structure and process of care, physical dimensions of care, psychological dimensions of care, social dimensions of care, spiritual, religious, and existential dimensions of care, cultural dimensions of care, taking care of a dying patient, and ethical and legal dimensions of care (Ferrell et al., 2018).

Data Analysis

The data were collected and analyzed simultaneously. Data management was done using the MAXQDA software (Kuckartz and Rädiker, 2019), and data analysis was done using the method proposed by Elo and Kyngas in preparation, organization, and reporting stages (Elo and Kyngäs, 2008; Elo et al., 2014). In doing so, each interview was read several times, so that the researcher gained an overall understanding of the content. Then, the meaning units were identified and the primary codes were extracted. After that, similar primary codes were classified in more comprehensive categories, and the main categories were extracted. Finally, all directed content analysis stages and results were abstracted and reported. Qualitative data analysis based on the method proposed by Elo and Kyngas has been presented in **Table 2**. In addition, an example of data analysis has been shown in **Table 3**.

Rigor

In order to evaluate the qualitative data, use was made of the criteria proposed by Lincoln and Guba (Guba and Lincoln, 1994). Data credibility was ensured through prolonged engagement with the data (10 months), continuous observation, peer review, interaction among researchers, reviewing the interview transcripts by the participants, and combination of several methods (interview and observation). Considering transferability, the participants' features and experiences were described in details. In addition, the reliability of the data was ensured via accountability. In other words, research process, data collection and analysis, coding, and formation of categories were reviewed by an external observer who was familiar with qualitative studies. Finally, all research processes were recorded and presented extensively to ensure confirmability.

Ethical Consideration

This study was approved by the Ethics Committee of Ahvaz Jundishapur University of Medical Sciences (IR.AJUMS.REC.1398.781). The ethical considerations in this study included the voluntary nature of participation in the research, provision of the participants with explanations about the study objectives, obtaining written informed consent forms, confidentiality of the participants' information, anonymity, the participants' right to withdraw from the study, and trustworthiness in using information and resources.

RESULTS

This study was conducted on 19 participants with the mean age of 46.05 ± 10.98 years. The participants' characteristics have been presented in **Table 1**.

TABLE 1 | Demographic characteristics of the participants in the study.

Number	Participate	Age(year)	Gender	Marital status	Level of education	Job	Ratio with caregiver	Stage of patient
1	Formal caregivers	58	Male	Married	Fellowship	Neurologist	Patient	Stage 1–4
2	Formal caregivers	38	Female	Married	Ph.D.	Doctor	Patient	Stage1–4
3	Formal caregivers	54	Female	Married	Fellowship	Faculty member	Patient	Stage1–4
4	Formal caregivers	37	Female	Married	Ph.D.	University professor	Patient	Stage1–4
5	Formal caregivers	38	Female	Married	Ph.D.	University professor	Patient	Stage1–4
6	Formal caregivers	47	Female	Married	Ph.D.	University professor	Patient	Stage1–4
7	Formal caregivers	43	Female	Married	Master sciences	University professor	Patient	Stage1–4
8	Formal caregivers	32	Female	Married	Master sciences	Nurse	Patient	Stage1–4
9	Informal caregivers	48	Female	Married	Diploma	Housewife	Father	Stage 3
10	Informal caregivers	71	Male	Married	Bachelor	Civil Engineer	Spouse	Stage 3
11	Informal caregivers	35	Female	Married	Bachelor	Housewife	Mother	Stage 2
12	Informal caregivers	52	Female	Single	Ph.D.	Nurse	Father	Stage 3
13	Informal caregivers	45	Female	Single	Master sciences	Teacher	Father	Stage 3
14	Informal caregivers	56	Female	Married	Illiterate	Housewife	Father	Stage 3
15	Informal caregivers	45	Female	Married	Diploma	Housewife	Father	Stage 4
16	Informal caregivers	65	Male	Married	Diploma	Housewife	Father	Stage 2
17	Informal caregivers	35	Female	Married	Diploma	Housewife	Mother	Stage 4
18	Informal caregivers	33	Female	Married	Illiterate	Housewife	Father	Stage 4
19	Informal caregivers	43	Female	Married	Illiterate	Housewife	Father	Stage 4

TABLE 2 | Qualitative data analysis process based on Elo Kyngas method.

Preparation stage	Select the analysis unit	After converting the interviews to text format, explicit content (same as the text of the interviews) and hidden content (non-verbal behavior of the participants) were analyzed and semantic units were identified.
	Find the logical connection of the data with the whole subject	The text of the interview was read several times by the researcher in order to gain a continuous and prolong engagement with the data.
Organizing stage	Create an analytical matrix	The structure and process of care, the physical aspect of care, the Psychological and psychiatric aspects of care, Care of the patient nearing the end of life, the social aspect of care, the cultural aspect of care, and the ethical-legal aspect of care were included as the main classes in the non-imposed matrix.
	Extract data from content based on categories	The main classes were formed based on conceptual and logical relationships with other classes, and finally the classes were identified based on the research framework.
	Grouping	The number of codes decreased by merging similar codes based on their differences and similarities into more general codes.
	Classification	The formed groups were classified based on their differences and similarities (merging similar groups).
	Abstraction	The revealed classes were placed in the main and primary classes of the analytical matrix.
Reporting stage	The sampling process, participants' characteristics, data collection, data analysis, and each of the main classes were reported in detail in the findings section.	

The codes extracted from the interviews revealed one main theme; i.e., "needs management with a holistic approach," which was in accordance with the NCP framework and consisted of seven main categories, namely "structure and process of care," "Psychological and psychiatric aspects of care," "Physical aspects of care," "Care of the patient nearing the end of life," "social aspects of care," "cultural aspects of care," and "ethical and legal aspects of care." Detailed information about the main categories and subcategories has been provided in **Table 4**.

Needs Management With a Holistic Approach

In this study, the participants believed that patients with Alzheimer's disease required needs management with a holistic approach in order to enhance their quality of life. This was in accordance with the NCP framework and contained seven categories and 16 subcategories. This theme referred to the care structure and process for management of physical, psychological, and legal problems, resolution of cultural, ethical, and social issues, reception of extensive support from families

TABLE 3 | An example of data analysis.

Theme	categories	subcategories	Primary codes	quotation
Social aspect of care	*Need for support on the part of the government and social systems*	*Need for support on the social systems*	**Establishment of supportive group groups** **Training through supportive group**	**We can use support groups to teach family and patient support (geriatrician).**

TABLE 4 | Themes, main categories, and sub-categories resulting from the content-driven analysis of the interviews.

Themes	Main categories	Sub-categories
Needs management with a holistic approach	Structure and process of care	Need for development of diagnostic and screening services Need for specialized care services Need for home care services Need for hospice services
	Psychological and psychiatric aspects of care	Need for Psycho-emotional needs management Need for Psycho-cognitive needs management
	Physical aspects of care	Need for controlling progressive physical symptoms Need for controlling the secondary complications of treatment
	Care of the patient nearing the end of life	Need for comfort care Need for selecting a preferred place for death
	Social aspects of care	Need for support on the part of the government and social systems Need for empowerment of informal caregivers
	Cultural aspects of care	Need for destigmatization Need for cultural interventions
	Ethical and legal aspects of care	Need for ethical care Attention to patients' undeniable rights

and social and governmental institutions, and empowerment of family caregivers.

Structure and Process of Care

This dimension included the assessment of palliative care, care program, and specific systems and processes for palliative care. This category consisted of four subcategories, namely "need for development of diagnostic and screening services," "need for specialized care services," "need for home care services," and "need for hospice services."

Need for Development of Diagnostic and Screening Services

Based on the study participants, diagnosis of Alzheimer's disease at the primary stages could impede the progression of the disease, which was considered one of the most effective strategies for preventing the incidence of the resultant disabilities. In this context, the participants referred to the differential diagnosis of Alzheimer's disease, prioritization of early diagnosis and identification of patients, and utilization of advanced tests for early screening of patients' problems including malnutrition and sleep disorders. For instance, one of the formal caregivers (geriatrician) maintained:

> *"Patients have to be screened in different areas before the incidence of further problems; for example, regarding old age, malnutrition, cognitive disorders, and sleep."*

Need for Specialized Care Services

"Need for specialized care services" was emphasized by the current study participants. This category referred to the required specialized care services, such as specialized rehabilitation services, empowerment of mental and cognitive status, psychiatric services, pharmacological treatments, visits by different specialists for controlling secondary problems, and provision of care services by day care, night care, and outpatient care centers. Considering the importance of pharmacotherapy, one of the formal caregivers (nurse) stated:

> *"Pharmacotherapy is needed for patients in the primary phases and mild to moderate stages of the disease and reduces the speed of disease progression."*

Regarding the importance of continuous visits by different specialists, one of the patients' spouse said:

> *"In order to control the secondary problems, patients have to be visited by different specialists. In addition, patients have to be visited by a neurologist on a regular basis."*

Need for Home Care Services

Home care, as a society-based care method, is one of the effective models in care provision that plays a key role in provision of comprehensive care services for patients with chronic diseases, including those suffering from Alzheimer's disease. In this disease, patients face challenges in doing their daily activities and as time goes by, a trained caregiver has to be with them at home or a nurse has to visit them at home on a regular basis. This category emphasized the necessity to design and develop home care services and to monitor the patient care process. In this respect, one of the patients' daughter mentioned:

> *"We need centers for home care… A new structure should be designed… We do not have a hospitalized patient… They are not patients referred to health centers. We have an elderly patient who has a problem and is at home and is living in the society."*

Need for Hospice Services

A large number of participants believed that due to the complex and variable conditions of patients with Alzheimer's disease who are in fact in the end stage of life, home care is not possible and they should be transferred to end-of-life care centers. In this regard, formal caregivers stated that the care services for patients at the end stage of life as well as their families had to be provided in form of palliative care in hospice centers. For instance, one of the formal caregivers (geriatrician) said:

> *"The important point is that taking care of these patients at home is impossible in the long run, it's really impossible. It's illogical to expect families to take care of these patients until the last stages. There should be a place, so that these patients can be separated from their families in the last stages."*

Psychological and Psychiatric Aspects of Care

This dimension involved the psychological and psychiatric aspects of care in the field of Alzheimer's disease and consisted of two subcategories; i.e., "need for psycho-emotional needs management" and "need for psycho-cognitive needs management."

Need for Psycho-Emotional Needs Management

All study participants believed that patients with Alzheimer's disease experienced various psycho-emotional disorders, including mood disorders, isolation, depression, anxiety, restlessness, behavioral disorders, aggression, confusion, and violent behaviors. These problems have to be identified in order to understand and respond to patients' needs, eventually providing the ground for providing them with comprehensive care services. In this respect, one of the patients' daughter stated:

> *"For example, now he himself asks me why he is like this. He is depressed. He is sad. He says that he was not like this before. This is one of the features of Alzheimer's disease. They become depressed first and then, the depression causes them not to be able to eat or sleep and weakens their physical and mental statuses."*

Need for Psycho-Cognitive Needs Management

The major complaints of the caregivers of elderly patients with Alzheimer's disease were related to cognitive issues, forgetting daily affairs, and inability to forge relationships with their families and other people. Since many cognitive deficiencies cannot be recovered in these patients, rehabilitation techniques are required to compensate for the deficiencies and to enhance the quality of life of patients and their families. In this context, one of the patients' daughter talked about her mother:

> *"She got Alzheimer's disease little by little… Now, she just remembers her childhood memories, her old memories. Her mental and cognitive status even gets worse over time. She cannot build relationships, particularly with her family members… This is our problem. She cannot build relationships with us and she asks a lot of questions… You explain, but they ask the same questions."*

Physical Aspects of Care

In this dimension, evaluation of palliative care, care planning, and treatment of physical symptoms were described by focusing on comprehensive patient care. This category included two subcategories, namely "need for controlling progressive physical symptoms" and "need for controlling the secondary complications of treatment."

Need for Controlling Progressive Physical Symptoms

The study participants stated that patients with Alzheimer's disease suffered from physical symptoms, including inability to do their daily activities (taking a bath and going to the toilet), loss of speaking ability, inability to stand up and walk independently, vulnerability to pain, urinary incontinence, weakness, cachexia, respiratory problems, panting, difficulty in swallowing, pressure ulcer, febrile infections, indifference to food, nausea, and vomiting, which had to be managed. In this regard, one of the patients' daughter said:

> *"For example, my mother used to forget time and place at the beginning. We were most bothered by changes in her sleeping hours; she was awake all through the night, but slept throughout the day. Little by little, she was not able to do her daily chores, and someone had to be with her."*

Need for Controlling the Secondary Complications of Treatment

Due to several pharmacological treatments, the disease complications are intensified in patients and the recovery process is slowed down. In the current study, some participants emphasized that unwanted complications occurred in patients with Alzheimer's disease because of the simultaneous consumption of different medications, suffering from different diseases at the same time, and reduction of body systems' functions. For instance, one of the patients' daughter mentioned:

> *"After taking sleeping pills, my father had a fall and had a femoral head fracture. He didn't get well afterwards. We could interact with each other before that incident, but we can't now."*

Care of the Patient Nearing the End of Life

This dimension emphasized the common symptoms and conditions in the last days and weeks of life, and consisted of two subcategories; i.e., "need for comfort care" and "need for selecting a preferred place for death."

Need for Comfort care

This type of care refers to improving the quality of life through relieving pain and other painful factors and providing practical, emotional, and spiritual support by focusing on patients' culture. In this study, a large number of caregivers believed that patients at the end stage of life needed different care services. These patients and their families needed tranquility, as well. One of the patients' daughter stated:

> *"My father's condition was really complicated. He as well as all family members needed to be understood and supported."*

Another patient's daughter also said:

> *"When we can't do anything for our father, why should we bother him? Some painful measures should be disrupted, so that he will be comfortable."*

Need for Selecting a Preferred Place for Death

One of the dimensions of end-of-life care was ensuring the patient's physical comfort at the time of death and selection of a preferred place for death. It also included preparing the family at this highly emotional and uncertain time. In this regard, one of the patients' daughter maintained:

> *"When patients are at the end stages of life, they experience a terrible death process. The preferences and needs of the patients and their families have to be taken into account by the treatment team; the place in which the patient experiences a more comfortable death, not to be left alone."*

Social Aspects of Care

This dimension of palliative care referred to the evaluation and elimination of the social needs of the patients and their families. Based on the participants' statements, two subcategories were formed as follows: "need for support on the part of the government and social systems" and "need for empowerment of informal caregivers."

Need for Support on the Part of the Government and Social Systems

Support on the part of social systems and entrusting some responsibilities to a group of individuals who can provide those in need with emotional protection and support resources were among the needs extracted from the present study results. Almost all study participants mentioned that inappropriateness of the price of the required medications and equipment to the patients' financial ability, lack of insurance coverage, and high costs of screening and diagnostic tests had caused numerous challenges for the patients and their families. Therefore, in order to afford the costs of life, medications, and treatments, patients need to be supported by the government, social institutions, support groups, peers, active NGOs, and charities. These support resources help individuals feel cared for, loved, and valued, thereby increasing their self-esteem. Considering the importance of NGOs, one of the formal caregivers (physician) maintained:

> *"NGOs should be asked for help in this area. In Iran, we don't have active NGOs in the field of Alzheimer's disease. If we can establish such centers; this was started in some parts of the country, but was not completed."*

Need for Empowerment of Informal Caregivers

This part emphasized the necessity of informational, mental, and emotional support for caregivers, which was in line with empowerment of informal caregivers for facing problems and managing care. The study participants believed that entrusting care to the informal caregivers who did not have the required knowledge and experience for taking care of patients with Alzheimer's disease could result in the quick progression of the disease. Hence, they stated that families had to be provided with information and support in order to provide appropriate care services, manage and control the disease complications, develop communication skills with patients, and manage the crisis and stress. Regarding the caregivers' inability to provide proper care, one of the informal caregivers who was responsible for taking care of one's father mentioned:

> *"The family caregiver didn't know what to do with the patient's constipation, how to prevent pressure ulcer in the patient with Alzheimer's disease, what to do with the patient's dry joints. S/he didn't even know how to provide the intended position."*

Cultural Aspects of Care

This dimension dealt with the effective cultural factors in the provision of palliative care and provision of patients and their families with services. It consisted of two subcategories, namely "need for destigmatization" and "need for cultural interventions."

Need for Destigmatization

Stigma has been defined as the outcomes associated with unawareness, prejudgment, and discrimination about an issue. Generally, stigma causes families to suffer from emotional experiences, such as disrespect, indifference, and discrimination. Besides, fear from stigma results in secrecy. Some participants in this study mentioned that they tried to hide their patients since they were afraid of being teased by others. This feeling of shamefulness due to the patient's repeated questions and abnormal behaviors as well as others' judgments could be attributed to the association of this disease with mental disorders. In this respect, one of the informal caregivers stated:

> *"Others' behavior is what makes us sad…For example, when I took my mother to other places, others said that it was very hard and asked us how we took care of the issue and how we tolerated her…Shouldn't we tolerate her? She is our mother; she has done everything for us for a lifetime. Now, it's our duty."*

Need for Cultural Interventions

In this part, the participants remembered the cultural items, which were less taken into consideration in the society. In fact, culture is a concept, which is taught and transferred to individuals through socialization. In this dimension, emphasis was put on training through the mass media in order to increase awareness about aging and the associated challenges as well as modification of the society's attitude toward Alzheimer's disease (destigmatization). Regarding the importance of training and Culturalization, one of the formal caregivers (physician) maintained:

> *"Culturalization should be done. This does not occur simply. It should be taught through the radio, TV, and mass media."*

Ethical and Legal Aspects of Care

The contents of this dimension included planning for advanced care services, decision-making for sensitive issues, and ethical and legal considerations with a focus on the necessities and ethical processes for supporting patients' independence. This dimension consisted of two subcategories; i.e., "need for ethical care" and "attention to patients' undeniable rights."

Need for Ethical Care

One of the axes of the patients' rights charter is the respect for patients' and their families' right for selecting and making decision about receiving healthcare services. For instance, the participants expressed that patients and their families had to be involved in making decisions about their treatment. In addition, families had to give their patients the right to maintain their independence in doing their daily activities. In this context, one of the informal caregivers (a girl taking care of her father) said:

> *"During his operation, for example, I said a hundred times that he had Alzheimer's disease, but it seemed as if I was talking to no one. Or during his femur fracture operation, no one asked my opinion as the caregiver, as the nurse. They do not explain what they are going to do for the patient. Nobody asks about the patient's history and conditions."*

Attention to Patients' Undeniable Rights

This category referred to decision-making about patients' legal issues, including management of their possessions, writing a will, and selecting a guardian and a main caregiver. In this regard, some participants mentioned the legal issues occurred for patients with Alzheimer's disease, because these patients have judgments and decision-making disorders and, consequently, are prone to financial abuse on the part of their acquaintances. For instance, one of the formal caregivers (geriatrician) maintained:

> *"Patients face increasing problems in keeping their daily financial accounts. Therefore, elderly people should be provided with the related information. There may be resistance on the part of elderly individuals or their acquaintances; e.g., writing a will, announcing a person as the guardian at the severe stage of the disease. All these should be taught and individuals should decide who to be the main caregiver."*

DISCUSSION

Identification of unmet palliative care needs among patients with Alzheimer's disease and consideration of the complexity of these needs can help healthcare providers to provide them with the care services appropriated to their needs. The present study aimed to determine the perceptions of unmet palliative care needs among the caregivers of patients with Alzheimer's disease. The codes extracted from the interviews resulted in the emergence of the theme "Structure and processes of care," "Physical aspects of care," "Psychological and psychiatric aspects of care," " Social aspects of care," " Cultural aspects of care," "Care of the patient nearing the end of life," and "Ethical and legal aspects of care" with 16 subcategories.

As mentioned above, the theme extracted in the present study was management of patients with Alzheimer's disease using a holistic approach Comprehensive management refers to a value-based decision-making framework, which integrates all planning dimensions and is one of the major principles of palliative care. The results of numerous studies have emphasized the necessity to comprehensively manage the behavioral problems, personal, care, and emotional activities, and social needs of patients with Alzheimer's disease (Cadieux et al., 2013; Sinvani et al., 2018; Zucchella et al., 2018). The findings of a prior research indicated that the Severe Behavior Response Teams (SBRT) model was used for patients with dementia in Australia. This model puts emphasis on out-of-home services and the patient's living place. In this model, services are provided by a multidisciplinary team in response to crises. Interventions are initially focused on the evaluation and management of behaviors and are completed by training and supporting caregivers (Macfarlane and Cunningham, 2017). In Iran, some programs have been developed and started for dealing with Alzheimer's disease. However, there is no comprehensive approach for management of the needs of these patients. Moreover, lack of care centers and home care systems for taking care of patients with Alzheimer's disease are considered the main obstacles against comprehensive management of the disease in Iran. Hence, this approach is recommended to be taken into account (Yektatalab et al., 2013).

"Structure and process of care" was among the main categories extracted from the interviews. Provision of comprehensive care services requires structures to take the responsibility of providing a part of the services. This construct emphasizes screening and early diagnosis, and indicates that hospitalization of patients at the specialized level depends on diagnosis at lower levels and treatment, pharmacological, and rehabilitation services are provided on the basis of patients' needs. Home care services, as society-based services, were also included in this category. In case patients do not have complicated needs, home care services can be provided, which is of interest to a large number of patients and their caregivers. This type of care refers to reception of a wide range of care services appropriated to patients' needs. There is limited evidence regarding society-based home care services for supporting patients with dementia. Dawson et al. (2015) demonstrated that home care services were accompanied by desirable outcomes in case they were provided on time and were responsive, flexible, and appropriated to individual needs (Dawson et al., 2015). This category also referred to hospice centers for patients with Alzheimer's disease at the end stage of life. Hospice centers provide high-quality spiritual, mental, social, and physical services for patients and their caregivers. The findings of the research by Mitchell et al. (2007) revealed that hospice service providers could respond to the unique challenges of end-of-life care for patients suffering from dementia (Mitchell et al., 2007). The studies performed in Iran have also emphasized the need for establishment of hospice centers for provision of care services at the end stages of life. Despite the increasing demands for care services at the end stages of life due to the increasing incidence of chronic and life-threatening diseases in Iran, there are no hospice centers for the patients and families who require optimum care services (Azami-Aghdash et al., 2015). The most important reasons for the lack of hospice centers in Iran include the lack of a proper ground, lack of expert human workforce, lack of comprehensive care programs, lack of appropriate guidelines, lack of policies, lack of free access to opioids, financial problems, economic problems, and cultural differences (Assocciation-Alzheimers-Iran, 2018; Zarea et al., 2020).

"Psychological and psychiatric aspects of care" and "physical aspects of care" were two other categories extracted in the current investigation. In order to organize integrated healthcare services for meeting the needs of patients with Alzheimer's disease, the variety of symptoms and needs in this population should be taken into consideration (Farmer et al., 2016; Commisso et al., 2017). In fact, designing person-centered rather than disease-focused care services is only possible through perception and prioritization of patients' symptoms and problems (Curnow et al., 2019). The Behavioral and Psychological Symptoms of Dementia (BPSD) include a wide range of physical, emotional, psychological, and behavioral symptoms (Tible et al., 2017) whose management and treatment are challenging due to the complex etiopathogenesis of the symptoms as well as multiple complications. Management of BPSD involves an accurate diagnostic investigation, exploration of the causes of Alzheimer's disease, and elimination of other causes such as delusion related to drugs consumption, simultaneous treatment of other physical diseases, effective control of pain or infections, and providing patients and their families with psychosocial therapies (Hersch and Falzgraf, 2007; Tible et al., 2017). In Iran, management of BPSD symptoms is mainly focused on pharmacological interventions. Expert advice and guidance, prefer non-pharmacological interventions as a first-line approach although the evidence for most non-pharmacological strategies is weak, their effectiveness is confirmed by long-term clinical experience. Medication for BPSD is offered frequently, but carries the risk of serious side effects (Savaskan et al., 2014; Deuschl and Maier, 2016). Nonetheless, patients suffering from Alzheimer's disease are vulnerable to the side effects of medications. Hence, non-pharmacological interventions are recommended to be performed for better management of physical and psychological symptoms amongst these patients.

"Care of the patient nearing the end of life" was the fourth dimension extracted in the present research. Taking care of dying patients is a huge challenge, which is done in a challenging environment. In this dimension, patient care involved "comfort care" and "selection of a preferred place for death," which was provided comprehensively with a focus on the patient's body and mind (Gillan et al., 2014). Generally, end-of-life care aims at providing the ground for a comfortable death, maintaining balance in patients' daily lives, and consoling their families (Iranmanesh et al., 2010). In fact, end-of-life care is a key component of taking care of elderly individuals. Nevertheless, evidence has indicated that individuals aging above 85 years had little access to specialized end-of-life care services (Hunt et al., 2014). Furthermore, the findings of some studies have shown that pain control through the use of analgesics, management of symptoms, palliative care, and Do Not Resuscitate (DNR) and Do Not Hospitalize (DNH) guidelines were carried out less among patients with Alzheimer's disease in comparison to other patients (Mitchell et al., 2004; Sampson et al., 2006).

"Social aspects of care" was another main category in the current research. One of the subcategories of this dimension was the "need for support on the part of the government and social systems." Accordingly, patients with Alzheimer's disease required formal care support or access to care services to meet their needs (Ball et al., 2015; Smith et al., 2015; Hynes et al., 2016). Creating and taking part in support groups formally, informally, and voluntarily could be a proper measure for gaining knowledge about coping strategies as well as emotional and informational support (Rosa et al., 2010). Based on the reports provided by Prince (2004), patients with Alzheimer's disease in Iran received lower social support compared to those living in developed countries (Prince, 2004). These results were in agreement with those of the present investigation (Górska et al., 2018; Yang et al., 2020). In the Netherlands, for instance, a Meeting Centers Support Program (MCSP) is used as a supportive approach for patients with mild to moderate Alzheimer's disease who live in the society as well as for their caregivers. This approach is a combination of recreational and psychotherapeutic activities for patients, psychotherapy groups for caregivers, social activities for patients and caregivers, and holding regular sessions in treatment centers (Dröes et al., 2000; Farina et al., 2006). Another subcategory of this dimension was the "need for empowerment of informal caregivers." The results of a previous research indicated that in case caretaking was carried out continuously by caregivers, lack of support programs on the part of the government, society, and family members could increase the burden of the caretaking responsibility and the probability of fatigue in the main caregivers, which might lead to misbehavior toward patients with Alzheimer's disease (Mohamadi et al., 2008). In another study, emphasis was put on emotional, mental, and informational support for caregivers. In addition, empowerment of caregivers for facing the problems associated with taking care of elderly patients as well as management of family caretaking were considered important (Mohammadi, 2008). In spite of the need for social support, there are no support programs, including financial, mental, and social support programs, on the part of government organizations, peers, families, and the society for patients with Alzheimer's disease in Iran.

Another category extracted in the present research was "cultural aspects of care," one of whose subcategories was the "need for destigmatization." Generally, diagnosis of Alzheimer's disease, similar to mental disorders, can be accompanied by considerable stigma, which may be attributed to the cultural beliefs about the etiology of the disease as well as to the patients' unsocial behaviors that can result from cognitive disorders

(Mukadam and Livingston, 2012; Navab et al., 2019). In this study, the caregivers' experiences indicated that they were afraid of being teased by their acquaintances as well as by the society and, as a result, attempted to hide the patients. In general, people living in collectivist cultures like the Iranian culture pay great attention to their relationships with other individuals. When patients are not able to build effective relationships with their caregivers and other individuals, their membership in the society will be questioned. Hence, these patients need to be accepted by their family members, the society, and the healthcare system (Yektatalab et al., 2013). Therefore, it is necessary to carry out cultural interventions, including training through the mass media, in order to improve the society's attitude toward these patients and their families. The results of a study explained that stigma among patients with Alzheimer's disease could be reduced via Culturalization, training, protesting against the existing inequities, and forging relationships with these patients (Mukadam and Livingston, 2012). Edney also disclosed that mass media could exert a great impact on people's belief systems. Thus, showing mental disorders and Alzheimer's disease was reported to be effective in the perception of the disease (Edney, 2012).

The last category extracted in the current study was the "ethical and legal dimensions of care." Since Alzheimer's disease has a progressive nature and patients' abilities are considerably decreased at each stage, adhering to ethical and legal considerations is essential in each stage of the disease. Khan Ahmadi et al. (2015) mentioned the ethical considerations related to patients with Alzheimer's disease in form of independence, care, participation, self-fulfillment, and dignity principles in three sections (care, treatment, and research) at the three stages of the disease (beginning, middle, and final) (Khan Ahmadi et al., 2015). In spite of the rich Iranian culture that values respect toward elderly people, the ethical and legal principles for this group are not taken into account nowadays. In some countries like England and Wales (2005), however, the Mental Capacity Act has been approved and executed for solving the ethical and legal problems of patients with Alzheimer's disease. This program provides these patients with a framework to decide for themselves and to choose representatives for making decisions about their behaviors. This includes basic decisions about care, treatment, financial issues, and daily affairs (Kritika Samsi, 2012). Unfortunately, these issues are not common in Iran and even a power of attorney can be revoked in case insanity is diagnosed in any of the parties. This is in fact a legal gap, which requires quick modification.

STUDY LIMITATIONS

Even though qualitative studies show the participants' in-depth experiences, they may suffer from the non-generalizability of the results. One of the limitations of the current study was that most of the participants were female. Another significant study limitation was related to the selection of caregivers in the research setting, which was faced with problems due to the COVID-19 pandemic.

CONCLUSION

The study findings demonstrated that patients with Alzheimer's disease suffered from numerous physical, mental, emotional, and cognitive problems. In addition, disease symptoms and complications, lack of a holistic and comprehensive care system, social stigma, and lack of acceptance by the society, cultural, ethical, and legal issues associated with the disease, lack of support resources, and the economic burden of the disease caused patients and their families to face various challenges. Thus, paying attention to patients' unmet needs could result in beneficial effects for both patients and their informal caregivers. These needs can be perceived by determining the experiences of formal and informal caregivers. This issue is of particular importance in Iran, which is faced with the aging phenomenon. Overall, identification of patients' unmet needs can help health systems to take effective measures for meeting those needs and strengthening caregivers for providing patients with comprehensive care services.

DATA AVAILABILITY STATEMENT

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

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Ethics Committee of Ahvaz Jundishapur University of Medical Sciences (IR.AJUMS.REC.1398.781). The ethical considerations in this study included the voluntary nature of participation in the research, provision of the participants with explanations about the study objectives, obtaining written informed consent forms, the confidentiality of the participants' information, anonymity, the participants' right to withdraw from the study, and trustworthiness in using information and resources. The patients/participants provided their written informed consent to participate in this study. Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article.

AUTHOR CONTRIBUTIONS

MG, HA, and MR in critical revisions for important intellectual content and administrative/technical support and supervised the work. All authors involved in the study conception, design and contributed to the data collection, and analysis.

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Preferred Place of Death in Adult Cancer Patients: A Systematic Review and Meta-Analysis

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Background: Identifying the preferred place of death is a key indicator of the quality of death in cancer patients and one of the most important issues for health service policymakers. This study was done to determine the preferred place of death and the factors affecting it for adult patients with cancer.

Methods: In this systematic review and meta-analysis study four online databases (PubMed, Scopus, web of science, ProQuest) were searched by relevant keywords. Quality assessment of papers was conducted using Newcastle-Ottawa (NOS) criterion. Odds ratios, relative risks, and 95% confidence intervals were determined for each of the factors extracted from the investigations.

Results: A total of 14,920 participants of 27 studies were included into the meta-analysis. Based on the results, 55% of cancer patients with a confidence interval [95% CI (41–49)] preferred home, 17% of patients with a confidence interval [95% CI (−12%) 23)] preferred hospital and 10% of patients with confidence interval [95% CI (13–18)] preferred hospices as their favored place to die. Effective factors were also reported in the form of demographic characteristics, disease-related factors and psychosocial factors.

Conclusions: This study showed that more than half of cancer patients chose home as their preferred place of death. Therefore, guided policies need to ensure that the death of the patients in the preferred place should be considered with priority.

Systematic Review Registration: https://www.crd.york.ac.uk/prospero/display_record.php?ID=CRD42020218680, identifier: CRD42020218680.

Keywords: palliative care, end of life, cancer, hospice, preferred place of death, terminally ill, hospital, supportive care

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INTRODUCTION

Cancer is considered the second leading cause of death worldwide. In 2018, about 9.6 million people of the world have died by cancer, which was almost one in six worldwide deaths (WHO, 2021). Of these, approximately 1.6 million patients did not die in their preferred place (Kern et al., 2020). Patients with advanced cancer show that despite the fact that the majority of cancer patients prefer to die in the preferred place (Neergaard et al., 2011; Hyun et al., 2013; Gomes et al., 2015; Vidal et al., 2020), a small number of these patients die in the preferred place (Chen et al., 2014; Howell et al., 2017).

The place of death has passed through three evolutionary periods in societies. In the first period, people often died at home due to poor access to health resources. Secondly, deaths in hospitals have increased due to advances in the health system, but in the last decade, the emphasis has changed to the quality of end-of-life care, and by developing home care, it has been emphasized to people's preferences to die at home. During different periods, social norms play a role in determining the place of death (Gu et al., 2007).

The place of death is very important in the allocation of medical resources and has recently received a lot of attention from palliative care specialists (Hyun et al., 2013; Cabañero-Martínez et al., 2019). In recent decades, this place has been a hospital for the majority of cancer patients (Alonso-Babarro et al., 2011; Gomes et al., 2015). In fact, the actual place of death is 10 to 35% of patients at home and 50 to 60% occurs in the hospital (Bell et al., 2010).

The preferred place of death (PPOD) means the desire of people to die in one place or death of people in a favorite place (Yamagishi et al., 2012). Over the last few decades, the ability to select and control the PPOD has increasingly been considered as a key indicator for increasing the quality of death and one of the criteria of a good death (Ali et al., 2019); Therefore, health care policymakers attribute great importance to determining PPOD. Focusing on PPOD increases the quality of end-of-life care, respect for patients' preferences, as well as proper distribution of health and medical resources to successfully implement palliative care (Gu et al., 2015; Ali et al., 2019). In a systematic review, Bell et al. studied the correlation between the preferred and actual place of death (Bell et al., 2010). Gomes et al. (2013) In a systematic review of meta-analysis, reveal that not only the majority of cancer patients but also other patients with life-threatening issues choose the home as their preferred place of death. The use of home-based end-of-life care has very beneficial effects on the physical, psychological, social, and economic dimensions of patients. Also it reduces the costs of the health system and hospital complications, shortens the length of hospitalization, and prevents re-hospitalization of patients. Also, this method of care can facilitate the continuity of care after discharge and patients benefit from the facilities of different centers (Kerr et al., 2014; Lustbader et al., 2017; Heydari, 2018). However, the home is not always the best place to die because access to home-based palliative care and care services varies from country to country (Chen et al., 2014). Due to the different PPOD, the present study was conducted to determine the PPOD and the factors affecting it in adult patients with cancer.

METHODS

Study Design

This systematic Review study was performed based on the guidelines of Preferred Reporting Items for Systematic Reviews and Meta-Analyzes (PRISMA) (Liberati et al., 2009). Also was approved by the ethics committee of Baqiyatallah University of Medical Sciences (Ethics code: IR.BMSU.REC.1399.425). The study protocol is registered in PROSPERO with the code CRD42020218680 (the access link:https://www.crd.york.ac.uk/prospero/display_record.php?ID=CRD42020218680).

Type of Study and Participants

Observational studies including cross-sectional, case-control, cohort that explicitly state the PPOD of cancer patients or provide data to calculate this index have been selected. Review articles, case reports, case series, and clinical trials were not included. The abstracts of studies published at conferences, case studies, reviews, qualitative studies, gray studies, and letter to the editor due to lack of use of primary data (PPOD) separately were excluded. Also to accurately and without bias determine the patient's preferences studies focusing on pediatric cancer patients (individuals under 18 years of age), presenting secondary and tertiary by nurses, physicians or family caregivers, and inability to differentiate reported outcomes for cancer patients were excluded. Primary studies were performed on cancer patients of any race, ethnicity, and one of the two sex groups of men or women or both of them, were entered into the study.

Sampling Method and Sample Size

Sampling methods in studies were randomly systematic review (probable) (simple random sampling, systematic random sampling, stratified random sampling, cluster random sampling) or primary studies using non-random (non-probable) sampling methods (quota sampling), convenience sampling, purposive sampling, self-selection sampling, and snowball sampling) or public call announcements or a combination of them were entered into the study.

Selection Criteria

Original articles published in English without time-limitation were identified according to search criteria's. Duplicate sources were removed using EndNote X8 software. In the screening stage, the titles and abstracts of the articles were reviewed. Selected studies were divided into three categories: related, unrelated, and unreliable. Articles reported by both unrelated researchers were excluded from the study. Then in the selection stage, the full text of the articles independently were investigated by two researchers (A.F. and S.B.). All disagreements at any stage were resolved by discussion and agreement between the two researchers. In case of disagreement between the researchers, a third person was used as a judge and the result was reported as a statistical Kappa coefficient after general agreement. Data extraction and quality assessment studies were performed by two researchers (A.F and S.B.).

Search Strategy

The studies were searched in four databases: PubMed/Medline, Scopus, web of science, and ProQuest on October 22, 2020, without any time-limitation. The keywords for this systematic study were a combination of Mesh Term and Free Text words (**Table 1**). In the case of encountering a study in accordance with the objectives of the study, in the case of lack of access to the full text of articles, unpublished data or the existence of erroneous and ambiguous data, an email was sent to the corresponding

TABLE 1 | Search strategy.

Search engines and databases: PubMed, Scopus, web of science, ProQuest
Limits: Language (Only resource with at least an abstract English)
Date: Up to 22 October 2020
Strategy: #1 AND #2 AND #3 AND #4 AND #5 AND #6
#1… cancer OR neoplasm OR tumor OR malignancy OR carcinoma
#2… Death OR dying OR die
#3… Place OR location OR site
#4… palliative OR hospice OR terminal OR "End of life" OR supportive OR "Terminally ill"
#5… choice OR prefer OR decision OR wish
#6… Hospice OR Hospital OR Home

author of the article and three more emails were sent at intervals of 1–10 days. If no message was received from the author of the article after 3 emails, the article was deleted. Any disagreement was resolved by agreement of the two researchers (S.B, A.F) and in case of disagreement, the opinion of the third informed person was the criterion for decision making.

Assessment of the Risk of Bias

After reviewing the purpose of the studies and inclusion criteria, a total of 27 studies in terms of quality was separately evaluated by two researchers (A.F. and S.B.). All disagreements were resolved by discussion and agreement between the two researchers. In case of disagreement between the researchers, a third person was used as a judge. The quality of these articles was assessed using the Newcastle-Ottawa Edited Scale (Observation Studies Version) (Wells et al., 2000). In this scale, articles were assessed based on 4 criteria including representativeness of the samples, sample size (non-respondents, and measurement tool), comparability (a section including review of confounders and other influencing factors), and results (from two aspects: Assessment of the outcome and statistical test) were studied. Based on the Newcastle-Ottawa scale, articles were rated from zero (weakest study) to 10 (strongest study). For data maintenance, studies with a score lower than the mean score (less than a score of 4) were considered low quality. None of the 27 studies were excluded due to low quality.

Data Extraction

Data extraction was separately conducted by two researchers A.F. and S.B. using a researcher-made form. Initially, an article was assessed as a pilot with this form; then was done for other articles. Each researcher used a data extraction form for their articles and the two lists were compared. All disagreements between the researchers were resolved by mutual agreement. In case of disagreement between the researchers, a third person was used as a judge and the result was reported after general agreement. Data related to the author, year, place of study, year of study, sample size, study design, preferred place of death (Home, Hospital, Hospice), and study quality was extracted. Using this form, the preferred place of death of patients with cancer was extracted and the results of the studies were classified into different factors.

Statistical Analysis

Meta-analysis was performed using "meta" command in STATA 16 software. According to the existence of heterogeneity between studies, data were pooled using a random effects model by dersimonian- larid method. The heterogeneity of studies was assessed by the Cochran Q statistic. We planned to test the statistical heterogeneity with the Q test ($\chi 2$, I2, and Tau-squared statistics). The index I2 was interpreted by the following guide (Deeks et al., 2019).

(I2: 0–40; mild, I2: 40–70; moderate, I2: 70–90; sever, and I2; 90–100; highly sever).

The findings were considered heterogeneous if the *P* value was <0.1. Moreover, I2 was utilized to provide a model of the degree of inconsistency between the results of the studies. A value of 0% indicated no observed heterogeneity, whereas larger values showed increasing heterogeneity. Moreover, Egger test was used to determine publication bias in results. In case of encountering a duplicate article, only one duplicate study was used in the relevant composition. If the data is a graph, Web plot Digitizer software at: https://apps.automeris.io/wpd, was used, and if it is not a graph, corresponded with the responsible author. If no response was received within three different time intervals of 10 days, the data related to the initial objectives of the study were deleted.

RESULTS

Studies Identified

One thousand five hundred fifty-three articles were found after searching. Endnote software version 8, (End Note. Thomson Reuters, X8) was used to organize information. Using the mentioned software and reviewing the title and abstract of articles, 522 duplicate articles were removed. Then, the title and abstract of 1031 articles were reviewed by researchers (A.F) and (S.B). A total of 857 unrelated articles were deleted in accordance with the objectives of the study. At this stage, if a study is suspicious, the full text of the article was reviewed by the researchers. In the next step, a search was performed to access the full text of the articles, access to the full text of 3 articles was not possible, and finally, the full text of 171 articles was reviewed. By considering the inclusion and exclusion criteria in accordance with the objectives of the study, Articles due to lack of research results specifically for cancer patients ($n = 71$), Lack of focus on choosing the preferred place of death ($n = 49$), reporting secondary results by a person other than the patient ($n = 9$), review, qualitative studies, letter to the editor ($n = 16$), the inability to differentiate outcomes for cancer patients ($n = 3$) was eliminated. To ensure the retrieval of all articles, the list of sources of the final articles was also manually searched and 5 more articles were added to the final articles. Finally, 27 studies were finalized. The process of entering studies based on inclusion and exclusion criteria was shown in **Figure 1**.

Articles' Descriptive Characteristics

Table 2 summarizes the characteristics of the studies selected for meta-analysis. In this table, the author, year of publication, sample size, study design, study time, study population, country,

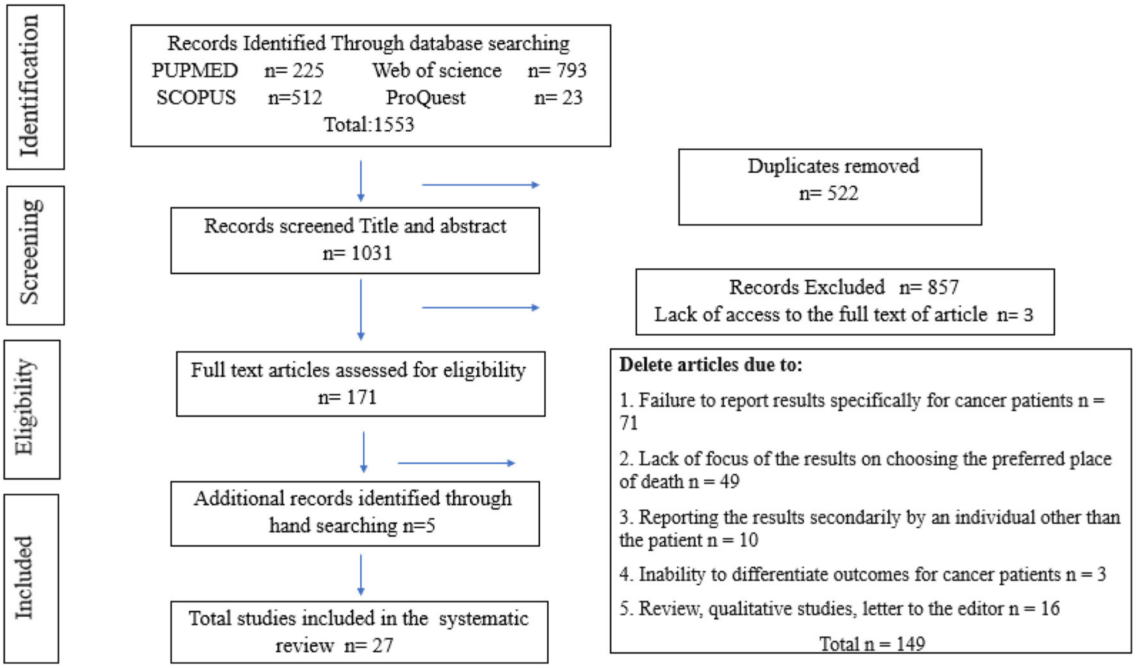

FIGURE 1 | PRISMA flowchart.

continent, and preferred place of death (home, hospital, hospice) were separately expressed for each study. The number of participants in all studies was 14,920. Based on the year, studies were conducted in different years, the number of which is based on the year of study, including 1995 to 2005: 13 studies, 2006-2010: 5 studies, and 2011-2020: 7 studies. In 2 studies, the time of the study was not mentioned. By continent, 11 studies were conducted in Asia, 4 in the Americas, 8 in Europe, 3 in Africa, and 1 in Australia. Both men and women participated in all studies. According to the study design, 15 studies were cross-sectional, 11 were cohort studies and 1 was case-control studies. Of the 27 studies that reported Home: 55% of cancer patients with a safe interval [95% CI (61-49)] listed home as the preferred place of death; of the 21 studies that reported Hospital, 17% of patients with a confidence interval [95% CI (12-23)] preferred hospital as their favored place of death, and of the 12 studies that reported Hospice, 10% of patients with a confidence interval [95% CI (8-13) preferred hospice centers as their preferred place of death.

Prevalence Preferred Place of Death

Prevalence of Home-Based PPOD

In 27 studies, the preferred place of death in home was reported. Based on the prevalence Home PPOD test of heterogeneity ($\chi 2 = 1502.50$, chi-square DF = 26, $P \leq 0.001$) and heterogeneity indices [I2 = 98.27% and tausquared = 0.025], we used a random-effects model to calculate the prevalence. The pooled prevalence of Home PPOD in these studies was 55% [95% CI (49–61)]. Moreover, Egger test ($z = -2.17$, P value = 0.029) showed that there was publication bias in results. **Figure 2** shows the preferred place of death at home in all studies.

Prevalence of Hospital-Based PPOD

In 21 studies, the preferred place of death in hospital was reported. Based on the prevalence Hospital PPOD test of heterogeneity ($\chi 2 = 1859.20$, chi-square DF = 20, $P \leq 0.001$) and heterogeneity indices [I2 = 98.92% and tausquared = 0.015], we used a random-effects model to calculate the prevalence. The pooled prevalence of Hospital PPOD in these studies was 17% [95% CI (12–23)]. Moreover, Egger test ($z = 2.32$, P value = 0.020) showed that there was publication bias in results. **Figure 3** shows the preferred place of death at hospital in 21 studies.

Prevalence of Hospice-Based Preferred Place of Death

Figure 4 shows the preferred place of death at hospice in 12 studies. Based on the prevalence PPOD Hospice test of heterogeneity ($\chi 2 = 237.00$, chi-square DF = 11, $P \leq 0.001$) and heterogeneity indices [I2 = 95.36% and tausquared = 0.001], we used a random-effects model to calculate the prevalence. The pooled prevalence of PPOD Hospice in these studies was 10% [95% CI (8–13)]. Moreover, Egger test ($z = 7.39$, P value = $P \leq 0.001$) showed that there was publication bias in results. **Figure 4** shows the preferred place of death at Hospice in 21 studies.

Factors Influencing the Choice of Preferred Place of Death in Cancer Patients

Various studies have identified different contributing factors to the preferred place of death for cancer patients (Ikezaki and Ikegami, 2011). These factors play an important role in choosing the preferred place of death in cancer patients. Studies have been considered influential the factors such as early diagnosis, marital

TABLE 2 | Characteristics and results of included studies.

*N	Author Year	Continent country	Study design Study time Sample size	PPOD of Home N (%)	PPOD of Hospital N (%)	PPOD of Hospice N (%)	** NOS Score
1	Kern et al. (2020)	Europe Switzerland	Cross Sectional 2015–2016 *n*: 116	34	9	**NR	5
2	Portorani et al. (2020)	Asia Iran	Cross Sectional (Short report) 2018 *n*: 274	176	NR	NR	7
3	Sheridan et al. (2020)	Europe UK	Prospective Cohort 2004–2012 *n*: 453	184	80	82	8
4	Alsirafy et al. (2019)	Africa Egypt	Cross Sectional 2014–2018 *n*: 272	253	19	NR	8
5	Blanchard et al. (2019)	Africa Southern Africa	Prospective cohort 2016-2018 *n*: 191	127	NR	NR	7
6	Shen et al. (2018)	Africa South Africa	Cohort 2016–2017 *n*: 221	127	51	4	7
7	Skorstengaard et al. (2017)	Europe Denmark	Cross Sectional 2013–2015 *n*: 81	29	1	33	7
8	Howell et al. (2017)	Europe UK	Cohort 2005–2010 *n*: 142	65	40	24	7
9	Gu et al. (2015)	Asia China	Prospective cohort 2007–2012 *n*: 522	280	204	NR	7
10	Guerriere et al. (2015)	America Canada	Prospective cohort 2010–2012 *n*: 302	185	***NR	NR	8
11	Chen et al. (2014)	Asia Taiwan	Cross Sectional 2003–2004 *n*: 2034	1114	483	53	6
12	Lee et al. (2014)	Asia Taiwan	Cross Sectional 2009–2011 *n*: 439	212	85	NR	7
13	Aoun and Skett (2013)	Australia Australia	Cross Sectional 2009–2010 *n*: 43	19	4	11	7
14	Jeurkar et al. (2012)	America USA	Retrospective Cohort 2000–2008 *n*: 5837	4336	35	254	8
15	Ikezaki and Ikegami (2011)	Asia Japan	Retrospective Case control 2005 *n*: 1664	810	207	NR	8
16	Ishikawa et al. (2013)	Asia Japan	Cross Sectional 2011 *n*: 258	123	NR	NR	7
17	Blaney et al. (2011)	Europe Ireland	Retrospective Cohort 2007 *n*: 283	173	36	41	7
18	Alonso-Babarro et al. (2011)	Europe Spain	Prospective cohort 2004–2006 *n*: 380	182	NR	NR	7
19	Nakamura et al. (2010)	Asia Japan	Cross Sectional 2005–2006 *n*: 92	37	18	NR	7

(Continued)

TABLE 2 | Continued

*N	Author Year	Continent country	Study design Study time Sample size	PPOD of Home N (%)	PPOD of Hospital N (%)	PPOD of Hospice N (%)	** NOS Score
20	Stajduhar et al. (2008)	America Canada	Cross Sectional 2001–2003 *n*: 56	28	17	NR	7
21	Hsieh et al. (2007)	Asia Taiwan	Cross Sectional NR *n*: 46	34	12	NR	8
22	Kui et al. (2005)	Asia Korea	Cross Sectional NR *n*: 371	175	118	54	7
23	Tang et al. (2005)	Asia Taiwan	Cross Sectional 2003–2004 *n*: 559	341	135	11	7
24	Thomas et al. (2004)	Europe UK	Cohort 2000–2002 *n*: 41	10	0	8	8
25	Tang and McCorkle (2003)	America USA	Prospective cohort 2001–2002 *n*: 127	111	3	10	7
26	Gyllenhammar et al. (2003)	Europe Swedish	Cross sectional 1999 *n*: 221	81	NR	NR	7
27	Lee and Pang (1998)	Asia Singapore	Cross sectional 1995 *n*: 44	23	15	NR	7

[*] *Number.*

[**] *Newcastle—Ottawa Quality Assessment Scale.*

[***] *No Report.*

status, early referral to palliative care specialists, socioeconomic status, age (Bell et al., 2010), gender, place of residence (urban or rural), education, job status, and knowledge of the concept of death in choosing the preferred place of death (Foreman et al., 2006). According to the factors in different studies, factors affecting the choice of preferred place of death in cancer patients were divided into Three groups (Demographic characteristics, disease-related factors and psychosocial factors).

Demographic Characteristics

In 3 studies "age" (Blaney et al., 2011; Jeurkar et al., 2012; Blanchard et al., 2019), in 2 studies "gender" (Kui et al., 2005; Sheridan et al., 2020), in 1 study "area of residence" (Gu et al., 2015), in 2 studies "level of education" (Chen et al., 2014; Gu et al., 2015), in 1 study "race" (Jeurkar et al., 2012), in 3 studies "the status of life," (living alone or with other family members) (Gyllenhammar et al., 2003; Guerriere et al., 2015; Gu et al., 2015) in one study, "the initial place of patient care" (Jeurkar et al., 2012) was significantly associated with the choice of the preferred place of death in cancer patients.

In the study by Blaney et al. (2011) younger patients were more likely to choose the hospital. In the study by Jeurkar et al. (2012) younger patients chose home more as the preferred place of death, in the study by Blanchard et al. (2019) older patients chose home more as the preferred place of death. In a study by Kui et al. (2005) women were less likely than men to choose the home as a preferred place of death, and In a study by Sheridan et al. (2020) women were more likely to discuss on preferred place of death. In a study by Gu et al. (2015) and Chen et al. (2014) patients with a lower level of education chose home as their preferred place of death.

In a study by Gu et al. (2015) patients living in rural areas chose home as their preferred place of death. Also, in this study, patients living with family members were more likely to choose the home, unlike the study of in the study by Guerriere et al. (2015) patients living alone were less likely to choose home as their preferred place of death. In the study by Jeurkar et al. and Gu et al. married people were more likely to choose home as their preferred place of death (Jeurkar et al., 2012). In a study by Jeurkar et al. (2012) whites' patients were more likely than blacks to choose the home as their preferred place of death.

Disease-Related Factors

In 3 studies "type of cancer" (Chen et al., 2014; Howell et al., 2017; Blanchard et al., 2019), 2 studies "time of diagnosis" (Chen et al., 2014; Gu et al., 2015) 1 study "prognosis of the disease" (Jeurkar et al., 2012), 1 study of patient symptoms such as pain (Blanchard et al., 2019), 1 study "medical treatment", 2 studies "disease awareness" (Gyllenhammar et al., 2003; Blanchard et al., 2019), 1 study of "satisfaction with treatment" (Gu et al., 2015), 1 study "the functional status" (Jeurkar et al., 2012) of patients was

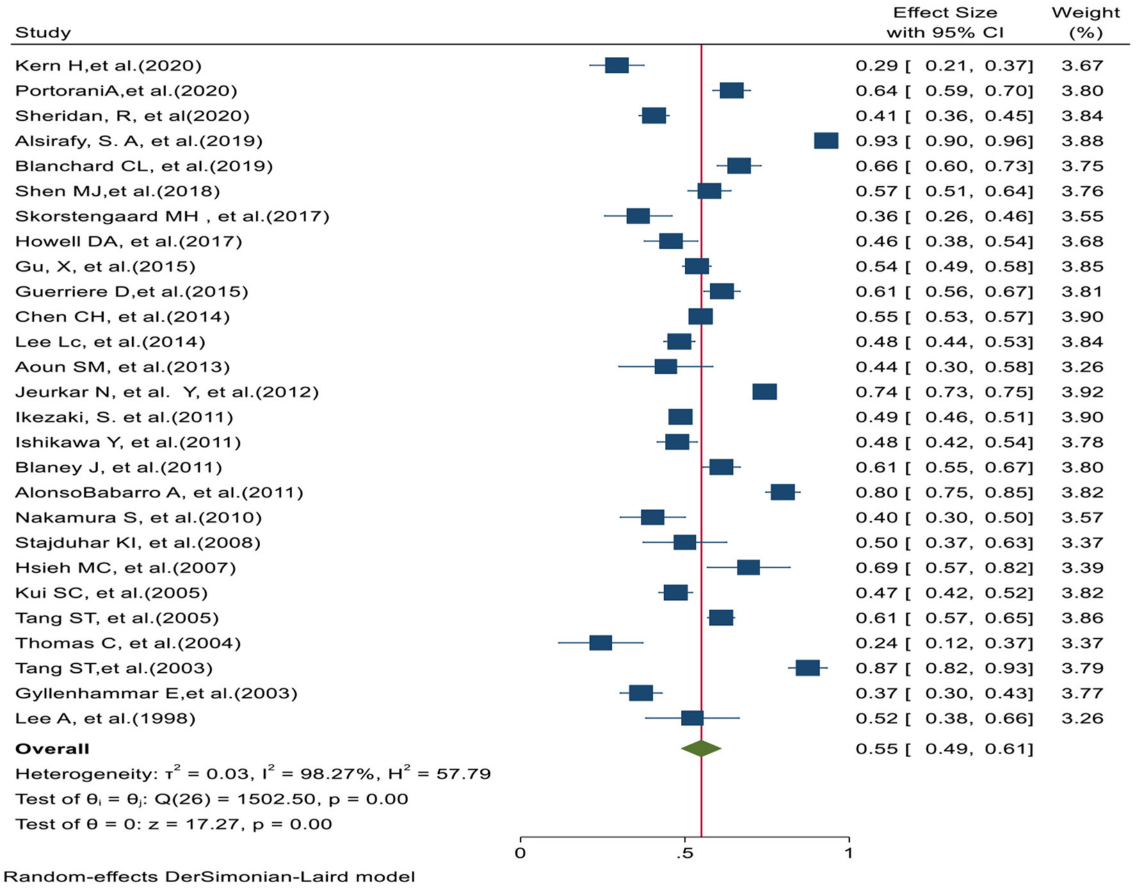

FIGURE 2 | Forest plot for the prevalence of Home-based PPOD.

a significant related to the choice of preferred place of death in patients with cancer.

In the study of Chen et al. (2014) patients with liver and pancreatic cancer, and in the study of Blanchard et al. (2019) patients with lung or breast cancer were more likely to choose home as their preferred place of death.

In the study by Blanchard et al. (2019) patients who knew their prognosis were less likely to choose home as their preferred place of death. In the study by Jeurkar et al. (2012) patients with severe to moderate pain were less likely to choose home as their preferred place of death; Also in this study, patients with better and more independent functional status were less likely to choose the home, and patients who used morphine to relieve pain were more likely to choose home as their preferred place of death (Blanchard et al., 2019). In the study by Gyllenhammar et al. (2003) and Blanchard et al. (2019) people who were aware of their illness were more likely to choose home as their preferred place of death. In the study by Gu et al. (2015) patients who were satisfied with their treatment status and patients who were in the poor physical condition and survived for more than 6 months from the diagnosis were more likely to choose home as their preferred place of death.

Psychosocial Factors

Psychological factors influencing the preferred place of death in cancer patients was including not imposing burden on family and other caregivers (Lee and Pang, 1998; Kui et al., 2005; Tang et al., 2005; Yamagishi et al., 2012), Patients' level of anxiety (Skorstengaard et al., 2017), awareness of the incurability of the disease (Gomes et al., 2015), participation or non-participation of social workers (Kern et al., 2020), Being with the family in the last days of life (Kui et al., 2005), having independence in doing daily tasks at the end of life (Tang et al., 2005, 2006), environmental security (Lee and Pang, 1998) and intimacy and emotional connection with family members (Lee and Pang, 1998).

DISCUSSION

The importance of patients' preferences is considered an important result of palliative care, so it is important to understand the preferred place of death and the factors that affect it in end stage cancer patients. Because the choice of this place is directly related to the spiritual and physical peace of patients and their families (Cabañero-Martínez et al., 2019). Therefore, this systematic review and meta-analysis study was performed to

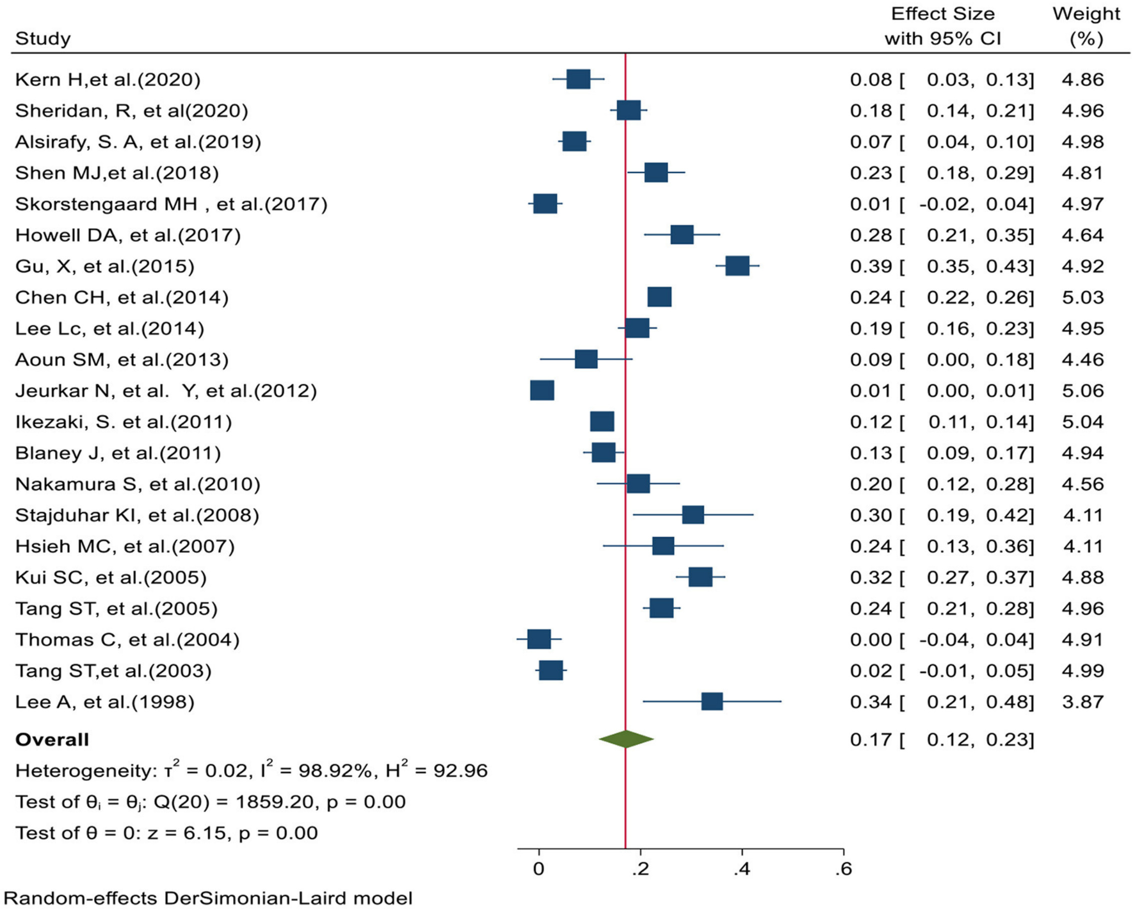

FIGURE 3 | Forest plot for the prevalence of Hospital-Based PPOD.

investigate the preferred place of death and the factors affecting it in adult patients with cancer.

Recent studies have also shown that preference for place of care and place of death is not a fixed concept and can change over time through discussion between health care professionals and patients (Butow et al., 1997; Munday et al., 2009). Of the 27 studies that reported Home: 55% of cancer patients with a safe interval [95% CI (61–49)] listed home as the preferred place of death; of the 21 studies that reported Hospital, 17% of patients with a confidence interval [95% CI (12–23)] preferred hospital as their favored place of death, and of the 12 studies that reported Hospice, 10% of patients with a confidence interval [95% CI (8–13) preferred hospice centers as their favored place of death. Results of the study by Jeurkar et al. (2012) show that in the United States, of 5,837 patients under study, about 56.5% with a confidence interval [95% CI (1.77–2.76)] of patients preferred to die at home (Jeurkar et al., 2012). In the study by Ikezaki and Ikegami (2011) on cancer patients in Japan, half of the patients and 42% of families preferred to die at home, when the patient and family preferences were different, it was the patient who preferred to die at home (Ikezaki and Ikegami, 2011). Another study In Taiwan, conducted by Chen et al. (2014), shows that more than half of the participants (54.7%), preferred to die at home (Chen et al., 2014). The results of a meta-analysis by Suzanne Rainsford et al. show that most patients reported a preferred place for care and death in the hospital and at home (Rainsford et al., 2016). Another systematic study reveals that home is the preferred place of death for most cancer patients worldwide (Bell et al., 2010). Debra A Howell et al. in a systematic study and meta-analysis show that Debra A Howell et al. (2010). In a systematic study and meta-analysis show that most patients with hematologic malignancy die in hospital more than twice as often as patients with other cancers. Given that the house is usually considered as their preferred place of death. If the patient prefers to die at home, he or she should be respected as much as possible, for example, some patients with moderate to severe pain can stay home with effective pain management. The patient may not be in a position to express his or her wishes, and the family's preference and capacity for care should be considered (Leff et al., 2000; Jack, and O'BRIEN, 2010; Silveira et al., 2010). In addition, the home may not have enough facilities to meet the needs of the dying person, people who can provide end-of-life care, and the resources of the health care system (Gomes and Higginson, 2008). The results of a study by Vidal

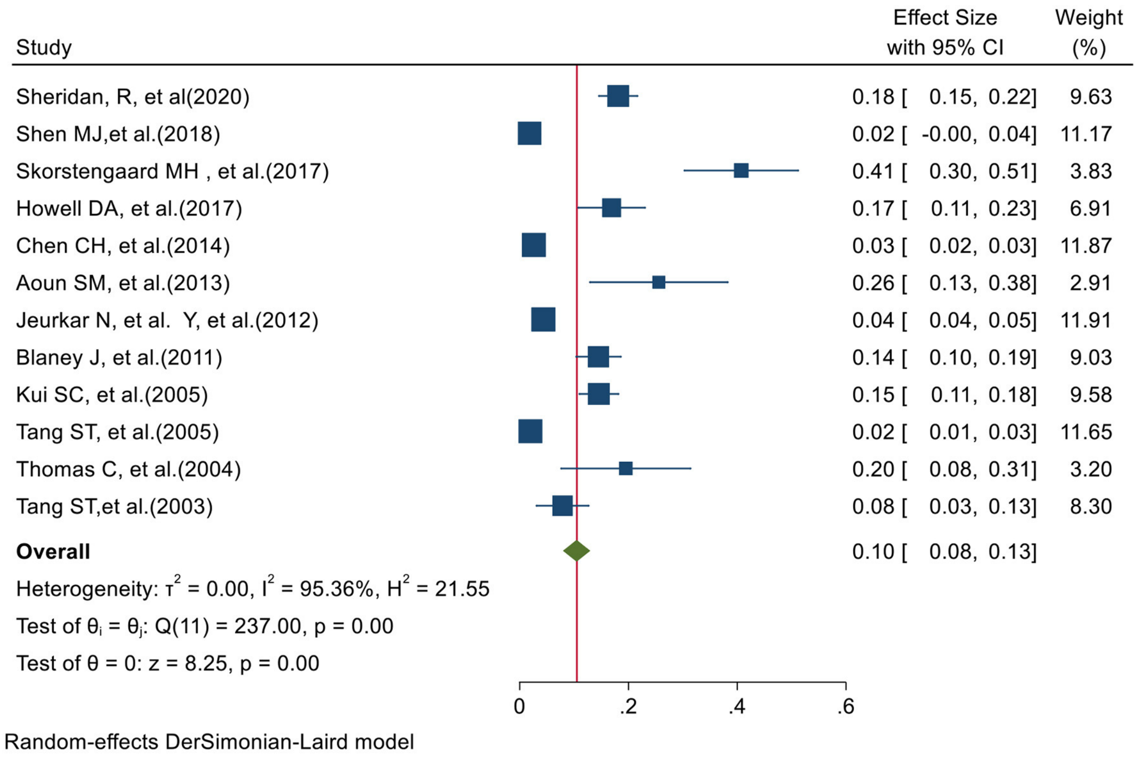

FIGURE 4 | Forest plot for the prevalence of Hospice-based PPOD.

et al. (2020) show that a significant number of patients have no preference or prefer to die in hospital, especially in patients who have already examined home care, have financial and social problems, or have uncontrolled physical and mental symptoms and distress. In many countries, such as the United Kingdom, Japan, South Korea, Greece, and Italy, the number of deaths at home is declining, and in some other countries, such as the United States and Canada, guidelines have been developed to reduce deaths at home (Higginson et al., 2013). Therefore, health care professionals should provide the patient and family with prognostic information and discussions about end-of-life care to facilitate their understanding of the patient's preferred place of death.

In the second part of the study, the factors related to the preferred place of death were investigated. Due to the lack of similarity between different studies, it was not possible to perform meta-analysis in this dimension, and the results have been reported qualitatively and do not necessarily indicate a specific direction and cause. Because each person's preferences are unique and influenced by a variety of factors, identifying priority predictors of the preferred place of death is useful for understanding how patients make decisions about where to live at the end of their lives. But the results of studies have mentioned many different factors. The present study reported the effective factors in the preferred place of death of cancer patients in two groups of demographic characteristics, disease-related factors. Disease-related factors highlight the issue of palliative care time. Patients in the more advanced stages of the disease with uncontrolled mental, physical, and social symptoms may die less at home because they have multiple treatment options, even in the advanced stages of the disease, and the responsibility of caring for family members is less. In most individual factors, the preferences, desires, and inclinations of the patient play an important role in achieving the place of death. In a systematic study, other factors influencing the preferred place of death in the two groups of maintaining patient individuality include demographic variables (relatively stable and unchangeable characteristics in patient identity), personal factors reflecting patients' beliefs, desires, and internal resources for adaptation, and underlying environmental factors (Gomes and Higginson, 2006). The results of diffusion bias according to Egger test in three death places were significant in terms of patient (home, hospital, and hospice) preferences.

Regarding the psychological factors affecting the preferred place of death, researchers report severe heterogeneity and the results are qualitatively reported, so the evidence was inconclusive. According to a study (Lee and Pang, 1998; Tang et al., 2005, 2006), intimacy, emotional connection with the family and having independence in doing daily tasks at the end of life are among the psychological factors affecting the preferred place of death. Solomon et al. reported in 2013 that examining family relationships emotionally, from the patient and family member's perspective, may enrich their understanding and ability to help patients die at home (Solomon and Hansen, 2015). Support from family members may require a good family relationship and mutual trust. Qualitative findings suggested that caregivers commit to providing care and to address the patient's preference to be at home, to then become aware of the complexities involved. Victoria Turner et al. showed the main factor affecting access to preferred place of death was social support; people with fewer informal carers were less likely to die in their preferred location. This highlights the importance of good communication of preferences and concerns between patients and caregivers throughout the process, and the need for practical and emotional support to caregivers, to meet the patient's preference when possible and to minimize the risk of difficult bereavement for caregivers (Gomes et al., 2013). Moreover, human factors such as social support and career resilience, plus the availability of resources such as care staff and hospice beds, as important factors in achieving preferred place of death (Turner and Flemming, 2019). Other sociological factor is Patients' level of anxiety. The participants in Pradilla study that experienced emotional symptoms such as depression and anxiety preferred dying in a health care environment. Their assumption is that these people are more prone to seek and need professional assistance (Pradilla et al., 2011). This stresses the role of the mental health professional when taking care of a terminal patient, as reported in the review by Gibson et al. (2006). Also, helping the next of kin to be aware of the impending death may increase the chance to die at home and based on preferred place (Lee et al., 2014).For a better hospice care service, it is essential to inquire patients or their relatives on preferred place of death while concerning the influences of other factors. Therefore, identifying the factors affecting the preferences of patients in the end stages of life and their families is a necessity that should be considered.

In our study, heterogeneity levels were calculated using Q and I2 tests, the level of which was reported to be high in terms of home death place (I2 = 98.27%), hospital (I2 = 98.92%), and hospice (I2 = 95.36%), respectively. One of the reasons for the increase in the level of heterogeneity can be the combination of different studies with different sample sizes. In the present study, the lowest sample size consisted of 41 samples and the highest sample size consisted of 5,837 samples.

ADVANTAGE AND LIMITATIONS

In our study, priori registration in PROSPERO system based on the principle of comprehensiveness and quality, data combination and investigation of the amount and possible causes of heterogeneity, as well as more inclusive search based on the use of synonymous detection systems Thesaurus Mesh and, Emtree, examining large databases such as PubMed, Scopus, web of science, ProQuest with extensive search time, using the opinion of experts, without time and space limitations, this systematic review can be compared to previous studies have a more comprehensive review of initial studies in the field. In the second part of the study, the factors related to the preferred place of death were investigated. Due to the severe heterogeneity between studies in this dimension, the results were qualitatively reported, which is one of the limitations of this study. Given that more than half of patients chose home as their preferred place of death, it is suggested that future studies on cost-effectiveness and health resource allocation be devoted to home-based end-of-life care.

CONCLUSION

Based on the results of the present study, more than half of cancer patients in the later stages of life prefer to die at home. Also, the factors affecting the individual's preferences in deciding to choose the preferred place of death have been classified into two groups of demographic characteristics, disease-related factors. Considering that one of the goals of palliative care and end-of-life care program is to increase the quality of life of patients and their families, for this reason, guided policies to ensure the death of patients in the desired place according to the patient's condition should be a priority. Therefore, health care professionals should be aware of the palliative care needs of patients.

DATA AVAILABILITY STATEMENT

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

AUTHOR CONTRIBUTIONS

AF and SB conceptualized and designed the study, retrieved data, and rechecked the data. MS, AV-A, AF, and SB conducted study analyses. AF, SB, and MR did the initial drafting. AF, MR, HA, MS, AV-A, and SB critically assessed the data and provided intellectual inputs. All members approved the final draft.

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REVIEW
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Loneliness in Bereavement: Measurement Matters

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The role of loneliness in the bereavement experience has been reported as substantial, with the death of a close person leaving a considerable void in the life of the bereaved. Yet, there is lack of agreement about its precise role and, notably, whether loneliness should be included as a core symptom for diagnosis of grief complications. The ongoing threat of heightened social isolation due to the COVID-19 pandemic underlines the need to understand the impact of loneliness, and to accurately chart its prevalence, intensity, duration, and associated difficulties in the context of bereavement. Assessment issues are central to this endeavor. In this article, we review the scientific literature to examine how loneliness after bereavement has been operationalized and measured. Sixty-three articles analyzing 51 independent datasets were reviewed. Results show major disparities: approximately half of the projects assessed loneliness by means of one of two validated scales (spanning different versions); the remainder included only single- or few-item measures. Diverse instructions, content and answer categories were used. While one size does not fit all, awareness of assessment options and dis/advantages may aid selection of the most appropriate measure, to suit the goals of a particular study and the specific groups under investigation. Our conclusion is that, in selecting a loneliness measure, health care professionals should come to their own well-informed decision, aided by the information provided in our review.

Keywords: loneliness, measurement, assessment, social isolation, bereavement, grief, prolonged grief disorder, complicated grief

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INTRODUCTION

Loneliness is generally understood to be a distressing reaction accompanying the perception that one's social needs are not being met by one's interpersonal relationships (Hawkley and Cacioppo, 2010). It has been established as a risk factor for compromised health and well-being, not only in terms of general mental and physical illness, morbidity, and mortality, but also for specific problems such as reduced daily functioning and suicidal ideation, risky health behaviors, age-related disease such as Alzheimer's, and various physiological indices of ill-health (Hawkley and Cacioppo, 2003; Leigh-Hunt et al., 2017; Brown et al., 2018; Wang et al., 2018). A range of theoretical approaches has been offered in the scientific literature to help explain such phenomena and manifestations of loneliness, with major contributions including the classic attachment theory extension by Weiss (1973); the cognitive discrepancy approach of Perlman and Peplau (1981) and the evolutionary perspective of Cacioppo et al. (2014). For an extended review of these theories, see De Jong-Gierveld et al. (2018) and Marangoni and Ickes (1989).

With respect to bereavement, research has confirmed that loneliness is frequently one of the major challenges experienced following loss (Vedder et al., 2021). Particular research attention has been paid to its impact on older persons who have lost their partner (Lund, 1989; Utz et al., 2014). There are indications that loneliness may play a key role in depression after the loss of a close person, as well, functioning as a key symptom that may lead from bereavement to the development of other listed depressive symptoms (Fried et al., 2015). Loneliness is also associated with complications in grieving, including post-traumatic stress disorder, depression, as well as other mental and physical health problems (e.g., Simon et al., 2014; Erzen and Çikrikci, 2018; Asch et al., 2021).

A recent review of empirical studies of loneliness in bereavement testifies to the greater frequency and/or intensity of loneliness among bereaved compared with non-bereaved groups (Vedder et al., 2021). Yet, loneliness does not appear consistently in lists of core symptoms in diagnostic manuals of mental disorders. Different sets of criteria for disturbed grief have been proposed (Boelen and Lenferink, 2020). In the forthcoming text revision of the 5th edition of the Diagnostic and Statistical Manual for Psychiatric Disorders (DSM-5-TR; American Psychiatric Association and Association, 2013; Prigerson et al., 2021), "Intense loneliness (i.e., feeling alone or detached from others) as a result of the death" is listed as a symptom among the criteria for Prolonged Grief Disorder (PGD). Similarly, "Loneliness" is one of the four separation distress symptoms of Complicated Grief (CG) proposed by Shear et al. (2011). By contrast, there is no symptom of loneliness among those for PGD as described in the 11th edition of the International Classification of Diseases (ICD-11; World Health Organization, 2019).

One plausible reason for the discrepancy in recognition of loneliness as a key feature of complications in grief is that there is still quite limited knowledge regarding the nature, prevalence, and intensity of loneliness and loneliness-related problems encountered by bereaved people. In our view, matters relating to assessment contribute to this shortage of information. Examination of the general (i.e., non-bereavement-specific) loneliness literature points to a lack of agreement on how best to measure loneliness or what precisely such assessment(s) should incorporate (for example, whether to measure a single or multiple related constructs; Cramer and Barry, 1999). This likely also reflects the difficulties associated with capturing the multidimensionality and complexity of the construct (Yanguas et al., 2018). Nevertheless, providing a useful starting point for further consideration, there is reasonable consensus regarding a general definition of loneliness (for a review of definitions, see: De Jong Gierveld, 1998; Hawkley and Cacioppo, 2010). According to Valtorta and Hanratty (2012), *"One of the most widely-used definitions has loneliness as a subjective negative feeling associated with a perceived lack of a wider social network (social loneliness) or the absence of a specific desired companion (emotional loneliness)"* (p. 518).

There are good reasons to address issues of assessment, particularly in the context of bereavement. For example, not only is there need to reach consensus across different diagnostic sets of criteria for grief complications (Lenferink et al., 2021), but in the changed world of the COVID-19 pandemic, loneliness is likely to become an even greater issue, particularly among the bereaved. Predictions are that the lack of physical presence and support from family and friends will intensify feelings of loneliness even more than in non-pandemic times (Stroebe and Schut, 2020), with evidence beginning to confirm such prognoses (van Tilburg et al., 2020). Given these concerns, research needs to provide a body of sound information on the prevalence and intensity, correlates, and consequences associated with loneliness in bereavement. In order to do so effectively, accurate measurement is needed.

The purpose of this study was to examine the assessment of loneliness in bereavement; to summarize the current state of knowledge; to chart how loneliness has been operationalized and interpreted to date; and to suggest ways forward for future research. To do so, we reviewed the existing body of scientific literature available on this topic. More specifically, we sought to explore:

- Which measures/questionnaires have been used?
- How have they (differentially) assessed loneliness?

METHOD

Information for the present review was drawn from published, empirical studies designed to examine loneliness among bereaved people that we considered in our systematic review on the prevalence, correlates, and intervention efficacy of loneliness in bereavement (Vedder et al., 2021). Measurement issues, such as widespread use of single-item measures and heterogeneity in validated scales, arose from the results of that review but were beyond its scope, suggesting the need for finer-grained examination of the assessment of loneliness.

As described in detail in Vedder et al. (2021), studies conducted before March 12th, 2020 were included. The studies were selected according to the guidelines of the Preferred Reporting Items for Systematic Reviews and Meta-Analyses statement (PRISMA; Moher et al., 2009). The search in Psychinfo, Web of Science, Scopus, Medline, and PubMed returned 8,119 articles, of which 5,600 duplicates were removed, leaving 2,519 (31%) articles for screening. Following title and abstract screening, 312 (12%) articles were retained, and after full-text screening there were 63 articles that conformed to the set criteria for inclusion, representing 51 independent datasets. In all, 16,558 bereaved persons participated in the studies; their mean weighted age was 65 years ($SD = 14$). There were almost equal numbers of cross-sectional ($N = 30$, 59%) and longitudinal ($N = 25$, 49%) studies, but very few ($N = 5$, 10%) investigating the efficacy of interventions. Note that this sum exceeds the number of 51 independent datasets, as some were used for more than one study, with different designs. For the present review, articles were scrutinized to extract information on the measures used in each of the studies. Information was compiled, listing different versions of established scales, their instructions, the particular questions asked in single- or few-item measures, and whether the latter were self-constructed or derived from established scales. Answer categories in terms of response options, and the frequency and/or intensity of experienced loneliness were listed.

RESULTS

An overview of the measures used in the reviewed studies is included in the supplementary table 1 of Vedder et al. (2021). **Tables 1–4** below summarize this information according to the type of measure employed to examine loneliness in the bereavement studies. As can be seen, a variety of scales has been used for assessment. Surprisingly, only about half of the studies used a validated measure, including either the UCLA Loneliness Scale (UCLA-LS; Russell et al., 1978; Russell, 1996; Elphinstone, 2018) or the De Jong Gierveld Loneliness Scale (DJG-LS; De Jong Gierveld and Van Tilburg, 1999, 2010). Other validated scales used (never more than once among those found through our search system) were the modified New York University Loneliness Scale (Rubenstein and Shaver, 1982), Emotional and Social Loneliness Scale (Russell et al., 1984; Robinson et al., 2013) and Emotional/Social Loneliness Inventory (Vincenzi and Grabosky, 1987). We consider the frequently-used scales next, before turning to the use of single- or few-item measures.

Scales Measuring Loneliness in Bereavement

A third of the studies ($N = 17$, 33%) used a version of the UCLA-LS, a well-established measure of loneliness (see Russell, 1996, for portrayal of the questionnaire). The scale has shown dependably high internal consistency, with a coefficient alpha of >0.90 in college student populations and good retest reliability after 12 months ($r = 0.73$) (Cramer and Barry, 1999). Nearly all of the studies using a version of this scale were conducted in the U.S. ($N = 15$, 29%), the country of origin of the scale developers. The UCLA-LS was based on an early conceptualization of loneliness (Russell et al., 1978). The emphasis on uni-dimensionality is noteworthy (Russell, 1996, p. 30), contrasting with the emotional and social loneliness dimensionality of the DJG-LS (see below). Russell et al. (1980), see also Russell et al. (1984) further described a cognitive model of loneliness in which the perception is that “social relationships are “too few,” and people feel “lonely” (Russell et al., 1980, p. 472). Thus, consistent with what is usually understood as the nature of the experience, these researchers placed emphasis on the person’s own subjective perception of loneliness. It is worth noting that intensity as well as frequency were incorporated (as reflected in the scale).

The second most-frequently used scale in bereavement studies is the De Jong Gierveld Loneliness Scale ($N = 8$, 16%) (DJG-LS; De Jong Gierveld and Van Tilburg, 1999, 2010). Internal consistency is reported to be good, particularly among older adult samples, with Cronbach’s alpha coefficients ranging from 0.80 to 0.90 (De Jong Gierveld and Van Tilburg, 1999; for comparison of the UCLA-LS with the DJG-LS see Penning et al., 2014). Overall, authors have concluded the scale demonstrates reliability and validity (De Jong-Gierveld and Kamphuis, 1985; Pinquart and Sörensen, 2001a,b; Gierveld and Tilburg, 2006). Nearly all of the studies using this scale were conducted in The Netherlands ($N = 6$, 75%), again the country of origin of the scale developers. The DJG-LS followed a definition of loneliness as “the manner in which the person perceives, experiences, and evaluates his or her isolation and lack of communication with other people” (De Jong-Gierveld, 1987, p. 120). The DJG-LS therefore emphasizes some rather different components of loneliness (e.g., communication) compared with the UCLA-LS. However, quite similarly to the DJG-LS, loneliness is here considered to be a subjective negative feeling originating from perceived deficits in social relationships, indicating a lack of intimacy or support in relationships, one that reflects a person’s social participation and isolation. The scale also adopted the developers’ so-called “cognitive theoretical approach” (De Jong Gierveld and Van Tilburg, 1999), which emphasizes the discrepancy between what the person *desires* in terms of interpersonal affection and intimacy and what one *perceives they actually have*. Again, this is similar to the cognitive approach reflected in the UCLA-LS. Furthermore—and uniquely at the time the scale was developed—De Jong Gierveld and colleagues built on Weiss’s (Weiss, 1973) constructs of social and emotional loneliness, as compared with the unidimensional measurement approach reflected in the UCLA-LS.

Turning to the actual use of these loneliness scales in bereavement research: Within the empirical studies examined, different versions of the UCLA-LS–including various shortened versions–have been utilized (see **Table 1**; adapted from Vedder et al., 2021). Instructions ask respondents to indicate how often each of the statements applies to them. Items are typically rated on a 4-point scale, from “never feeling this way” to “often feeling this way.” Inspection of these scales shows that the items do not include the actual word *loneliness* (we consider this strategy in Discussion), not even in the instructions, in order to reduce response bias. Instead, they cover aspects such as: lacking companionship, having no one to turn to, feeling isolated, feeling unhappy being so withdrawn. As such, they reach beyond the above-cited definition of loneliness (Valtorta and Hanratty, 2012), to include social isolation as well as related concepts that could function as moderators, mediators, or outcomes (e.g., the included item about unhappiness). Such extensions in the scope also represent a systematic difference compared to the single- or few-item measures of loneliness (discussed below). As Cramer and Barry (1999, p. 493) note, the UCLA-LS does not specify a time frame for respondents, so it remains “unclear whether a state or trait measure has been designed.” There appears to be no norm or reference information concerning UCLA-LS scores, limiting the extent to which scores from a particular study can be compared with those found for other populations or relevant sub-groups. Nor do there appear to be agreed-on points for designation of intensity, for example, to indicate absence of, mild, moderate or high levels of loneliness (Russell et al., 1978, 1980; Weeks et al., 1980). Given the number of different versions of the UCLA-LS that have been used by researchers, it is also difficult to compare results across studies.

As with the UCLA-LS, different versions of the DJG-LS have been employed in bereavement research, with most using the full 11-item scale, while some used the six-item version (see **Table 2**). Like the UCLA-LS, this scale does not include the

TABLE 1 | Versions of the UCLA Loneliness Scale (UCLA-LS) used to measure loneliness (after bereavement).

UCLA-LS	# Items	Scale	Range
Revised UCLA-LS or UCLA-LS Version 3 (Hansson et al., 1986; Murphy, 1986; Gfellner and Finlayson, 1988; Kovarsky, 1989; Byrne and Raphael, 1997; Henderson et al., 2004; Stein et al., 2009; Knowles and O'Connor, 2015; Yan and Bonanno, 2015; Spino et al., 2016; Cao et al., 2020)	20 items	never, rarely, sometimes, often/always	0–80
UCLA-LS-Short Form (Lund et al., 2010)	13 items	never, rarely, sometimes, always	13–52
UCLA-LS-8 (Lee, 2019)	8 items	strongly disagree (1)—strongly agree (5)	8–40
UCLA-LS-4 (Zettel and Rook, 2004)	4 items	agree a lot (1)—disagree a lot (5)	4–20
UCLA-LS-4 (Morgan et al., 1997)	4 items	never, rarely, sometimes, often	4–16
UCLA-LS-5 (Sun et al., 2012)	5 items	agree almost always (1) to almost never (5)	5–25
UCLA-LS-3 (Carr et al., 2018)	3 items	hardly ever, some of the time, or often	0–9

TABLE 2 | Versions of the De Jong Gierveld Loneliness Scale (DJG-LS) used to measure loneliness (after bereavement).

DJG-LS	# Items	Range	Cut-off
DJG-LS (Stevens, 1995; Van Baarsen et al., 1999; De Groot et al., 2006; Onrust et al., 2010; Merz and De Jong Gierveld, 2016; Szabo et al., 2019)	11	0–11	0–2 not lonely; 3–8 moderately lonely; 9–11 strongly lonely;
DJG-LS Short scale (Spahni et al., 2015; Chow et al., 2019)	6	0–6	0–1 not lonely; 2–6 lonely

TABLE 3 | Single items derived from validated measures to assess loneliness (after bereavement).

References	Scale	Single-item measure
Robinaugh et al. (2014) and Fried et al. (2015)	Center for Epidemiologic Studies Depression Scale	"I feel lonely"
D'Epinay et al. (2003)	Self-Assessing Depression Scale	"I feel rather isolated, rather lonely, even among friends"
Pan (2020)	Inventory of Complicated Grief	"I feel lonely a great deal of the time ever since... died"
Waldrop (2007)	Brief Symptom Inventory	"Feeling lonely," "Lonely with people"
Xiang et al. (2016)	Psychosomatic Situation Scale	"Feeling lonely," "Lonely with people"
Oechsle et al. (2020)	Distress Thermometer	Participants can answer "yes" or "no" to whether it was loneliness causing their distress.

word *loneliness* at all, covering such aspects as having no one to share problems with, experiencing a sense of emptiness, feeling rejected, and having no one to trust completely. Thus, the scope seems somewhat broader than the definitions provided by the authors, raising questions about content validity. Instructions were slightly more elaborate than those for the UCLA-LS, explaining that the statements were made by individuals who had previously shared their experience with the researchers, and asking respondents to indicate the extent to which they applied to their own situation, the way they currently felt. However, as with the UCLA-LS, no specific time frame was included. In contrast to the use of a four-point Likert scale for the UCLA-LS, answers for DJG-LS items are scored by circling the statements that applied to the respondent's situation. Notably, a cut-off point relating to severity was provided (De Jong Gierveld and Van Tilburg, 1999). Furthermore, the DJG-LS and its shorter versions enable overall loneliness (from the total scale) as well as emotional and social loneliness (from subscales) to be calculated, facilitating comparison across studies.

Single-Item or Few-Item Measures to Assess Loneliness in Bereavement

More than a third of the studies used single- or few-item measures to assess loneliness ($N = 19$, 37%). Such items were used across more countries than either of the above validated scales: Half of the studies ($N = 9$, 50%) were conducted in various European countries, a lesser proportion was conducted in the U.S. ($N = 7$, 39%). The remainder were either conducted in

TABLE 4 | Self-constructed items to measure loneliness (after bereavement).

References	Single-item measure
Abrahams (1972)	Help-request used and categorized in (1) lonely requires a listener and (2) lonely wants to meet people
Arling (1976)	Two items (1) "Do you have as much contact as you would like with a person that you feel close to- somebody that you can trust and confide in?" (2) "Do you find yourself feeling lonely quite often, sometimes or almost never?"
Atchley (1975)	Participants could answer if they feel lonely "Lots, Some, Hardly Ever or Not all"
Bahr and Harvey (1979)	Two items (1) from Bradburn (1969): "During the past week, how often did you feel very lonely or remote from other people?" (3-point scale). (2) item "Are you as involved in community life as you would like to be?" (yes/no).
Caserta and Lund (1996)	Single item (1 = not at all lonely, 7 = very lonely)
Grimby (1993)	No scale but three dimensions of grief reactions (as rated by psychologists). Under which "low mood" = dysphoria, loneliness, crying and pessimism.
Eckholdt et al. (2018)	Two items (7-point scale)—Emotional loneliness "I feel lonely even when I am with other people" and Social loneliness "I have no really close friends."
Kivett (1978)	Single item "Do you find yourself feeling lonely quite often, sometimes, or almost never?" 3-point scale
Lichtenstein et al. (1996)	Loneliness: Single item "How often do you feel lonely?" 3-point scale "almost never or never" (0) to "almost always or always" (3).
Savikko et al. (2006)	Loneliness: 1 item "Do you suffer from loneliness?" 1 = seldom or never, 2 = sometimes and 3 = often or always
Stroebe et al. (1996)	Self-constructed measure of social (2 items) and emotional (2 items) loneliness (based on Weiss, 1982), yes/no answers
van der Houwen et al. (2010)	Emotional loneliness, 2 items: "I feel lonely even if I am with other people"; "I often feel lonely." 7-point scale ranging from 1 (totally disagree) to 7 (totally agree).

China ($N = 2$, 11%) or internationally-administered ($N = 1$, 6%). A number of the short measures were derived from existing scales ($N = 7$, 14%; see **Table 3**). Instructions (not listed here) varied according to the overall focus of the existing scales from which they were derived, and/or the interests of the particular researcher who developed the short measure.

Other studies—accounting for ∼one-quarter of the total ($N = 12$, 24%)—used items that were self-constructed by the authors (see **Table 4**). A noteworthy difference between these single- or few-item measures and the two validated scales discussed above is that their items all refer explicitly to loneliness, with each of them also employing Likert-type scales. For example, Eckholdt et al. (2018) used a 2-item, 7-point scale of emotional loneliness: *"I feel lonely even when I am with other people"* and social loneliness: "*I have no really close friends."* Savikko et al. (2006), used a 1-item, 3-point one: *"Do you suffer from loneliness?"* with answer categories from 1 = seldom or never, 2 = sometimes, 3 = often or always.

GENERAL COMMENTS

Some general comments on scale use are called-for, to elaborate on certain emerging patterns that arose during the course of this review. We noted a number of differences between the various measurement instruments in terms of content, instructions, and answer categories. We also noted that there were different applications of measures across countries, with more international coverage in the use of single- or few-item measures than for either of the validated scales. Given these results, we explored other potential differences: We considered it plausible that single- or few-item measures might have been employed mostly in the years prior to the establishment of the validated scales; this, however, was not the case. More than half of the studies employing single- or few-item scales ($N = 10$, 56%) were conducted since the year 2000, well after both validated scales were developed. We also considered the possibility that researchers would choose to ask older participants fewer questions, to avoid burdening them with more items (or even to limit response burden for participants in general). This hypothesis was also not supported; the mean ages of participants in studies using the longer scales or short-item scales did not show any notable differences. We reflect on these results further in the Discussion section.

Overall, given the diversity of measurement options described above and in **Tables 1–4**, it is difficult to establish precise rates for prevalence or intensity of loneliness in bereavement, which is clearly relevant for clinical awareness and intervention planning (for further discussion: Vedder et al., 2021). From single-item measures, for example, we mainly learn about percentages of people who are lonely, i.e., how prevalent loneliness is. Prevalence and intensity could be investigated by using the UCLA-LS, but in the absence of any agreed-upon cut-off point, researchers are left to derive their own interpretations, or find other studies using comparable samples and measurement to derive their own conclusions. We also noted the wide variety of versions of measurement instruments that researchers have used. This could lead to different claims about the impact of loneliness in bereavement. For example, with the 6-item DJG-LS version, one can only infer "lonely" vs. "not lonely," whereas with the full 11-item version, four grades of loneliness (not lonely, moderate lonely, severe lonely and very severe lonely) are identifiable. Finally, it is noteworthy that approximately half of the reviewed studies ($N = 26$, 51%) employed scales consisting of fewer than 10 items. This might reflect the necessity for researchers to tailor their study design to the specific characteristics of their participants, and/or to reduce participant burden.

DISCUSSION

Our review of studies of loneliness in bereavement revealed diversity with regard to many central features relating to its assessment. Several of these are noteworthy: There is an abundance of different (versions of) scales and items concerning loneliness; a wide variety of instructions and answer categories are used; there is limited information about how to interpret scores, intensity or cut-off points for clinically-relevant levels of loneliness. Furthermore, the use of different scales and/or versions results in lack of comparability between different studies, and across different populations or sub-groups. Examination of the full body of evidence also shows that, depending on which measure is employed, one might draw different conclusions about prevalence and/or intensity of loneliness. These problems associated with the assessment of loneliness after bereavement add to other methodological limitations of the empirical studies, discussed elsewhere (see Vedder et al., 2021). For instance, the majority of these studies lack non-bereaved control groups and longitudinal assessments.

A series of critical questions remain: What can we learn from the way loneliness has so far been measured in empirical studies on bereavement? How can researchers build on existing information to improve assessment and, consequently, contribute to knowledge in general about the role of loneliness in bereavement? Several points relating to the assessment of loneliness must be taken into account for future research:

Definition of Loneliness

First, it would be advisable to come to a clearer agreement on the definition of what loneliness is—and is not. The widely-used definition by Valtorta and Hanratty (2012) reflects basic components identified in both theorizing and empirical research. Details of the measures included in our review seem in line with this general definition: They focus on subjective negative feelings and some enable distinction of social from emotional loneliness. The latter distinction seems critical when considering loneliness after bereavement, in line with theoretical formulations. In bereavement, one has lost an attachment figure (Weiss, 1973; Bowlby, 1980) a person to whom one was closely bonded, suggesting the centrality of emotional rather than social loneliness (although social loneliness may be particularly felt among older bereaved persons as their networks decrease). Indeed, there is some empirical support for this supposition (Stroebe et al., 1996; Guiaux, 2010). De Jong Gierveld et al. (2006), provide further discussion of the need for distinction of social from emotional loneliness.

Thus, it may be helpful to use the description by Valtorta and Hanratty (2012) as a working definition for further research; others have endorsed this strategy for research on loneliness (Fakoya et al., 2020). However, openness to alternatives may also be called for. Cramer and Barry (1999) have provided a valuable evaluation and comparison of the various self-rating scales (including the UCLA-LS and DJG-LS). They recommend use of the Social and Emotional Loneliness Scale for Adults (SELSA; DiTommaso and Spinner, 1993, 1997). They add the factor "family loneliness," a feature that is well in-line with our argument that emotional loneliness is of particular interest when studying bereavement. Furthermore, Bandari et al. (2019) are currently reviewing definitions of loneliness and, on the basis of the accumulated knowledge, are planning to propose a new definition that may better reflect the scientific construction of what loneliness incorporates.

Avoidance of Conceptual Overlap

There is need to avoid conceptual overlap with related constructs such as social isolation and social support. Yanguas et al. (2018) state that "…various researchers have referred to "loneliness" and "social isolation" indistinctly. Others, however, find both terms very different from each other. Making accurate evaluations depends on a clear definition of the concept of loneliness, with special awareness of its multidimensionality and its differences with respect to related concepts (social isolations or a lack of social support)" (p. 302–303). Our review showed that some measures cover the latter dimensions in their items, while others do not. Researchers may want to adapt the content of the scales to conform to the narrower definition of loneliness, in order to better distinguish it from related—yet distinct—constructs.

Selection of an Appropriate Measurement Instrument

Our results showed that two scales, the UCLA-LS and the DJG-LS, have each been quite widely adopted. A comparative examination of the UCLA-LS with the DJG-LS concluded that the latter has better utility, at least for use among middle-aged and older persons (Penning et al., 2014). However, some of the DJG-LS items border on different domains (e.g., feeling rejected or not having people one can trust). Such items might best be deleted, and/or items from the UCLA-LS that are complementary to those on the DJG-LS added (requiring new validation). The DJG-LS has the advantage of enabling examination of emotional loneliness and social loneliness as well as overall loneliness.

Deciding whether to assess loneliness using a validated instrument or a single/few-item measure requires careful weighing of pros and cons. There is much to support the use of an established scale. Multiple-item usage increases reliability and validity; errors and the specificity that are inherent in single items are averaged out (Bowling, 2005). For such reasons, instruments with carefully selected items are *almost* always considered superior to single item measures in scientific research. However, the qualification *almost* may be particularly appropriate for assessing loneliness in bereavement. For instance, the average age of participants in the reviewed studies was 65 years, suggesting a wide interest in loneliness among older bereaved persons, for some of whom research participation may be more challenging than for younger persons. When study participants are overwhelmed by the length of an interview or the cognitive demand of responding to multiple scale items, using multi-item scale measures may not be the better choice: One well-answered item clearly has superiority over a number of unanswered or incorrectly answered ones. In addition to feasibility, when the goal is to assess the general feeling of loneliness rather than distinguish different dimensions of it, one or a few items may be sufficient. The fact that approximately half of the reviewed studies used ten items or fewer may reflect this dilemma. For some purposes, few items may be able to capture loneliness as well as the score on a longer loneliness scale. Finally, the choice depends on the broader aims of the study (e.g., symptomatology in general vs. loneliness in particular).

In summary: Choice of one of the validated scales or a single/few-item measure likely reflects the distinct aims and scopes of the studies, as well as the targeted participants. Indeed, different types of assessment may be called for under specific circumstances. There is no one-size-fits-all solution to the measurement of loneliness in bereavement.

On the Use of an Indirect or Direct Measure

Another issue that the scope of the reviewed studies raises is whether one should ask directly about being lonely, or whether some proxy is preferable[1]. On the one hand, one might argue that people in general—and perhaps bereaved people in particular—are likely to know whether or not they feel lonely (and/or how intensely); thus, there is no clear detriment to asking directly about loneliness. However, on the other hand there is risk of social desirability bias, and demand characteristics or perceived expectations may prevail (e.g., a societal norm may prevent someone from admitting to being lonely). In this case, indirect questions may garner more accurate information about loneliness from respondents than direct questions. An example of an indirect item (from DJG-LS) is: "I miss having a really close friend."

Apart from the content of the items, we noted that response categories used in the scales and single/few-item measures varied among the reviewed studies, ranging from simple "yes"/"no" options to Likert scale usage and to further sub-categorization (e.g., collapsing a larger range of scores into three categories, such as "not lonely," "moderately lonely" and "strongly lonely"). Again, there would be advantages to coming to agreements where possible on this point, to enable comparison of results across studies. More research is needed to evaluate the pros and cons of different response option formats.

Comparing Measures

To work toward selection of the most appropriate, reliable, and valid measurement of loneliness for any particular investigation, one strategy would be to compare results on the existing instruments with single- or few-item measures. None of the studies we reviewed used both single/few-item measures in addition to validated scales.[2]. For practical purposes, if a short measure is indicated, the best-loading items of emotional and social loneliness could be selected. However, this may result in a further expansion of loneliness measures. On the other hand, a validated pool of items could be established from which clinicians and researchers may select. In addition, we refer above to the review of loneliness scales by Cramer and Barry (1999), a valuable source for considering the relative merits of different assessment instruments.[3]

It would also be useful to examine the benefits of the self-constructed items, that is, ones created by the investigators themselves, based on their understanding of "loneliness" (**Table 4**) compared with those selected from the established, more generic scales (**Table 3**), such as the Center for Epidemiologic Studies Depression Scale (CES-D) or Symptom Checklist-90 (SCL-90), ones which have also already been used in bereavement research. This would help to improve standardization and optimization of single/few-item measurement of loneliness across studies, allowing for comparison across study contexts and (sub)populations.

Limitations

Additional limitations to the data set and our review process need to be mentioned. First, our criteria limited the scope of investigation: notably, they restricted the review to adults, excluding bereaved children; only pre-COVID-19 studies were covered; only English language articles were included. Second, our chosen focus was on bereavement and not on other situations or diagnostic categories; comparisons across such domains would also be enlightening (for review of relationships between loneliness and psychiatric as well as physical disorders: Mushtaq et al., 2014). As a result of these selection criteria, the number of participants in the studies reviewed ranged from 30 to 2,018, and most participants were bereaved through loss of their spouse. The average age of sample participants, as noted above, was 65, limiting our ability to examine loneliness in bereavement across different age groups. However, there was no preferred use of specific assessments with regards to study-size or target group. Finally, regarding the available studies: to the best of our knowledge, no scientific investigation has yet been undertaken to compare the relative usefulness of the different measures to assess loneliness in bereavement. For recommendations to be made, such systematic, comparative investigations of the measures are needed.

CONCLUSION

The variety of assessment tools used in the reviewed studies makes it difficult to come to conclusions about the prevalence, intensity, and influence of loneliness in bereavement. Taking the effect of loneliness on quality of life and health seriously, selection of the most reliable, valid, and feasible measure(s) is needed to

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[1] Useful discussions relating to this issue have appeared in the literature (Cramer Cramer, K.M., and Barry, J.E. (1999). Conceptualizations and measures of loneliness: A comparison of subscales. *Personality and Individual differences* 27(3), 491-502. doi: https://doi.org/10.1016/S0191-88699800257-8.) (e.g., ibid., Victor et al., 2005). Measuring loneliness in later life: a comparison of differing measures. *Reviews in Clinical Gerontology* 15(1), 63. doi: https://doi.org/10.1017/S0959259805001723, van Tilburg et al. (2020). Loneliness and Mental Health During the COVID-19 Pandemic: A Study Among Dutch Older Adults. *The Journals of Gerontology: Series B*, gbaa111,. doi: https://doi.org/10.1093/geronb/gbaa111.

[2] For further information from a recent non-bereavement-specific study of loneliness which did so, see von Soest et al. (2020). Development of loneliness in midlife and old age: Its nature and correlates. *Journal of Personality and Social Psychology* 118(2), 388-406. doi: http://dx.doi.org/10.1037/pspp0000219, ibid.

[3] For a review on loneliness measures in children and adolescence, see Cole et al. (2021). A Systematic Review of the Development and Psychometric Properties of Loneliness Measures for Children and Adolescents. *International Journal of Environmental Research and Public Health* 18(6), 3285. doi: https://doi.org/10.3390/ijerph18063285.

move the field forward (e.g., toward finer-grained examination of variables/mechanisms influencing loneliness or for planning psychotherapeutic intervention). While our original aim in reviewing the assessment of loneliness was to determine which (possibly adapted) instrument might be chosen for universal, common use, closer examination of the instruments suggested that "one size does not fit all"; different measures are needed for different purposes, and for application among different subgroups. Thus, our review should be taken as a source of presently available information, as well as a discussion of the issues currently facing researches of loneliness in bereavement. Additional research and reflection is needed to work toward more concrete guidelines to establish which instrument to use in what context; our inventorization is a suggested step in this direction.

AUTHOR CONTRIBUTIONS

AV and MS drafted the manuscript. PB and KB discussed and commented on the text, providing suggestions for improvement. All authors were involved the conceptualization and discussion as well as critical revision of the manuscript and approved the final version of the article.

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Practical Measures for Dealing With the Struggles of Nurses Caring for People With Amyotrophic Lateral Sclerosis Comorbid With Cognitive Impairment in Japan

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Amyotrophic lateral sclerosis (ALS) is a devastating neurodegenerative disease for which there is currently no cure. This study aimed to explore the situations with which nurses struggled, their implemented practical measures, and the challenges they experienced when caring for patients with ALS comorbid with cognitive impairment (hereinafter, targeted patients). In this qualitative study, we conducted a survey with nurses (n = 121) experienced in caring for ALS patients; the survey contained a free-writing section in which participants described their struggles regarding care delivery for these patients. To collect data on practical measures that nurses had already implemented or wanted to propose regarding care delivery for the targeted patients, we conducted four focus group interviews (n = 22). We used a qualitative inductive approach to extract the categories. Fifty-eight nurses (49.6%) completed the free-writing survey section. The situations in which nurses struggled in care for the targeted patients were organized into three categories: “Patients’ strong persistency on specific requirements for nursing assistance in their daily lives,” “Patients’ problematic behaviors toward nurses,” and “Struggles in communicating with and understanding patients’ wishes.” Nurses reported these situations as stressful, and they affected care quality. The practical measures implemented when caring for the targeted patients were organized into five categories: “Cognitive impairment assessment,” “Care delivery to deal with patients’ strong persistency on specific requirements for assistance in their daily lives,” “Communication,” “Supporting the decision-making process,” and “Collaboration between the hospital and the community.” Multidisciplinary collaboration in the hospital, and collaboration between the hospital and the community from an early stage is necessary to share the results of the assessment and diagnosis of cognitive impairment. Our evidence underlines that guideline and care manual establishment may lead to improved care delivery and to the unification of care deliveries to respond to patients’ strong persistency.

Keywords: amyotrophic lateral sclerosis, cognitive impairment, comorbid, nursing, neuroscience, care

INTRODUCTION

The prevalence of neurodegenerative diseases increases with age (De Ronchi et al., 2005). Recent data shows that the number of patients with amyotrophic lateral sclerosis (ALS) has been increasing, albeit it remains a rare disease (Talbott et al., 2016). ALS is a devastating neurodegenerative disease that affects the motor system, being characterized by progressive neuro-deterioration (Miller et al., 2009). It can evoke upper and lower limb dysfunction, dysphagia, and speech impairments, making activities of daily life arduous (Hobson and McDermott, 2016). Unless tracheostomy positive pressure ventilation (TPPV, i.e., invasive mechanical ventilation, which requires delivering breath through a tracheostomy tube) is initiated for dealing with patients' respiratory failure, ALS patients usually die within a 3- to 5-year period (Miller et al., 2009).

Meanwhile, the prevalence of multimorbidity among older adults is expected to increase in the near future. From 2015–2035, the proportion of individuals with more than four diseases is expected to almost double, and two-thirds of the patients are expected to have some sort of cognitive impairment and depression (Kingston et al., 2018).

Despite the traditional view that ALS is a neurodegenerative disease that does not impact cognition, it is often comorbid with cognitive impairments (Goldstein and Abrahams, 2013). Specifically, research shows that approximately 50% of ALS patients have cognitive dysfunction and 10–15% have dementia (Lomen-Hoerth et al., 2003). Frontotemporal dementia (FTD) has received much attention from scholars studying dementia in ALS patients, with up to 15% of FTD patients and 30% of ALS patients experiencing overlapping features (Lomen-Hoerth, 2011).

Numerous scholars have studied on either care delivery for people with cognitive impairment or for people with ALS. Caring for a person with either dementia or ALS is mentally and physically demanding (Bromberg et al., 2011; Maayan et al., 2014), and caring for a person with both is considered to be even more challenging. Psychological stress in people who provide care for ALS may impact well-being in ALS patients and, possibly, management of symptoms in ALS patients (Siciliano et al., 2017).

However, few studies have focused on care for the people who care for those who have ALS and cognitive impairment, even though there are numerous pathological, clinical, and genetic studies. The burden of care for caregivers of a person with both ALS and FTD is high (Lillo et al., 2012; Cui et al., 2015; Caga et al., 2019). Nurses, as professionals in both medicine and care, support many informal caregivers and collaborate with certified home caregivers on a long-term basis in delivering care for the ALS patients with cognitive impairment.

Moreover, Japan was shown to have a higher rate of patients with ALS undergoing TPPV when compared with other developed countries, and a subset of these Japanese patients was shown to extend their lives for more than 10 years (Tagami et al., 2014). The motor impairment severity may be associated with cognitive impairment severity (Chiò et al., 2019).

With advance in medical technology and care as well as the advent of a super-aged society, the number of ALS patients with comorbid cognitive impairment is expected to increase. To ensure that these patients receive optimal care, we see the need for clearly understanding the current status of nursing care delivery for ALS patients with comorbid cognitive impairment. The purpose of this study was to clarify the situations with which nurses struggled, effects of situations nurses struggled with in caring for these patients on the quality of care, the practical measures implemented or proposed by nurses, and the challenges they experienced when caring for patients with ALS comorbid with cognitive impairment.

MATERIALS AND METHODS

Design

This study was a cross-sectional, qualitative descriptive study in which a questionnaire survey and focus group interviews were conducted. In order to collect a wide range of data from a large number of nurses, we thought that a questionnaire survey method with a free-writing section would be suitable. Following this survey, focus group interviews (FGI) were conducted to collect a rich dataset, because focus groups facilitate open discussion sharing of nurses' experiences and opinions.

Sample and Data Collection

We conducted a cross-sectional survey using an anonymous questionnaire in November 2018. Through purposive sampling, we recruited 172 nurses experienced in care delivery for ALS patients from two hospitals, 38 home visiting nursing stations, and 11 public health centers in one prefecture in the Kanto region. Regarding questionnaire distribution, head nurses distributed them to hospital nurses, with potential participants having been asked to complete the questionnaire and return it in a collection box; meanwhile, home health and public health nurses received and returned the questionnaire by mail. The questionnaire asked participants to indicate their characteristics (workplace, years of nursing experiences, and the number of patients with ALS they have supported) and fill in the freewriting section on situations in which nurses often struggle to deliver care for ALS patients with comorbid cognitive impairment.

In total, we collected 117 valid questionnaires, with 58 of them containing responses in the free-writing section (response rate: 49.5%). Among participants, 32 (55%) worked in a hospital, 19 at home visiting nursing stations (33%), and 7 (12%) in public health centers that care for patients with intractable neurological diseases. The mean number of years of nursing experience was 16 years (range: 6 months to 32 years). Ten nurses (17%) cared for one to five ALS patients, 14 nurses (24%) cared for six to ten patients, and 34 nurses (57%) cared for more than 11 ALS patients.

One year after a questionnaire survey, FGI were conducted to collect data on the care delivery experience and practical knowledge of nurses experienced in caring for the targeted patients. FGI is a qualitative methodology used to conduct in-depth data collection through group dynamics; it enables scholars to obtain detailed data inaccessible through quantitative research methods, as well as broad and dynamic data inaccessible through solo interviews (Anme, 2001). The participants were recruited from the same facilities as in the questionnaire survey. The directors or hospital head nurses informed their staff about the study. The inclusion criterion of FGI was to have experience in support for patients with ALS. In total, 23 individuals expressed interest in participating in the focus group interviews. However, one nurse could not attend to the interview because of an emergency call. Accordingly, 12 home care nurses, five hospital nurses, one nursing consultant, two nursing educators, one neurologist, and one medical social worker were included in the study. One participant was in her 30s, and all the others were in their 40s or 50s. All but one of the participants were female. To avoid uneven distribution of work locations, they were divided into four groups, with each group ranging from five to six participants.

Prior to discussion onset, the first author shared the results of the questionnaire survey (i.e., on situations with which nurses struggles in caring for the targeted patients) with the participants in order to get the discussion started smoothly and to have participants review the survey results. Each group was moderated by T.O., H.K and two other research team members who were a nurse certified by the Japanese Society of Intractable Disease Nursing.

The moderator followed an interview guide, which included the following questions: "Introduce yourself and, if you have already faced any situations with which you struggled regarding care delivery for ALS patients comorbid with cognitive impairment aside from those in the survey results report, share them with us;" "Share practical measures you have implemented, may want to propose, and discuss what would be better strategies;" and "Share any challenges you perceive regarding care delivery for ALS patients when they have a comorbid cognitive impairment." The moderators raised follow-up questions based on participants' responses and discussions; each focus group interview lasted approximately 60 min. The data source was the record made by the scribe, supplemented by each moderator of the groups to make it easier to read or understand.

Data Analysis

We conducted descriptive statistics for examining the following participants' characteristics: the number of nursing care experiences, the number of ALS patients they delivered care for, and their workplaces. We used qualitative inductive analysis for assessing the responses to the free-writing survey section. This qualitative data analysis was carried out based on the method proposed by Greg (2007), which aims to clarify actual situation from the emic viewpoint, and is suitable for this study.

The procedure of the analysis was as follows. All responses were reviewed several times to obtain the sense of the whole. The first author broke down each description into one sentence with one meaning. These descriptions were categorized by assessing their similarities and differences to extract subcategories. Similar subcategories were grouped together to derive categories. All co-authors examined the first author's analysis processes separately, and repeatedly discussed and confirmed the appropriateness of the descriptions, subcategories, and categories. Afterward, all authors scrutinized the relationship between situations and their influences on care quality, and expressed these relationships visually through a diagram.

Similarly, we used the qualitative inductive approach for the FGI records. The records from each group were merged for each question. Each description was shortened to one sentence with one meaning, ensuring that no meaning would be lost. Those descriptions were then categorized into subcategories and categories based on their similarities and differences.

Rigor

Trustworthiness or rigor of a study refers to the degree of confidence in data, interpretation, and methods used to ensure the quality of a study (Polit and Beck, 2014). This study employed constant comparative analysis to ensure trustworthiness for analysis of both the survey data and the interview data. The codes, subcategories, and categories were repeatedly scrutinized back and forth. The contents and classification of the categories were discussed with the authors with a master's degree or Ph.D. degree who had substantial experience with qualitative-inductive analysis and further validated by an experienced researcher. In addition, member checking was employed for the survey analysis. Member checking, participant validation is a technique for exploring the credibility of results (Birt et al., 2016). The report followed the Standards for Reporting Qualitative Research (O'Brien et al., 2014).

Ethical Considerations

This study was approved by the ethics committee of the institution to which the first author is affiliated. On the cover of the questionnaire in the survey, participants were informed study aims, methods, and ethical considerations. Specifically, they were assured that confidentiality would be maintained, participation and withdrawal from research were entirely voluntary, and that informed consent would be considered to have been granted with the return of the questionnaire. For the FGI, before their onset, we explained the same items to potential participants and obtained informed consent from all the participants.

RESULTS

Situations With Which Nurses Struggled in Care for People With ALS Comorbid With Cognitive Impairment

The data analysis for the free-writing section in the survey yielded three categories, nine subcategories, and 52 descriptions. Hereinafter, subcategories are denoted by 《 》, and descriptions are shown in 〈 〉 (**Table 1**). We describe each category below, with explanation by using some main descriptions.

Category: Patients' Strong Persistency on Specific Requirements for Nursing Care

This category consisted of three subcategories. The first was 《Frequent and time-consuming positioning caused by patients' persistency in body posture change》, which was extracted from four descriptions; 〈It takes a lot of time to get some patients to be

TABLE 1 | Situations with which nurses struggle in caring for people with ALS comorbid with cognitive impairment.

Category	Subcategory
Patients' strong persistency on specific requirements for nursing care	• Frequent and time-consuming positioning caused by patients' persistency in body posture change
	• Struggles in helping patients accept care delivery changes related to disease progression
	• Nurse exhaustion owing to time consuming care delivery caused by high cre demand and detailedness
Patient's problematic behaviors toward nurses	• Patient harassment toward nurses
	• Preference for particular nurses
Struggles in communicating with and understanding patients' wishes	• Struggles in helping patients understand, and assessing their comprehension of, ALS
	• Struggles in communicating with patients owing to patient forgetfulness
	• Struggles in supporting patients' decision-making regarding medical interventions
	• Concerns about increased burden on family caregivers who must make proxy decisions

satisfied with the body positioning⟩, ⟨Some patients are unusually picky about changes in their body position⟩, ⟨The frequent demands for repositioning from one ALS patient interferes with nurses' care delivery for other patients⟩, and ⟨Some patients frequently press the nurse call button to request fine, millimetric repositioning no matter what time⟩.

The second subcategory was ⟪Struggles in helping patients accept care delivery changes related to disease progression⟫, which was extracted from five descriptions including, ⟨They have a strong desire to eat and drink, despite the risk of aspiration⟩, and ⟨Some patients want to use the regular toilet even if they are fully dependent on assistance⟩.

The third subcategory was ⟪Nurse exhaustion owing to time-consuming care delivery caused by high care demand and detailedness⟫, which was extracted from seven descriptions; for example, ⟨The burden of care is so high that nurses become exhausted⟩, ⟨Nurses take much time to satisfy patients' requests because these patients are very detailed in their care requirements⟩, and ⟨Nurses spend so much time responding to the request of one patient that they have no time to care for others⟩.

Category: Patients' Problematic Behaviors Toward Nurses

This category consisted of two subcategories. The first was ⟪Patient harassment toward nurses⟫, which was extracted from four descriptions; ⟨There is rejection and verbal abuse toward unexperienced nurses⟩, ⟨Some patients treat nurses like idiots⟩, ⟨Some patients harass nursing staffs they do not like⟩, and ⟨Several nursing staff are experiencing psychogenic symptoms owing to harassment from patients⟩.

The second subcategory was ⟪Preference for particular nurses⟫, which was extracted from three descriptions; for example, ⟨Some patients interfere with the nursing care of other patients owing to preferring a particular nurse⟩, and ⟨Some patients require to change the nurse, because they don't like that nurse⟩.

Category: Struggles in Communicating With and Understanding Patients' Wishes

This category comprised four subcategories. The first subcategory was ⟪Struggles in helping patients understand, and assessing their comprehension of, ALS⟫, which was extracted from three descriptions; ⟨I had to explain to the patient the nature of one's disease many times because of one's poor understanding⟩, ⟨They stubbornly believe that they will be able to do what they were able to do before they got sick.⟩, and ⟨They cannot understand that ALS causes dysphagia and gait problems.⟩.

The second subcategory was ⟪Struggles in communicating with patients owing to patient forgetfulness⟫, which was extracted from 13 descriptions, including ⟨They struggle to convey what they want owing to their forgetfulness⟩, and ⟨They have difficulty selecting characters on the communication board owing to patient forgetfulness, even though their hands can point out letters⟩.

The third subcategory was ⟪Struggles in supporting patients' decision-making regarding medical interventions⟫, which was extracted from nine descriptions; for example, ⟨We need to consider dangerous actions, such as deliberate self-extubation, to support the decision-making process⟩.

The fourth subcategory was ⟪Concerns about increased burden on family caregivers who must make proxy decisions⟫, which was extracted from four descriptions, such as ⟨We thought that the cognitive decline and the progression of ALS would make it difficult for family caregivers to confirm the patient's preferences for ventilator use, which would lead to an increased burden on the family⟩.

We further clarified how these three situations affected care quality by carefully examining each description, extracting the content that indicates the influence on care. These situations evoked dilemmas for nurses, as they were caught between meeting patients' needs and ensuring patient safety. They also interfered with care delivery to other patients, evoked the physical and mental exhaustion of nurses, and led to an increasing number of stressful situations for nurses. Hence, these situations

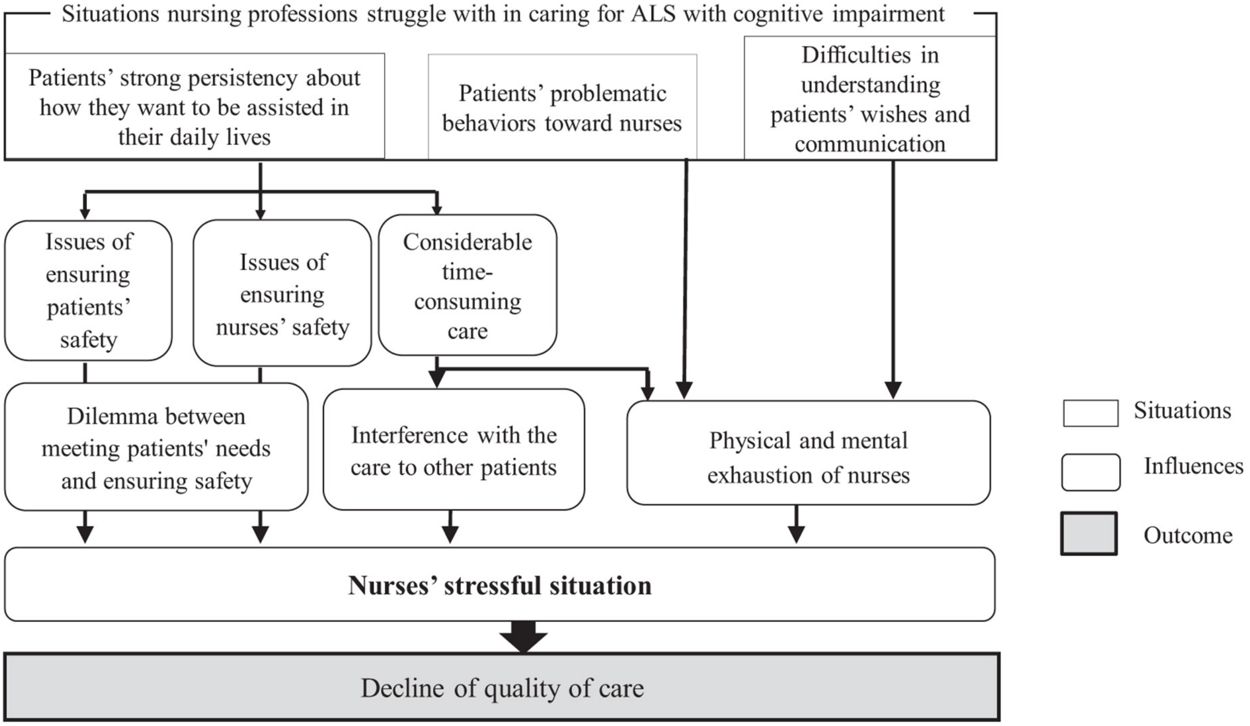

FIGURE 1 | Effects of situations nurses struggle with in caring for the patients with ALS and cognitive impairment on the quality of care.

may have had a negative effect on care quality, as shown in **Figure 1**.

Practical Measures Implemented or Proposed by Nurses

Regarding practical measures implemented or proposed by nurses for care delivery to patients with ALS comorbid with cognitive impairment, we extracted five categories, 13 subcategories, and 33 descriptions (**Table 2**).

Category: Cognitive Impairment Assessment

This category comprised four subcategories. The nurses mentioned that they paid attention to changes in both physical and cognitive aspects and collected information on patients' cognitive impairment from a multifaceted perspective; specifically, from the perspective of family members and other professionals. Additionally, they mentioned conducting objective cognitive assessments periodically.

Category: Care Delivery to Deal With Patients' Strong Persistency on Specific Requirements for Assistance in Their Daily Lives

This category comprised three subcategories. The first was ⟪To unify the care methods for responding to a patient's persistence in specific care requirements among care providers⟫, which was extracted from two descriptions; ⟨The contents of care to be provided should be unified among nursing staffs⟩, and ⟨It would be good to create a manual to unify the care method among all care providers⟩. The second was ⟪To respond to position change requests with large movements⟫, which was extracted from one description, ⟨It is a good practice to move the patients significantly, before making fine, millimetric adjustments⟩. The third subcategory was ⟪To ensure that nurses' considerations reconcile with patients' wishes⟫, which was extracted from one description, ⟨We need to discuss with the patients, in advance, about when and the amount of time we can be available for them, and acquire their consent⟩.

Category: Communication

This category comprised two subcategories: ⟪To create a verbal way to explain the cognitive impairment⟫, and ⟪To devise communication methods through non-verbal means⟫. If nurses tell the patients that they suffer from dementia, the patients may express that they no longer want to go to the hospital for examination, so home care nurses try to talk to them creatively. An example would be something like, ⟨People of your age need to check for brain atrophy owing to aging⟩. Nurses also mentioned that, ⟨Verbal explanations alone will not solve the problem, so we use gestures and facial expressions, creating non-verbal ways to explain something⟩.

Category: Supporting the Decision-Making Process

This category comprised one subcategory: ⟪To keep in mind the implementation of advance care planning in hospital and home care settings⟫. As the nurses experienced ⟪Struggles in supporting patients' decision-making regarding medical interventions⟫, they described the following: ⟨We need to give opportunities for family members to discuss about

TABLE 2 | Participants' reports on practical measures to care for ALS patients comorbid with cognitive impairment.

Category	Subcategory
Cognitive impairment assessment	To pay more attention to cognitive aspects
	To understand how much patients have changed by asking about it to their family
	To confirm patient's cognitive impairment with other professionals
	To conduct objective evaluations periodically
Care delivery to deal with patients' strong persistency on specific requirements for assistance in their daily lives	To unify care methods for responding to a patients' persistence in specific care requirements among care providers
	To respond to position change requests with large movements
	To ensure that nurses' considerations reconcile with patients' wishes
Communication	To create verbal way to explain something
	To devise communication methods through non-verbal means
Supporting the decision-making process	To keep in mind the implementation of Advance Care Planning in hospital and home care settings
Collaboration between hospital and community	To strengthen the multidisciplinary collaboration in the hospital and the collaboration between the hospital and the community
	To restructure care methods when patients are hospitalized for respite care or clinical examination
	To ensure the collaboration between the hospital and the community regarding decision-making support on patient's medical treatment

patients' wishes before cognitive impairment onset⟩ and ⟨We need to support the decision-making processes and connect with the community⟩.

Category: Collaboration Between the Hospital and the Community

This category comprised three subcategories: ⟪To strengthen the multidisciplinary collaboration in the hospital and the collaboration between the hospital and the community⟫, ⟪To restructure care methods when patients are hospitalized for respite care or clinical examination⟫, and ⟪To ensure the collaboration between the hospital and the community regarding decision-making support on patient's medical treatment⟫. The participants remarked that in hospitals, unlike home care settings, they deal with numerous patients, hindering the ability of hospital staff to respond to patients' individual wishes in detail. Still, the hospital setting was also described to be a facilitator of nurse-physician collaboration. In this situation, patients often have little choice and are forced to accept nurses' requests to change care methods.

Moreover, the nurses described that, while most patients stay in hospitals for short periods, hospital staff can be involved intensively in and available for 24 h a day to caring for patients. Accordingly, our study participants remarked that the strength of hospital care should be utilized to assess patients' cognitive function level. They also emphasized the need to restructure care methods upon patient hospitalization for clinical examination or respite care; they believed that this may help to ensure that the care delivered is relevant to one's physical independence level. For this to be operationalized, they remarked the need for a greater collaboration between the hospital and the community.

Challenges in Care for ALS Patients With Comorbid Cognitive Impairment

Two categories were extracted from the analysis of FGI records: challenge of integrating *cognitive impairment assessment* into ALS care and challenge of *unification of nursing staff to respond patients in the same way*. The former was extracted from one subcategory, ⟪Challenges to implement the cognitive impairment assessment⟫. This subcategory had two descriptions: ⟨The content of neuropsychological tests is too demanding and I am not comfortable conducting them⟩, and ⟨When family members are asked to perform neuropsychological tests at home, it is necessary to consider the influence of the family's psychological situation⟩.

The second category comprised two subcategories. One subcategory was ⟪Challenges in responding to and experiencing dilemmas regarding patient requests among nurses⟫, which was extracted from four descriptions, including ⟨There are many times when the nursing staff disagree on how to care for the patient⟩, ⟨There are nurses who have a strong sense of mission to care for ALS patients and try to meet all their needs to any extent⟩, and ⟨The nurses always feel uncomfortable and face dilemmas about meeting all patient requests⟩. The second subcategory was ⟪Challenges in responding to requests from other care providers to comply with the patient's requests⟫, which was extracted from two descriptions; ⟨Nurses may struggle to deal with the family members who wish to fulfill patient's requests that seem unreasonable⟩, and ⟨Nurses may often be requested to care for the patient's needs and reminded that this is the responsibility of nurses by other care providers, although it may be difficult to deal with patients' needs〉. The value of people involved are diverse, and unifying them is a heavily difficult task.

DISCUSSION

This study clarified that the situations in which nurses struggled to care for the targeted patients were related to the following: patients' strong persistency on specific requirements for nursing assistance in their daily lives, patients' problematic behaviors toward nurses, and struggles in communicating with and understanding patients' wishes. These situations increased nurses' stress and may have facilitated the deterioration of care quality. Moreover, we extracted five practical measures to deal with these some problems and two challenges that nurses may experience. Those findings were integrated into the following discussion for better care of ALS patients with cognitive impairment.

Need to Increase Awareness of Nurses Regarding Cognitive Impairment in ALS

In this study, the participants reported struggling with patients' persistency on specific requirements regarding their nursing care. Indeed, the non-adherence to treatment of individuals with comorbid ALS and cognitive impairment was shown to be twice as high as that of ALS without dementia (Cui et al., 2015). Thus, caregivers of such patients often experience a heavy care burden (Lillo et al., 2012; Caga et al., 2019). Moreover, FTD was shown to be associated with personality changes, irritability, poor insight, perseverance, obsession, disinhibition, and altered social conduct (Phukan et al., 2007). Despite the scientific evidence on these associations, on the practical setting, nurses may interpret some of these behaviors as stubborn expressions of patients' autonomy (Bromberg and Bromberg, 2017), potentially not recognizing that the patients experience not only ALS but also a cognitive impairment. Therefore, we suggest for nurses to pay more attention to the possible presence of FTD in ALS patients.

In concordance with this suggestion from our study, a past research described that, in clinical settings, Japanese neurologists should pay more attention to the cognitive and behavioral aspects of patients with ALS (Watanabe et al., 2020). Another study conducted with Japanese nurses clarified that, although 60% of them were aware of the cognitive impairments associated with ALS, they only learned about this within the last 1 or 2 years and through their own clinical experience (Ushikubo et al., 2021). Therefore, it seems that both nurses and neurologists need to pay more attention to cognitive function in ALS patients.

Conducting Appropriate Assessment of Cognitive Impairment in Delivering Care for ALS Patients

Our study participants struggled with patients' problematic behaviors, such as harassment. In relation to this finding, a prior study has assessed coping strategies of neurology nurses experiencing abuse from patients and families, identifying that over 96% of the nurses reported some form of verbal abuse and over 60% reported some form of physical abuse; to deal with this abuse, nurses utilized avoidance coping strategies (Trahan and Bishop, 2016). The literature recommends that nursing professionals should develop nursing practice that eliminates the use of avoidance as a way of dealing with abuse or these problematic behaviors (Trahan and Bishop, 2016). Thus, we see a critical need for assessing patients with ALS for cognitive impairment; this may support nurses, other healthcare professionals, and informal caregivers by ensuring that they have more information on and are better prepared to care for these patients. This may help reduce the stress surrounding patient care in such complex settings.

Hence, it seems important for nurses to acquire skills that allow for them to evaluate different types and levels of cognitive impairment. On the topic, the literature recommends professionals to use several batteries of evaluations for ALS-FTD, which include the Mini-Mental State Examination (Folstein et al., 1975), the Frontal Assessment Battery (Dubois et al., 2000), the Japanese version of the ALS-FTD Questionnaire (Watanabe et al., 2016), and the Montreal Cognitive Assessment (MoCA) (Nasreddine et al., 2005). Of these, the MoCA has been regarded as the most sensitive clinical scale for evaluating cognitive impairment in ALS (Nagashima et al., 2019). Meanwhile, in our study, the nurses pointed out cognitive impairment assessment as a challenge, reporting many barriers to this end. For example, they observed that the motor (e.g., hand weakness, dysarthria) or verbal communication deficits often found in ALS patients may interfere with the performance of cognitive impairment tests. Furthermore, the application of a screening test may upset or even offend some patients, and so careful consideration is required.

Understanding Patients' Wishes in the Communication Difficulties

Moreover, nurses struggled to communicate with and to understand patients' wishes. When dealing with ALS patients, healthcare professionals often need to introduce treatments in a timely manner to ensure symptom alleviation and optimize conditions (Hogden et al., 2021). To ensure that this is done according to patient's wishes, ALS patients need to understand their medical conditions and be able to decide about their medical treatment. Health care professionals identified that the patient's acceptance of the disease and its progression by both patient and caregiver were one of the most important factors to support decision-making (Martin et al., 2016). Surrounding this topic, a prior study showed how the inability of patients to communicate may lead to their family members making proxy decisions, placing a great burden on the family (Foley and Hynes, 2018). To deal with this issue, the nurses in our current study reported attempting to communicate with patients through verbal and non-verbal methods. Thus, we suggest the need to provide specialized communication support for nurses dealing with both cognitive impairment and bulbar symptoms of ALS patients. Multidisciplinary collaboration, including occupational and speech therapists may be essential in such settings (De-Bernardi-Ojuel et al., 2021). Furthermore, past research remarked the need to integrate advanced care planning ever since the diagnosis, as this may allow for family caregivers to be relieved from the burden of making proxy decisions and for patients to get involved in the decision-making process regarding their medical treatment (Bromberg and Bromberg, 2017).

Seamless Care With Multifaceted Perspective and Multidisciplinary Cooperation Between Hospital and Community

This study showed that community collaboration is a potential practical measure to be used by nurses for both decision-making process about medical interventions and care method changes. Based on our participants' remarks, collaboration between the hospital and the community and multidisciplinary collaboration is essential to understand the patient's value, preferences, and goals to support their advance care planning (Seeber et al., 2019).

In the home care setting, nurses are easier for listen to the true feelings of the patients. However, it is also easier for patients to escalate their demands and home care nurses experience difficulties in asking patients for changes in care delivery. Therefore, the optimal way for delivering care to such patients may be to use a seamless, multifaceted perspective and multidisciplinary cooperation. This may allow for stakeholders to use the strengths of both the hospital and the community and to cover for their respective weaknesses.

Limitation

There were several limitations in this study. The FGI was very active, so we could have used a longer timeframe, which could ensure that we reached data saturation. In addition, to determine whether there are other situations with which nurses struggle with and whether nurses utilize other types of practical measures, we suggest for future studies to include larger sample sizes and a sample that is representative of various Japanese regions. As ALS is a rare disease as designated by the Japanese government and it is even more difficult to secure a sufficient number of nurses with experience in supporting ALS patients with cognitive impairment, this study employed purposive sampling to efficiently secure participants. Furthermore, the sampling was limited to the region as this research planned to conduct FGI following the questionnaire survey. We recommend to conduct a national survey or to recruit participants with the cooperation of several regional nursing associations or home care nurse councils. Those may be able to secure a larger number of participants. Even if participants are dispersed all over the country, it is feasible to hold FGI or individual interviews as online meetings grow in popularity.

Conclusion

This qualitative study clarified three types of situations with which nurses struggled during the care for people with ALS comorbid with cognitive impairment, as well as five categories of practical measures. Further, nurses deemed that the cognitive impairment assessment for ALS patients and the unification of care deliveries to respond patients' strong persistency were challenging issues. The multidisciplinary collaboration between the hospital and the community may be important to ensure that the diagnosis of cognitive impairment is appropriately shared between stakeholders and to ensure that they are aware of the condition of these patients. In addition, all nurses need to respond to patients' excessive demands and patients' harassment to nurses in same way. The people involved with patients have different care policies, such as establishing a standard care for all, under their various views, but their unification is important. This study suggests to establish care guideline or care manuals, which may lead to the provision of an optimal care. It needs to include cognitive impairment assessment, unification of care delivery to respond to patients' strong persistency, coping for problematic behavior to nurses, communication that covers both language and cognitive impairments caused by ALS, and the decision-making support, by collaborating between hospital and community.

DATA AVAILABILITY STATEMENT

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

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Ethical Review Board for Medical Research Involving Human Subjects in Gunma University. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

MU conceptualized the research, designed the study, gathered and analyzed the data, and drafted the manuscript. EN, TO, and HK contributed with data collection, data analysis, and a critical revision of the manuscript. All authors contributed to the article and approved the submitted version.

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Explore the Bereavement Needs of Families of Children With Cancer From the Perspective of Health Caregivers: A Qualitative Study

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Introduction: The experience of bereavement is associated with severe physical, psychological, social and spiritual reactions in the parents of children with cancer. Because of that, the families of these children need to receive bereavement services. The aim of this study was to explore the bereavement needs of families of children with cancer from the perspective of health caregiver as people who have a close relationship with the child and the family.

Methods: This qualitative descriptive study design in. In total 15 semi-structured interviews were conducted using a purposive sampling in 2018. Interviews were recorded and transcribed and conventional content analysis was used to analysis the data. The Trustworthiness of the data were assessed according to the criteria of Lincoln and Guba.

Results: From the data analysis, needs of the bereaved family were categorized in three dimension including "achieving peace," "Abandoned family access to care," and "continuing care." The category of "achieving peace" includes spiritual and existential support, companionship with the family, contact with other bereaved families, support in passing and accepting the bereaved and continuing empathetic communication with the family, the category "Abandoned family access to care" includes the promotion of family self-control, awareness of end-of-life care to the family, and the category of "continuing care," includes formal and informal family care and individualized care.

Conclusion: It is necessary for the care team to pay special focus to family considering the needs of the family about the death of the patient and the challenges of the family bereavement period. It is recommended that members of the health care team should be trained in assessing family needs, identifying risks of adverse outcomes, continuing care, and providing resources during bereavement. The needs of the bereaved family should also be addressed in their care plan.

Keywords: pediatric cancer, death, bereavement, family, grief, care, self-control

INTRODUCTION

Pediatric death by cancer is a serious problem worldwide, with more than 413,000 death caused in 2020 (The American Childhood Cancer Organization, 2021). Annually up to 2,000 children under 15 years are diagnosed with cancer in Iran (Pakpour et al., 2019). Cancer is the second leading cause of death in children under 14 in Iran. It causes about 4% of deaths of children under 5 and 13% of deaths of children aged 5–10 years (Kashani, 2012).

Cancer causes physical, psychological, social, economic, and emotional reactions for child and family (Zarit, 2004; Lu et al., 2009; Valizadeh et al., 2014) that children's reactions to these stresses include physical problems, depression, anxiety, fear, behavioral, and communication problems, decreased self-esteem and social interactions, and retrograde behaviors that are affected by the child's developmental stages and adaptability, duration of stress, and Its intensity (Kirsch et al., 2003; Yahia et al., 2015) and imposes a heavy burden of care on the family (Zaider and Kissane, 2010).

The death of a child due to cancer is a devastating event that leads to prolonged bereavement (Snaman et al., 2020), complex traumatic reactions to bereavement (Gilmer et al., 2012), dissolution of the attachment relationship with the child, and feeling guilty. Grief is defined as an internal experience in reaction to the loss of something loved and valued (Näppä et al., 2016). Parental grief is prolonged 5–7 and can intensify during significant periods (e.g., holidays), a concept known as "regrief" (Gilmer et al., 2012).

Bereavement is mourning with a strong sense of loss and sadness that occurs with death and is a process in which life continues without the presence of the person (Pakseresht et al., 2018). These severe and prolonged reactions to grief are common among family members but sometimes can be debilitating (Lichtenthal et al., 2015). All people grieve differently; some need professional help while others are resilient in their loss and do not require special interventions (Näppä et al., 2016).

Numerous studies have examined the psychological challenges such as depression and anxiety for parents with pediatric cancer (Kreicbergs et al., 2004; Kim et al., 2013), grief (Lannen et al., 2008), existential anxiety, identity, and spiritual challenges (Lichtenthal et al., 2015), guilt (Surkan et al., 2006), post-traumatic stress disorder, negative impact on copping, adverse effects on health, social interactions, and high levels of psychological distress that often require professional assessment and assistance (Gilmer et al., 2012; Kim et al., 2013). Parents who have lost a child to cancer, they feel that if they could prevent cancer or death, they often experience self-blame and guilt (Eslahkar et al., 2019). However, despite this misery, most grieving individuals do not develop mental and or physical complications. A minor number of significant other in the bereavement period show increased risk for hospitalization and death as well as depression, mental illness, and substance abuse (Näppä et al., 2016).

The presence of such symptoms, as well as the disease and its treatments side-effects, the high cost of treatment, psychological and social consequences of the disease of the child and family, require comprehensive care in the form of supportive care. This type of care supports the family and those around the dying or deceased child (Pakseresht et al., 2018). Communication, continuity of care by caregivers, as well as child's physical and mental symptoms management, which are very important for parents in the later stages of a child's life, are aspects of parental care during the bereavement period before the child's death (van der Geest et al., 2016). Family support at the end of a child's life and after death contributes to the bereavement process in families, as well as providing an opportunity for the family to share their feelings with those who understand them (Pakseresht et al., 2018). Attention to, emotional, physical, environmental and psychosocial needs are the criteria considered during family bereavement (Suttle et al., 2017). Despite this need, care for bereaved parents did not exist widely 10 years ago, and pediatric providers might not have knowledge, even when bereavement care programs exist (Spraker-Perlman et al., 2021).

According to a study, families who do not receive professional bereavement care live in a state of prolong uncertainty and anxiety. However, in addition to complex responses to loss, many parents have responded to this loss in constructive ways (Eslahkar et al., 2019). Parental demographics, such as age, gender, race, and ethnicity, affect the grief experience and bereavement outcomes (Snaman et al., 2020). One study found that parents over 30 years old showed better psychosocial adjustment than younger parents following the death of a child from cancer (Morrow et al., 1981). Reynolds et al. argued that grief and depression are normal emotions in bereavement, but that symptoms of grief resolve more slowly than medically treated depression (Reynolds et al., 2004).

Bereaved parents have identified a need for improved bereavement support, emphasizing the important role that the healthcare team and medical institution serve in their grief journey. Despite this need, care for bereaved parents did not exist widely 10 years ago, and pediatric providers might not have knowledge, even when bereavement care programs exist (Spraker-Perlman et al., 2021). There is a wide variety of bereavement programs where services can include a range of telephone or letter calling programs, grief support groups (siblings or parents), or meetings with professionals to discuss the death of a child or be the results of the autopsy (Widger et al., 2012; Yahia et al., 2015). Bereavement groups are believed to be beneficial as preventive interventions from social and economic standpoints. Participation is likely to be more acceptable and less threatening to potential recipients than professional interventions linked to psychiatry. Costs can be low, since groups are usually led by staff or volunteers rather than mental health professionals (Näppä et al., 2016). An experience not often discussed among providers is the personal feelings of grief that may come when a patient dies. Although providers of all disciplines have varied encouragement in engaging with the experience that may be personally felt when caring for those who are at end of life, the nurturing of individual self-care can be of benefit in regards to tenability and viability within such work (Jonas et al., 2018).

Among the developed countries, Britain has standards of bereavement care, published in 2001 (Kirsch et al., 2003; B. S.

A. a and Care, 2013). In Iran, a special, targeted, and codified care program is not designed for a caring family bereavement, and families are practically abandoned after the death of a child (Rassouli and Sajjadi, 2014). The only cases observed are limited counseling to families after the death of a child in some centers (Pakseresht et al., 2018), including the Sherwin Charity, which provides bereavement care for the family as a group therapy by volunteer psychologists, and Due to the lack of sufficient evidence in assessing the need for these services, the country's policymakers are not aware of the necessity and priority of such services (Knapp, 2009).

Since one of the best ways to improve the quality of care is to identify the needs of the patient and family from different dimensions, examining the current state of bereavement care in the country from the perspective of nurses who have the most contact with the patient and family can be a step toward development of this type of care in medical centers.

Awareness of the needs of the child and family during bereavement provides a unique care plan for each family and prevents wastage of time and money following unscheduled care. Therefore, the present study was conducted to explore the bereavement needs of families of children with cancer from the perspective of nurses.

METHODS

Design and Setting

In this present qualitative conventional content analysis 15 nurses were selected based on purposive sampling. This approach is used in situations where there has been previous research on a phenomenon but it needs further description (Potter and Levine-Donnerstein, 1999). Participants were selected from pediatric oncology ward of Mofid Hospital in Tehran (As a referral center from all regions of the country) and outpatient clinic of Shafa Hospital of in Ahvaz (**Table 1**). The research environment was the natural areas of employment or care in which study participants were routinely present.

TABLE 1 | Demographic characteristics of care providers.

No	Sex	Age (year)	Education (level)	Work experience (Years)	Interview time (Minutes)
1	Female	32	B.A.	3	45
2	Female	35	M.A.	2	27
3	Female	28	B.A.	2	31
4	Female	24	B.A.	3	26
5	Female	23	B.A.	2	15
6	Female	38	M.A.	9	17
7	male	32	B.A.	3	15
8	Female	27	M.A.	5	19
9	Female	42	B.A.	15	24
10	male	29	B.A.	4	25
11	Female	30	B.A.	3	24
12	Female	40	B.A.	14	18
13	Female	38	B.A.	10	33
14	Male	33	B.A.	10	37
15	Female	36	B.A.	12	21

Participants and Sampling

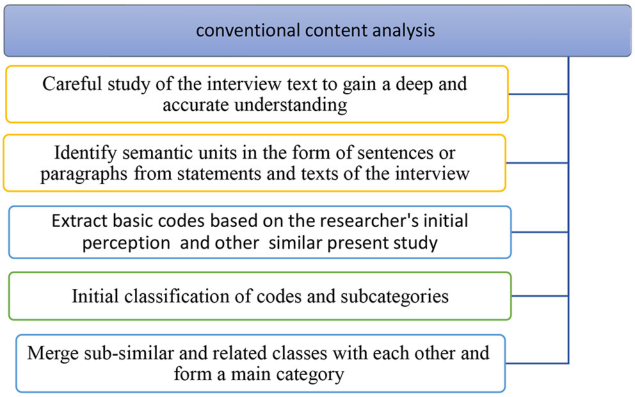
These were the 15 official nurses in the pediatric cancer ward who had the most contact with the bereaved families and were selected based on purposive sampling. One of them was the headers. In the study of Spraker-Perlman et al. (2021), 13 mourning parents participated in the study, which despite the small number of participants, rich results were obtained in the study (Spraker-Perlman et al., 2021). The inclusion criteria was having experience in pediatric cancer wards for at least two year, Being formal and willingness to participate in study. Exclusion criteria included reluctance to interview and its continuation, failure to deal with the bereaved families of children with cancer. The first participants were a 32-year-old nurse who had 3 years of experience in the pediatric hematology department of Mofid Hospital in Tehran. This nurse had a close relationship with children with cancer and their families and provided complete and comprehensive information about patients and families to the researcher and was eager to conduct the interview.

The reason for choosing nurses instead of bereaved parents was that the nurses were in contact with families who had a child with cancer at the end of life or all of bereaved parents went to the hospital. So they could identify different needs in different families. While a mourning family could only share their special needs with us. Another reason for not choosing breaved parents was the lack of appropriate mental conditions for the interview. Interviews with nurses were able to gain a professional perspective on the needs of bereaved families.

Data Collection

Semi-structured and face-to-face interviews were conducted by the first author in 2018. The first author was a doctoral student in nursing who had learned the method of interviewing during her studies from professors and the research team consisted of thesis supervisors and consultants who had sufficient experience in interview analysis. A guide to interview questions was prepared prior to the interview to gather comprehensive information. The interviews lasted between 15 and 45 min. All participants in this study were interviewed only once. Before conducting each interview, the researcher, by attending the research environment, while introducing himself/herself and explaining the nature and main purpose of the research, has identified the appropriate participant and while receiving written consent by specifying its provisions, the appropriate time and place agreed upon by the parties were determined based on the desire and preference of the participant. For the interview, the researcher introduced himself while explaining the main purpose of the research. Then the main question of the research was asked; "What is your experience in caring for families who have a child with cancer at the end of life or have lost their child?" What do you think are the needs of bereaved families? Probing questions then arose from experience of participants e.g., "What did you do as a health caregiver?" "Explain the current process of providing care for

TABLE 2 | Conventional content analysis.



conventional content analysis

Careful study of the interview text to gain a deep and accurate understanding

Identify semantic units in the form of sentences or paragraphs from statements and texts of the interview

Extract basic codes based on the researcher's initial perception and other similar present study

Initial classification of codes and subcategories

Merge sub-similar and related classes with each other and form a main category

TABLE 3 | Categories and subcategories.

Category	Subcategory
Achieving peace	Spiritual and existential support
	Companionship with the family
	Contact with other bereaved families
	Support in passing and accepting the bereaved
	Continuing empathetic communication with the family
Abandoned family access to care	The promotion of family self-control
	Awareness of end-of-life care to the family
continuing care	Formal and informal family care
	Individualized care

bereaved families" Was asked. Then the participant was asked to comment if they had any other points or questions. Interviews continued until data saturation. In this study, the needs of bereaved families at the end of child's life and also after the child's death were examined.

Data Analysis

In order to analyze the qualitative data, conventional content analysis was used. This method was proposed in 2005 by Hsieh and Shannon (2005). This approach is used in situations where there has been previous research on a phenomenon but it needs further description (Potter and Levine-Donnerstein, 1999). In the present study, the entire text of the interview was read several times until a deep understanding was obtained. Then parts of the text were identified, marked, and turned into the smallest significant units (code) based on the researcher's initial perception. In the next step, the parts that were marked were encrypted based on predefined codes. Each part of the text that did not fit into this initial encryption was given a new encrypt. The author is inspired by other subject-related studies for coding. For this purpose, first, the text of the files was entered into Word software and then carefully read several times while key phrases and sentences were underlined. Then the codes (the main opinion of the participants) were extracted. The first level of the coding process began with identifying the meaning units, e.g., words, phrases, themes, and sentences that have specific meanings. Then the main concepts in each unit of analysis were called semantic units (**Table 2**).

Rigor and Trustworthiness

The Trustworthiness of the data were assessed according to the criteria of Lincoln and Guba (1985). These criteria including credibility, confirmability, dependability and transferability. In this study, long engagement with data was done by spending enough time to collect and analyze data, also review the data by supervisors and consultants and was used. Furthermore, the data were carefully examined by an external observer. In order to achieve the confirmability, all stages of the research, especially the data analysis in all directions were recorded in detail so that if another researcher wants to continue research in this field can easily understand.. The type of research and the characteristics of the participants were well described in order to make a good judgment about the transferability of the readers.

Ethical Considerations

The present study is part of the doctoral dissertation of the first author of the study, which has been approved by the ethics committee of Ahvaz Jundishapur University of Medical Sciences with No.IR.AJUMS.REC.1394.764. Interviews and audio recordings were conducted after participants' awareness of the purpose of the study and informed consent. Participants were also reassured that the recorded information would remain confidential, and it was announced that they could be excluded from the study at any time "without any penalty of sorts."

RESULTS

A total of 400 codes were extracted from the interview analysis. The coding process was a continuous comparative analysis and duplicate codes was eliminated and similar codes was merged. Three categories named "achieving peace," "abandoned family access to care," and "continuity of care" and 9 subcategories were extracted (**Table 3**).

Category 1: Achieving Peace

Although the health care team provides information and advice to the family, they ultimately prefer to choose options that provide peace. Participants considered the importance of providing peace for the family during the bereavement process. Subcategories of this category included "Spiritual and Existential Support," "companionship with the family," "Contact with other bereaved Families," "Support in passing and accepting bereaved" and "continuous empathetic communication with the family" that they were considered soothing by some of the participants.

Spiritual and Existential Support

Based on participants' experience, spiritual and existential support has an effect on the sense of hope, value, purpose in connecting with others, and connecting to greater power in the child and family. "...Some people deny the illness of their child or family member due to the thought that it is a punishment from God... Sometimes their beliefs become so weak that they do not believe in anything. When you want to convince them that this is not the case, your work is a heavy burden...” (Nurse 1).

> “One of the mothers of a child with cancer in the late stages of midnight said loudly, ‘God, you say you are more compassionate than my mother, so why?’ A mother does not want her child to see her like this... so how do you want my child to die?” (Nurse 5).
>
> “In my opinion, the existence of a psychologist, religious counselor, clergyman in these areas where mortality is very high and very effective. When you yourself have a strong belief in this field, you talk to the patient who comes to you and you will realize how calm your patient is.... There were families whose patients were on the verge of death. We easily persuaded him to sit on his head and pray for him. Easily accepted” (Nurse 9).

Companionship With the Family

Families deserve to be accompanied during the bereavement period and should be allowed to talk about their grief. Sharing emotions during bereavement with a friend or loved one may not help because people often try to comfort and the bereaved person stops sharing emotions. Almost all participants in this study emphasized the effect of health caregivers’ behavior and their companionship on increasing or decreasing family morale. In the participants’ view, accompanying and talking to the care team is more effective than accompanying those around them during the bereavement period.

> “Let’s give the patients’ companion more time and opportunity to see the patient in bed so that they can communicate more easily with each other. This is the only thing that can be done to provide comfort for them.” (Nurse 3).
>
> “Family members in bereavement period before death are such that they accept everything we tell them and do not allow an unprofessional person from their friends or family to get too close to them... short our words are valid for them and, they like us to accompany them” (Nurse 8).

Contact With Other Bereaved Families

Participants believed that having communication with families who have similar conditions in the bereavement period is efficient and comforting.

> “We often find the contact numbers of families whose children have died of cancer, and we tell bereaved families to call them and see how they cope.... You do not believe that we often find out that families are together, Befriending and helping each other to get back to a normal life and even think about getting pregnant again.” (Nurse 6).
>
> “I wish that conditions would be created in the hospital for families who are in the period of bereavement to come together and talk to each other... Do you know that these families are ashamed to communicate with those around them because of their childish illness..... Cancer It is a taboo for them. But if they see a family with the same conditions, their suffering may be reduced”...(Nurse 11).

Support in Passing and Accepting the Bereaved

Support from family, social resources, and health care personnel can be efficient in overcoming the bereavement phase and making it easier to accept grief, as well as reducing the burden of bereavement on the family.

> “We have to know if the bereavement keeps the family away from those around or not. Can the family do other children’s chores to go to school? Or is his work messed up? Therefore, the family needs support in the bereavement period to meet all its needs.” (Nurse 5).
>
> “A patient who has died, we set up with colleagues to go to the grave of the child in groups. Then their families even go to the ward 6, 7, 10, and 11 months later and appreciate how good it was. We went through a difficult time, but your memories are still in our minds” (Nurse 15).

Continuing Empathetic Communication With the Family, the Category

According to nurses’ experience, establishing a good relationship with the family after the child’s death can make the acceptance of death easier for the family.

> “There are many cases where the personnel have communication with the families after the child’s death. In some cases even if the patient died 2 years ago, but the mother still calls in a state of denial and says, Tell me what happened that night. This connection can help the family to accept the death of their child.” (Nurse 7).
>
> “Personally, I cannot communicate with them afterwards. Now a series of colleagues call and talk, now they ask, but I cannot communicate at all, after this happens and goes away, I want to call and ask how he is, I cannot. I can’t do anything with myself. I cannot at all... After this case, I can no longer communicate with my mother. ‘It ’s as if I do not know what to say to them,’ which is why they’re cut off.” (Nurse 4).

Category 2: Abandoned Family Access to Care

This category emphasizes the abandonment of the family after the child’s death. According to the participants’ opinion, after the child’s death, the family is abandoned, and no care is provided. This category includes “improving family self-control” and “Family Awareness of End-of-Life Care.”

Promotion of Family Self-Control

Family self-control involves professional communication with the family so that the family retains or acquires a sense of control over the situation.

> “After child’s death, families are practically abandoned. A noticeable thing that they do is supporting the family after death, for example, they called the father of the family, and he said that ‘Come here, I need you.’ They talk to them psychologically and calm them down.” (Nurse 7).
>
> “Families are confused in this situation and do not perform many of their roles in the home and community properly.... In these cases, it is better to give families specific responsibilities

> and the nurse has the role of counselor in empowering the family." (Nurse 4).

Awareness of End-of-Life Care to the Family

Participants stated that families in the bereavement period need to obtain information and training from the treatment team on pre-and post-bereavement condition management and various end-of-life counseling. Most families had no proper method to control bereavement before and after the child's death.

> "For example, I have taught many times, and I say that 'well, try to be calm and trust in God and do not cry too much, because your child hears your voice even in the last moment of his life' after a while, they continue the same routine as before, and there is no change. There must be a training program that is taken seriously and they can be influenced by it." (Nurse 6).
>
> "It should be a training program that is done seriously and families can be affected by it. It seems that the training we are giving seems to have no effect.... Now I do not know, maybe it is because of the little time we spend on them that I tell you, or because of the problems, or because it is too busy, I do not know, maybe for example the training should be from Be someone else who has an impact" (Nurse 8).

Category 3: Continuing Care

The development of a child with cancer is stressful. Family involvement in the disease and treatment process leads to the reduction of resistance to problems. This can imbalances in life and the need for care throughout diagnosing process of a child with cancer until the post-bereavement stages. This category includes "formal and informal family care" and "individualized care."

Formal and Informal Family Care

Participants believed that it was necessary to provide formal care by health care professionals, including physicians and nurses, as well as informal care by family caregivers in the face of the bereavement process.

> "It is true that we do things for the family in the hospital which may calm them down a bit. On the other hand, the main stage of their adaptation to this situation is when they go home and need professional people to be with them." (Nurse 12).
>
> "Many times, moms say that they call Famila to ask how we are, but now that they do not ask, they all get sore. Hey, they say, look what you have done, God has made you like this.... Or they say, every time they call, they say, 'How are you?' The disease is the same... I mean, in fact, there are a series of disorders that I feel are bothering the family and there is no informal support for them, so official health caregivers should provide support at home and in the personal life of the bereaved family" (Nurse3).

Individualized Care

Preparing for death, considering the specific needs of each child and family, and post-bereavement care are integral parts of care.

> "Families are very different from each other, one is rich, one is religious, and in short, they are different, and you do not know how to behave... For example, 1 day, I told a mother that there are charities that can help the families suddenly, the mother got angry and said, "Are we begging?" (Nurse 9).
>
> "The wishes of the bereaved families may be similar in general, but they may differ from person to person... There must be an expert who knows what to say through which door to enter and how to guide them, because they really say something that only A team of experts can understand their needs from what they say" (Nurse 12).

DISCUSSION

In this study, the bereavement needs of families who have a child with cancer were explained from the perspective of health caregiver. The needs of bereaved families include the three categories of "achieving peace," "Abandoned family access to care," and "continuing care."

Regarding the category of achieving peace, based on the experience of the participants, during the bereavement period, the internal management of the house suffers, and the family becomes paralyzed. Following the pressures of illness and treatment, the strength of the family weakens, and the process of life becomes difficult. So that in the face of this crisis, they are constantly asking for help and support to maintain and strengthen their cohesion. The reflection of needing help is the need for spiritual and existential support, companionship with the family, contact with other bereaved families, support in passing and accepting the bereaved and continuing empathetic communication with the family. Many participants believed in calming the patient and family in the end of life and considered proper communication, as well as the cessation of painful aggression. Spiritual care supports a sense of hope, value, the purpose of connecting with others, and connecting to greater power in the child and family. Some participants believed that the remembrance of God and religious practices heal to reduce the child's pain and illness, but the possibility and support are not provided in this regard. It seems that spiritual care for parents during a child's illness is necessary because they look to help in the body of religion. Nikseresht et al. (2016) also recommended the use of spiritual and religious approaches as an efficient way to deal with the stresses caused by cancer in Iranian society. Rassouli et al. also considered spiritual needs as one of the most important needs of Iranian cancer patients and their families (Rassouli and Sajjadi, 2014). In line with this, the results of a qualitative study showed that religion has known as the main theme of the supportive factor for pediatric cancer adaptation (Stanton et al., 2015).

Regarding companionship with the family, it can be said that almost all participants in the study emphasized the effect of health caregivers' behavior on increasing or decreasing the morale of the child and family. Esbensen et al. (2008) suggested that professional staff spend more time listening to patients and paying attention to their interests. But Support from professional staff has been shown to be necessary only when the family network is dysfunctional, with poor communication (Näppä et al., 2016). A review study by Laura et al. showed that one of the basic needs of families was caregiver communication skills with the family. In consist with the results of the present study, effective communication and method of telling the truth can have a positive outlook for the family (Kerr et al., 2004).

In the subcategory of contact with other bereaved families, participants believed that during the bereavement period, contact and communication with families with similar conditions are effective and comforting. However, during this period, some communications are strengthened, and some are reduced. In addition, aspects of family communication such as sharing feelings, appreciating each other, listening, and expression skills are important, positive, and supportive. The bereavement groups seemed to produce positive effects which could not be captured by the chosen outcome measures, such as a deeper insight into the grieving process and a feeling of joint experience in grief. It cannot be told how anxiety, depressive mood and grief would have evolved in participants without the bereavement intervention. The results of Nappa study indicate that we as health care staff do not have to worry about most of those persons who decline participation in bereavement groups, as they show less severe grief and anxiety than others (Näppä et al., 2016).

Support in passing and accepting the bereaved was another subcategory of achieving peace. The dissolution of the attachment relationship with the child causes severe anxiety and other negative emotions in parents. Parents may feel guilty about not being able to protect their children. Research emphasizes that bereavement is an injury that causes negative psychological and health effects (Rogers et al., 2008). Stroebe's study found that bereaved were more likely to commit suicide (Stroebe et al., 2005). Li et al. (2005) that the bereaved were at risk of psychiatric hospitalization, especially the mother. The risk of a mother's hospitalization may continue for more than 5 years after the child's death (Candy et al., 2015). However, Lindemann stated in the 1940s that if normal grief is shared with professional help, it is possible to settle an uncomplicated grief reaction in 4–6 weeks (Näppä et al., 2016). Danish researchers have found that the death rate of bereaved parents is higher than that of non-bereaved parents. Thus, bereavement harms social functioning and quality of family life (Li et al., 2003). Families bereaved by cancer also often struggle with isolation due to fear of burdening their support network with their persistent pain (Hinds et al., 1997) and experience decreased social support over time (Lichtenthal et al., 2015).

Regarding continuing empathetic communication with the family, it can be said that according to the participants, after the child's death, the families are practically abandoned and, no care is provided. Some health caregivers make limited phone calls to bereaved families without written instructions. In a study, the satisfaction and uniqueness of the role of volunteers in care, from the perspective of the patient and family have been mentioned (Candy et al., 2015). According to the cultural and religious context of the country and the existence of numerous charities, it can be adapted to the prevailing condition (Rassouli and Sajjadi, 2014). While there is no right or wrong way to child loss bereavement, the Australian Care Program helps families know which response is right or wrong and helps families to identify when family members need counseling and support (Palliative Care Australia, 2010). In Canada, research has shown that parents are constantly in need of bereavement services as part of their care. There is a wide variety of bereavement programs where services can include a range of telephone or letter calling programs, grief support groups (siblings or parents), or meetings with professionals to discuss the death of a child or autopsy results (Widger et al., 2012).

The second category was abandoned family access to care. Regarding the sub-category promotion of family self-control and awareness of end-of-life care to the family, participants acknowledged that despite providing care, factors such as child and family wandering in different areas of care, insufficient knowledge of medical resources, and spending too much time accessing services. The family faces problems that require coordination between different levels of treatment to receive ongoing care. Consistent with the present study, a study aimed at assessing the information and health needs of immigrants with cancer showed that the most information needs of patients were access to information about healthier living and disease control and management, including information needs related to nutrition, physical activity and pharmaceutics information (Riahi et al., 2016). Parents like to be involved in treatment decisions and know the risks and side effects of treatment. These cases are consistent with the results of the present study. It can be said that having information about the child's condition is essential for parents' peace of mind, feeling of being in control of the situation, staying optimistic and developing strategies that are in the best interest of the child (Kerr et al., 2004). In line with the findings of the present study, in a needs assessment in parents of children with cancer, information needs were recognized as a common need (Kerr et al., 2004). Moreover, a study showed that parents highly rated involvement in decision making regarding care and treatment of their child, although decision making during the palliative phase is acknowledged as being extremely difficult for parents (Hinds et al., 1997). In addition, although parents perceive their decisions around their child's care and treatment needs as representative of their child's needs (Van der Geest et al., 2014). There is evidence that parents' understanding about their child's prognosis may not always be realistic (Wolfe et al., 2000). Therefore, involvement and support of health care professionals remain crucial (Van der Geest et al., 2014).

The last category of this study was called continuing care. Formal and informal family care should be based on an understanding of the position of the family and the child in the family network, friends and community, interactions, and communication. Findings showed that having good relationships and sources of support from family and those around are factors in reducing the burden of disease on the child and family, and the absence of these positive factors harms the child and family. Social care is defined as the provision of people whom a person trusts and feels respected. This type of support in stressful situations such as cancer diagnosis is considered an important source of psychological support (Candy et al., 2015). Despite families' risk for poor outcomes, few bereavement follow-up programs and formal interventions to support them have been rigorously evaluated. A systematic review of studies on hospital-based bereavement programs concluded that such programs help families feel cared for, reduce their sense of isolation, and improve their coping (Donovan et al., 2015). The results of a study showed parents were positive about the collaboration between the tertiary care, local hospital, and community health care professionals and highly rated the frequency of consultations with health care professionals from the hospital in the period before their child died, in the time period between their child's death and funeral, and in the period thereafter (Van der Geest et al., 2014).

Regarding continuity of care, earlier reported benefits of continuity of care, for instance, reducing parental frustration and enhancing parents' confidence in quality of care could clarify why continuity of care during the palliative phase is related to lower levels of parents' long-term grief. In contrast with communication and continuity of care, Van der Geets study showed that the extent to which parents felt involved by health care professionals turned out not to be related with parental grief levels (Van der Geest et al., 2014).

Children and adolescents are in the midst of the process of physical, emotional, cognitive, and spiritual development and have different needs depending on their stage of development, and their care should be individualized. Their families also communicate in different ways, and their understanding of illness and death depends on their child's stage of development and experience. Therefore, many participants considered that family care based on their child's development is necessary. However, research findings indicate that the child's anxiety about hospitalization is closely related to parents' mental states. Parents' tendency to behave abnormally with a sick child, giving the child a special position, and being vulnerable has negative consequences and leads to the child's secondary mental disability.

CONCLUSION

Dedication to providing excellent care during treatment and into bereavement for seriously ill children and their family members is crucial for holistic, patient-, and family entered care. Parental grief is complex and support for bereaved parents is universally needed, albeit with individual differences (Spraker-Perlman et al., 2021). According to the challenges and needs of the family before and after the death of the child, the care team needs to pay special attention to the families of the bereaved patients. Therefore, it is recommended that members of the health care team be trained in assessing family needs, identifying risks from adverse outcomes, continuing care, and providing resources during bereavement. The needs of the bereaved family should also be addressed in their care program. The results of the present study can provide broader bereavement care, but more studies are needed to clarify other experiences and families' perceptions of the needs of the bereavement period. Limitations of this study included the impossibility of generalizing the results and the lack of cooperation of bereaved families to participate in the study.

DATA AVAILABILITY STATEMENT

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

AUTHOR CONTRIBUTIONS

MP: investigation, wrote the manuscript, and writing—review and editing. MR: conceptualization methodology and writing—review and editing. NR, SR, and SB: writing—review and editing. SM: project administration, conceptualization, methodology, and supervision. All authors contributed to the article and approved the submitted version.

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SYSTEMATIC REVIEW
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Levels and Determinants of Place-Of-Death Congruence in Palliative Patients: A Systematic Review

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Introduction: Congruence, understood as the agreement between the patient's preferred place of death and their actual place of death, is emerging as one of the main variables indicating the quality of end-of-life care. The aim of this research was to conduct a systematic literature review on levels and determinants of congruence in palliative patients over the period 2010–2021.

Method: A systematic review of the literature in the databases of PubMed, Scopus, Web of Science, PsycINFO, CINAHL, Cuiden, the Cochrane Library, CSIC Indexes, and IBECS. Information was extracted on research characteristics, congruence, and associated factors.

Results: A total of 30 studies were identified, mainly of retrospective observational design. The congruence values varied substantially between the various studies, ranging from 21 to 100%. The main predictors of congruence include illness-related factors (functional status, treatments and diagnosis), individual factors (age, gender, marital status, and end of life preferences), and environmental factors (place of residence, availability of health, and palliative care services).

Conclusion: This review, in comparison with previous studies, shows that treatment-related factors such as physical pain control, marital status, having a non-working relative, age, discussing preferred place of death with a healthcare professional, and caregiver's preference have been associated with higher levels of congruence. Depending on the study, other factors have been associated with either higher or lower congruence, such as the patient's diagnosis, gender, or place of residence. This information is useful for designing interventions aimed towards greater congruence at the end of life.

Keywords: palliative care, congruence, place of death, systematic review, patient preference, hospital, end of life

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INTRODUCTION

Place of death is a key consideration in end-of-life care. Dying in the desired or preferred place is an important consideration for patients, families and caregivers, and is therefore considered a quality indicator of palliative care (Ali et al., 2019). Historically, palliative care professionals have tried to ensure that people are cared for at home until the end of life, with dying at home being seen as an indicator of high-quality palliative care (Stajduhar and Davies, 2005). However, home is not the sole optimal option for providing quality end of life care, as other alternatives may be preferable such as hospitals, nursing homes, and hospices (Jordhøy et al., 2000; Sleeman et al., 2014). Interpreting the proportion of deaths at home as the exclusive indicator of high-quality palliative care implies that not dying at home is a failure of care and ignores the possibility that the patient may wish to die in a different place (Sekiguchi et al., 2014). For this very reason, congruence between the patient's preferred and actual place of death is increasingly being considered as a quality indicator of end of life care. As such, "congruence" is understood as the agreement between a patient's preferred place of death and actual place of death (Tang and Mccorkle, 2003). However, despite growing interest in this concept, research on the key determining factors remains scarce (Billingham and Billingham, 2013). To date, most published studies have focused on the relationship between the characteristics of the person with advanced illness at the end of life and the final place of death (Brazil et al., 2005), with widely varying results on congruence between the preferred and actual place of death (Bell et al., 2010). A clear understanding of congruence levels between the chosen and final place of death, as well as the associated variables, is critical to shaping the development of palliative care and improving end of life care (Rainsford et al., 2018; Wiggins et al., 2019).

In this context, predictors of place of death and congruence have traditionally been grouped into three categories: illness-related factors, individual factors, and environmental factors. Among the illness-related factors, the latest review and meta-analysis (Billingham and Billingham, 2013) identified cancer as having the highest congruence in final place of death (compared to other non-cancer pathologies). In terms of individual factors, ethnicity and the patient's previous preference have thus far been the variables included in earlier reviews (Bell et al., 2010). With regard to environmental factors, Bell et al. (2010) highlighted support from medical staff and being in a hospice, as well as family support.

Although the review by Bell et al. (2010) significantly advanced understanding of the factors influencing congruence, it is important to update the findings in light of subsequent research (Gomes et al., 2013; De Roo et al., 2014; De Boer et al., 2017), in which congruence between preferred and final place of death is identified as a key factor in the quality of end-of-life care. The General Medical Council's (2010) end of life best practise guidelines included the need to plan end of life care together with patients, so that members of the multidisciplinary teams caring for them can understand and address their wishes and needs improving the care to patients and families.

In light of the above, the present study aimed to conduct a systematic literature review for the years 2010–2021, following on from Bell et al. (2010) and analysing congruence levels between the preferred and actual place of death reported in the scientific literature, and the factors identified as determinants of congruence between preferred and final place of death among palliative patients.

MATERIALS AND METHODS

Search Strategy

A systematic search was conducted to locate articles examining the congruence of place of death in patients at the end of life and predictors thereof between July 2020 and February 2021.

The keywords used to carry out the search were grouped into three main categories: terms related to "palliative care," terms related to "patient preference" and terms synonymous with "place of death." Searches with terms located in the title/summary and/or descriptors of the same category were joined with the Boolean connector "OR." Once the three search categories were prepared, they were combined with the Boolean operator "AND." Lastly, the time period (2010–2021) and language limits were applied. The final search strategy is included as **Supplementary Material**. The search was carried out using the following national and international databases: PubMed, Scopus, Web of Science, PsycINFO, CINAHL, Cuiden, the Cochrane Library, Information and Documentation of Science in Spain (ÍnDICEs-CSIC) and Spanish Bibliographical Index on Health Science (IBECS). This strategy was complemented by a manual search of international journals: BMJ Support Palliative, Palliative Support Care, Palliative Medicine, the Journal of Palliative Medicine and the Journal of Palliative Ageing, which address the topic of interest. The bibliographical references of the articles included in this study were also examined with a view to including potentially original studies.

Inclusion and Exclusion Criteria

Inclusion criteria were as follows: (1) original studies analysing place of death congruence in patients at the end of life and/or its predictors, (2) studies conducted in populations over 18 years of age with both oncological and non-oncological pathologies, and (3) studies published in English, Spanish or Portuguese from 2010 onwards. Qualitative studies were excluded, as well as those that did not provide at least quantitative data on congruence in the final place of death.

Selection of Studies

Firstly, the records obtained from the electronic searches were assessed by two independent researchers (NOB and SGS) for eligibility, based on a review of the titles and abstracts. Those studies selected in this first phase were reviewed in full text to verify that they met the established inclusion and exclusion criteria. When consensus could not be reached between the two researchers (in six of the studies) a third researcher (VCC) was consulted. At this stage the three researchers arrived at a final consensus, which is shown in the results section.

TABLE 1 | Study characteristics.

Study	Design	Data collection (Months)	Sample size	Type of participants	Gender (% Women)	Age in years (SD)	Pathologies (%)	Quality STROBE/ CONSORT
1. Sheridan et al. (2021)	ROS	12	963	P	43	75.8 (NI)	Cancer: 100	20
2. Cai et al. (2020)	POS	26	290	P, CG	P: 53.8 CG: 67.2	P: 72.4 (12.38) CG: 59.3 (13.10)	Cancer: 100	21
3. Skorstengaard et al. (2020)	RCT	36	N: 205 n (intervention) 102 n (control): 103	P	50	69 (NI)	Cancer: 50.1 Heart disease: 16.7 Lung disease: 33.2	21
4. Ali et al. (2019)	ROS	60	2176	P	NI	NI	Cancer: 88 Non-cancer: 22	18
5. Blanchard et al. (2019)	ROS	22	191	P	55.5	57.6 (13.26)	Cancer: 100	19
6. Wiggins et al. (2019)	ROS	16	1047	P	64.6	<79 (DT 13.8)	Cancer: 10.0 Cardiac disease: 4.4 Vascular disease: 2.2 Respiratory disease: 1.7 Neurological disease: 1.6 Dementia: 75.8 Other: 4.3	19
7. Bannon et al. (2018)	ROS	6	467 (363 included in the analysis)	P	60.6	<69 (57.5%)	Cancer: 100	18
8. Chiba et al. (2018)	Mixed	12	18 P/CG 24 GP	P, CG, GP	P: 11.2 CG: 94.4 GP: 16.7	P: 71.9 (12.4) CG: 61.9 (12.9) GP: NI	Cancer: 100	18
9. Raijmakers et al. (2018)	ROS	17	797	P	P: 53.6 CG: 69.3	P <65 57.6% CG <75 27.2%	Cancer: 43.7 Stroke: 12.4 COPD: 10.9 Heart failure: 14.6 Dementia: 26.5	19
10. Higginson et al. (2017)	POS	17	138	P	49	74 (NI)	Cancer: 88.0 Non-cancer: 12.0	18
11. Lin et al. (2017)	ROS	55	481	P	39.3	70.6 (14.3)	Cancer: 70.9 Non-cancer: 29.1	21
12. Howell et al. (2017)	ROS	36	323	P	44.9	72.4 (12.7)	Cancer (haematological): 100	21
13. de Graaf et al. (2016)	ROS	6	130	P	52	72 (12.1)	Cancer: 89 Lung failure, COPD: 3 Renal failure: 1 ALS: 2 Heart failure: 1 Dementia: 1 Other: 2	21
14. Arnold et al. (2015)	ROS	12	1127	P	50	70 (13)	Cancer: 94 Other: 6	15
15. Burge et al. (2015)	ROS	24	1316 (605 included in the analysis)	P	51.3	79.1 (12.8)	Cancer: 38.1 Others: 61.9	21
16. Gage et al. (2015)	ROS	18	688	P	43.6	75.10 (NI)	NI	18
17. Ko et al. (2014)	ROS	36	695	P	43,3	≥ 65: 67.9%	Cancer: 100	19
18. Hunt et al. (2014)	ROS	7	1422	P, CG	P: 34.6 CG: 64.9	P: ≥ 60: 91.8% CG: ≥ 60: 55.5%	Cancer: 34.6 Cardiovascular disease: 24.9 Other: 40.5	20

(Continued)

TABLE 1 | Continued

Study	Design	Data collection (Months)	Sample size	Type of participants	Gender (% Women)	Age in years (SD)	Pathologies (%)	Quality STROBE/ CONSORT
19. Aoun and Skett (2013)	POS	18	43 (36 included in the analysis)	P	49	74 (10.1)	Cancer: 100	20
20. Brogaard et al. (2013)	POS	21	96	P	41.7	69.9 (NI)	Cancer: 100	20
21. Fischer et al. (2013)	POS	29	458	P	35	57.9 (14.8)	Cancer: 11 Other: 89	18
22. Janssen et al. (2013)	POS	24	265 (206 included in the analysis)	P	35.9	67.2 (13.1)	COPD: 41.8 Chronic heart failure: 29.6 Chronic renal failure: 28.6	21
23. Capel et al. (2012)	POS	24	788	P	NI	NI	Cancer: 93 Non-cancer: 7	21
24. Johnson et al. (2012)	POS	15	126 (80 included in the analysis)	P	38	78 (10.7)	Chronic heart failure: 100	21
25. Abarshi et al. (2011)	ROS	12	252 (165 included in the analysis)	P	55	> 65: 80%	Cancer: 38 Non-cancer: 62	20
26. Alonso-Babarro et al. (2011)	POS	36	380 (228 included in the analysis)	P, CG	P: 39.5 CG: 17.9	P: 66.76 (13.4) CG: 54,32 (14.4)	Cancer: 100	20
27. Escobar Pinzon et al. (2011)	CS	4	1378	P, CG	P:55.6 CG: 63.4	P: 77.6 (13.2) CG: 58.8 (12.8)	Cancer: 24.2 Dementia: 8.9 Cardiovascular disease: 8.4 Other: 12.0 Multimorbidity: 36.6 Missing/I don't know: 9.9	21
28. Gerrard et al. (2011)	ROS	2007: 6 2009: 6	n (2007): 236 n (2009): 275 Total: 511	P	2007: 50.5 2009: 42.0	2007: 78 (NI) 2009: 72 (NI)	2007: Cancer: 66 Other: 34 2009: Cancer: 76 Other: 24	20
29. Walker et al. (2011)	ROS	24	150	P	NI	NI	NI	19
30. Holdsworth and Fisher (2010)	ROS	6	298	P	NI	NI	Cancer: 80 Other: 20	18

RCT, randomised controlled trial; ROS, retrospective observational study; POS, prospective observational study; CS, cross-sectional study; COPD, chronic obstructive pulmonary disease; ALS, amyotrophic lateral sclerosis; NI, no information; RRS, rapid response service; P, patient; CG, caregivers; GP, general practitioners.

Data Extraction

In order to facilitate data extraction, three tables were created in which the main results were synthesised. **Table 1** includes information on the characteristics of the selected studies (author/s, year, research design, data collection time, sample size, sample characteristics, age, pathologies, and methodological quality assessed). **Table 2** includes the main results concerned with preferred place of death and actual place of death, as well as congruence and the appearance of associated factors. **Table 3** shows the main factors associated with place of death congruence, according to whether they were illness-related, individual or environmental.

Methodological Quality

The methodological quality of the selected studies was examined using the CONSORT and STROBE checklists based on study design (see **Table 1**). The ratings of each article included in the systematic review are available as **Supplementary Material**.

RESULTS

Search Results

One thousand four hundred forty-eight articles were initially retrieved from the nine databases and the additional manual search. After eliminating duplicates, 1,062 articles were retained, and then following the initial screening process, by means of title and abstract review, 481 articles were selected and subsequently reviewed in full text. Finally, a total of 30 articles meeting the inclusion and exclusion criteria were selected and included. In this final phase, 451 articles were discarded, either because they were not original studies or because they did not record information on congruence between preferred and final place of death (see **Figure 1**).

Characteristics of the Studies Included

The main characteristics of the studies in the systematic review are described in **Table 1** and include: the design, the number

TABLE 2 | Results on preferred place, final place of death, congruence, and the existence of associated factors.

Study	**Preferred place of death (%)**				**Actual place of death (%)**				**Congruence**	**Associated factors**	**Percentage calculation**
	Hospital	**House**	**Long-term care**	**Hospice/ PCU**	**Hospital**	**House**	**Long-term care**	**Hospice/ PCU**			
1. Sheridan et al. (2021)	17.7	40.6	14.1	18.1	58.0	20.0	11.0	10.2	66.1%	NI	PREF
2. Cai et al. (2020)	16.9	65.5	1.4	16.2	29.3	48.6	1.4	20.7	71.7% K: 0.527	1-2-3	TOT
3. Skorstengaard et al. (2020)	GI: 1.0	GI: 33.5	GI: 3.9	GI: 2.9	GI: 17.3	GI: 42.3	GI: 0.0	GI: 34.6	GI: 52.4%	NI	TOT
	GC: 1.0	GC: 34	GC: 2.9	GC: 4.9	GC: 27.1	GC: 17.0	GC: 8.5	GC: 32.2	GC: 34.6%		
4. Ali et al. (2019)	1.2	44.8	5.5	12.0	21.2	46.5	8.4	23.8	69%	NI	PREF
5. Blanchard et al. (2019)	–	54.5	–	–	–	40.3	–	–	47.6% K: 0.016 IC 95%: (−0.107)–0.139	1-2	TOT
6. Wiggins et al. (2019)	0.4	31.1	49.8	1.4	12.1	26.5	50.9	3.2	83.7%	1-2	TOT
7. Bannon et al. (2018)	3.5	74.7	1.7	6.0	43.7	38.1	9.0	9.2	Home: 53.4%	1-2-3	TOT
8. Chiba et al. (2018)	11.1	61.1	–	–	11.1	88.9	–	–	100%	NI	TOT
9. Raijmakers et al. (2018)	1.8	65.7	24.3	8.3	–	–	–	–	69.0%	1-2-3	TOT
10. Higginson et al. (2017)	4.0	56.0	2.0	22.0	34.0	21.0	6.0	39.0	23.0%	NI	TOT
11. Lin et al. (2017)	–	49.1	–	49.9	–	42.8	–	57.2	92.3%	2-3	TOT
12. Howell et al. (2017)	28.2	45.8	5.6	16.9	74.3	15.2	5.0	5.6	63.4%	NI	TOT
13. de Graaf et al. (2016)	–	72.0	–	21.0	6.0	70.0	–	24.0	86%	NI	TOT
14. Arnold et al. (2015)	0.7	37.0	2.4	60.0	–	–	–	–	85%	NI	PREF
15. Burge et al. (2015)*	15.9	73.9	10.3	–	51.5	30.4	17.9	–	51.9% K: 0.29	NI	PREF
16. Gage et al. (2015)	*0.0	*76.9	*0.8	*21.1	*7.7	*63.2	*4.5	*24.7	*69.2%	2-3	TOT
	**0.92	**51.5	**10.7	**35.8	**12.7	**26.3	**15.0	**46.1	**59.2%		
17. Ko et al. (2014)	–	100	–	–	16.7	76.0	1.3	6.0	76.0%	2-3	TOT
18. Hunt et al. (2014)	5.1	73.9	6.5	10.6	49.4	13.4	24.6	10.5	49.3% K: 0.034	NI	TOT
19. Aoun and Skett (2013)	8.0	56.0	–	25.0	22.0	14.0	–	56.0	41.2%	NI	PREF
20. Brogaard et al. (2013)	3.0	45.0	1.0	16.0	26.0	41.0	6.0	26.0	44.0%	NI	TOT
21. Fischer et al. (2013)	10.0	75.0	6.0	4.0	35.0	31.0	20.0	12.0	37.0%	2	TOT
22. Janssen et al. (2013)	33.3	51.5	–	–	57.5	27.3	–	–	39.4% K: 0.07	NI	TOT
23. Capel et al. (2012)	1.7	48.2	5.2	14.4	30.0	36.0	6.3	27.1	Home: 69% Hospital: 85.7% Long-term: 82.9% Hospice: 81.6%	NI	TOT
24. Johnson et al. (2012)	*n* = 4	*n* = 69	–	*N* = 12	*N* = 41	*N* = 35	–	*N* = 21	61%	NI	PREF
25. Abarshi et al. (2011)	–	–	–	–	28.6	43.7	21.4	6.3	No deaths identified in recent days: 21% Deaths identified in recent days 79%	NI	PREF
26. Alonso-Babarro et al. (2011)	–	80	–	–	–	72.4	–	–	89%	NI	PREF
27. Escobar Pinzon et al. (2011)	0.4	50.5	1.3	1.5	39.3	38.2	13.4	7.5	58.9% K: 0.14	2-3	PREF
28. Gerrard et al. (2011)	+9.0	+44.0	+11.0	+36.0	–	–	–	–	76.0%	NI	PREF
	++31.0	++24.0	++7.0	++38.0							
29. Walker et al. (2011)	–	78.6	–	21.4	27.0	35.0	11.0	27.0	85.7%	NI	PREF
30. Holdsworth and Fisher (2010)	0.7	26.8	1.3	9.7	17.8	36.6	8.4	37.2	61.7% K: 0.38	NI	TOT

*K, kappa; 95% CI, 95% confidence interval for cohen's kappa; GI, group intervention; CG, control group; NI, no information; PREF, calculated on those showing a preference; TOT, calculated on the total sample; *users rapid response service; **non-users rapid response services; +2007 data; ++2009 data; 1: illness-related factors, 2: individual factors, 3: environmental factors.*

TABLE 3 | Factors associated with congruence between preferred and final place of death.

Study	Factors associated with congruence between preferred and final place of death		
	Illness	**Individual**	**Environmental**
Cai et al. (2020)	• Patient functional status- OR: 1.02; IC 95%: 1.01–1.04	• Civil status (divorced, separated or widowed) - OR: 0.45; IC 95%: 0.36–0.56	• Intensity of home-based nursing visits+ OR: 1.02; IC 95%: 1.00–1.04 • Hours of personal support care+ OR: 1.09; IC 95%: 1.01–1.18
Blanchard et al. (2019)	• Use of morphine + OR: 1.87; IC 95%: 1.04–3.36	• Aged + OR: 1.03; IC 95%: 1.00–1.05 • Preference to die at home - OR: 0.44; IC 95%: 0.24–0.82	
Wiggins et al. (2019)	• Patient functional status (impairment) + OR: 1.82; IC 95%: 1.06–3.13 • Ceiling of treatment of symptomatic relief only+ OR< 0.2.65; IC 95%: 1.37–5.14 • Cancer diagnosis - OR: 0.52; IC 95%: 0.28–0.97)	• Cardiopulmonary resuscitation preference- OR: 0.32; IC 95%: 0.16–0.62 • Early POD register (51–250 days)- OR: 0.60; IC 95%: 0.38–0.94	
Bannon et al. (2018)	• Being unconscious during final week of life - OR: 0.1; CI 95%: 0.0–0.4	• Discussing POD with a HCP + OR: 4.7; IC 95%: 1.9–11.5 • Age (older than 80/younger than 70)- OR: 0.5; CI 95%: 0.2–1.0 • Being Presbyterian - OR: 0.30; IC 95%: 0.11–0.87	• Living in an affluent area + OR: 4.0; 95% IC 95%: 1.4–11.8 • Satisfactory care at home from a nurse + OR: 6.1; IC 95%: 2.5–15.2. • Caregiver preference of place of death (home vs. others) + OR: 17.7; IC 95%: 5.3–59.3
Raijmakers et al. (2018)	• Dementia diagnosis **+** OR: 3.33; IC 95%: 1.01–11.00 • Interaction effect: patients with dementia x preference to die at home - OR: 0.14; IC 95%; 0.04–0.56 • Stroke diagnosis - OR: 0.51; IC 95%: 0.26–0.98	• Having a partner+ OR: 2.03; IC 95%: 1.23–3.35 • PPOD Home - OR: 0.05; IC 95%: 0.02–0.12	• Have had contact with a general practitioner in the last week before death + OR: 3.85; IC 95%: 1.38–10.78 • Interaction effect with preference to die at home + OR: 6.48; IC 95%: 2.01–20.92 • High continuity of care + OR: 4.83; IC 95%: 2.36–9.89
Lin et al. (2017)		• PPOD Inpatient Hospice + OR: 17.37; IC 95%: 5.13–58.82	• Use of a High Intensity Hospice information system (HIS) + OR: 3.85; IC 95%: 1.19–12.40
Gage et al. (2015)		• Declare an initial PPOD Care Home + OR: 7.7; IC 95%: 2.5–23.4 • PPOD Own home - OR: 0.55; IC 95%; 0.3–0.8	• User of RRS+ OR: 2.1; IC 95%; 1.4–3.0 • Live with a carer + OR: 1.5; IC 95%; 1.0–2.2 • Number of days in the study - OR: 0.98; IC 95%: 0.98–0.99 • Place of residence (area 3) - OR: 0.54; IC 95%: 0.31–0.96
Ko et al. (2014)		• Aged (65-85) + OR (Belgium): OR: 0.4; IC 95%: 0.2–0.97 • Female – OR (The Netherlands): 0.1; IC 95%: 0.04–0.4 • Decision making capacity + OR (The Netherlands): 6.7; OR 95%: 1.5–29.0	• GP provision of palliative care + OR (Belgium): 9.9; IC 95%: 3.7–26.6 OR (The Netherlands): 9.7; IC 95%: 2.3–39.9 OR (Italy): 2.6; IC 95%: 1.2–2.5 • Average number of GP contacts in the 2nd, 3rd, and 4th weeks before death + OR (Italy): 0.1; IC 95%: 0.01–0.9
Fischer et al. (2013)		• Female + OR: 3.30; IC 95%: 1.25–8.72	
Escobar Pinzon et al. (2011)		• Having a non-working relative + OR: 1.79; IC 95%: 1.16–2.76 • Respondent and deceased lived together in one common household + OR: 2.28; IC 95%: 1.57–3.32	• Living in a rural municipality + OR: 1.88; IC 95%: 1.02–3.43 • Living in a rural town + OR: 2.30; IC 95%: 1.17–4.49 • Living in a small town OR: 1.95; IC 95%: 1.04–3.68)

+, positive association between the variables; -, inverse association between the variables; POD, place of death; PPOD, preferred place of death; HCP, health care professionals; GP, general practitioners; RRS, rapid response service.

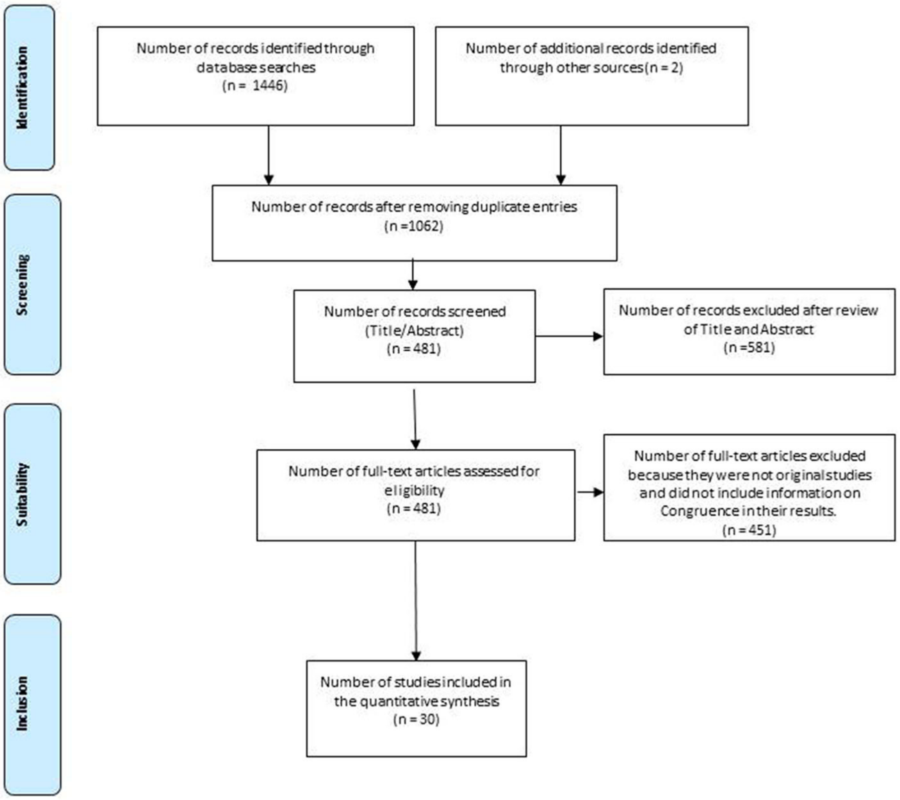

FIGURE 1 | Flow diagram of the selection of studies for the systematic review. Source: Moher et al. (2009).

of months of data collection, the total sample size (divided into groups when necessary), the type of participants (patients, caregivers, and/or general practitioners), the gender, age and main pathologies of the participants, and the results of the corresponding checklist. The research designs (see **Table 1**) were mainly retrospective observational studies ($n = 19$) and prospective observational studies ($n = 8$). Only one randomised controlled trial, one cross-sectional study and one mixed study were included. Data collection time for each study ranged from 4 (Escobar Pinzon et al., 2011) to 60 months (Ali et al., 2019). Sample sizes ranged from 18 (Chiba et al., 2018) to 2,176 individuals (Ali et al., 2019), with three studies having fewer than 100, 15 studies between 100 and 500 and 12 studies with a sample size >500 participants. Most of the studies had a sample with similar proportions of males and females. In six studies the female sample was over 60% and in four studies gender was not reported. The mean age was over 65 years in most studies ($n = 24$), and in four cases, age was not reported. A total of 10 studies exclusively analysed patients with cancer, while the rest included other chronic pathologies, or non-cancer pathologies such as dementia or stroke. In terms of the methodological quality of the studies, assessed through the different checklists, 29 of the 30 articles obtained scores greater than or equal to 18, with a range between 15 and 21.

Preferred Place, Actual Place of Death and Congruence

Table 2 shows the percentage of preferred place of death and actual place of death in each study. In addition, the congruence value (including the kappa value where reported), the method for calculating the congruence value (using either the total sample size or only those who indicated a preference), and the analysis of associated factors is included. Preferred place of death (see **Table 2**) is considered first of all. A preference for hospital was reported in 23 of the 30 studies, ranging from 0% in one of the groups in the Gage et al. (2015) study to 33.3% (Janssen et al., 2013). Most of the studies (29 out of 30) reported on preference for dying at home, ranging from 24% for one of the groups reported by Gerrard et al. (2011) to 78.6% (Walker et al., 2011). Preference for dying in medium to long stay facilities was reported in 19 of the 30 studies, ranging from 0.8% (Gage et al., 2015) to 49.8% (Wiggins et al., 2019). Finally, preference for dying in a hospice or palliative care unit was reported in 23 of the studies, ranging from 1.4% (Wiggins et al., 2019) to 60% (Arnold et al., 2015).

Of the 30 studies, 27 reported some data on actual place of death, with only three providing no information at all (Gerrard et al., 2011; Arnold et al., 2015; Raijmakers et al., 2018). The percentage of people who died in hospital ranged from 6% (de Graaf et al., 2016) to 74.3% (Howell et al., 2017), and was reported in 24 of the studies. Death at home was reported in all 27 studies, ranging from 13.4% (Hunt et al., 2014) to 88.9% (Chiba et al., 2018). The percentage of deaths in medium to long stay facilities was reported in 19 of the studies, ranging from 0% (Skorstengaard et al., 2020) to 50.9% (Wiggins et al., 2019). Finally, death in a hospice or palliative care unit was reported in 22 studies, ranging from 3.2% (Wiggins et al., 2019) to 57.2% (Lin et al., 2017).

All studies reported on the congruence index. However, some studies included an overall congruence percentage while others calculated specific congruence percentages according to place of death or other variables. In 19 of the 30 studies, the congruence percentage was calculated on the total sample, while in other cases it was calculated only on those who expressed a preference, or who were part of a sub-sample (see final column in **Table 2**). In the 19 studies that reported overall congruence percentages, these ranged from 21% (Abarshi et al., 2011) to 100% (Chiba et al., 2018). In addition, seven studies (Holdsworth and Fisher, 2010; Escobar Pinzon et al., 2011; Janssen et al., 2013; Hunt et al., 2014; Burge et al., 2015; Blanchard et al., 2019; Cai et al., 2020) provided Kappa agreement indices, ranging from 0.016 (Blanchard et al., 2019) to 0.527 (Cai et al., 2020).

Factors Associated With Congruence in Place of Death

Finally, the factors associated with congruence are outlined in **Table 3**. These factors were organised into three main categories: illness-related, individual, and environmental factors. For each of these categories, the odds ratio and their association with congruence (positive or negative) are included. Of the 30 studies included in the review, 10 reported factors that were predictors of congruence in place of death (see **Table 2**). Five identified illness-related factors as being risk variables (Bannon et al., 2018; Raijmakers et al., 2018; Blanchard et al., 2019; Wiggins et al., 2019; Cai et al., 2020), 10 identified individual factors (Escobar Pinzon et al., 2011; Fischer et al., 2013; Ko et al., 2014; Gage et al., 2015; Lin et al., 2017; Bannon et al., 2018; Raijmakers et al., 2018; Blanchard et al., 2019; Wiggins et al., 2019; Cai et al., 2020), while seven identified environmental factors (Escobar Pinzon et al., 2011; Ko et al., 2014; Gage et al., 2015; Lin et al., 2017; Bannon et al., 2018; Raijmakers et al., 2018; Cai et al., 2020).

Positive and negative (lack of congruence) associations were reported for the factors associated with congruence (see **Table 3**). In the first instance, a number of illness-related factors were identified. The patient's functional status had both a positive and negative association with congruence (Wiggins et al., 2019; Cai et al., 2020). Cancer diagnosis (Wiggins et al., 2019) and stroke (Raijmakers et al., 2018) were negatively associated with congruence, while dementia (Raijmakers et al., 2018) was positively associated. Treatment-related factors, such as morphine use (Blanchard et al., 2019) and ceiling of treatment of symptomatic relief (Wiggins et al., 2019), were positively associated with congruence. Finally, the patient's level of consciousness during the final few days was negatively associated with congruence (Bannon et al., 2018).

Secondly, with respect to individual factors, marital status (being in a relationship) was positively associated with congruence in three studies (Escobar Pinzon et al., 2011; Raijmakers et al., 2018; Cai et al., 2020), as were having a non-working relative (Escobar Pinzon et al., 2011) and age (Ko et al., 2014; Bannon et al., 2018; Blanchard et al., 2019). Significant variability was found in relation to preference for place of death. Preference for dying at home (Gage et al., 2015; Raijmakers et al., 2018; Blanchard et al., 2019), preference for cardiopulmonary resuscitation, and early registration of place of death (Wiggins et al., 2019) were negatively associated with congruence. Preference for dying in a hospice (Lin et al., 2017), having discussed preferred place of death with a healthcare professional (Bannon et al., 2018), and having decision-making capacity (Ko et al., 2014) were positively associated with congruence. Other variables had no clear association, for instance being female, which was both positively (Fischer et al., 2013) and negatively (Ko et al., 2014) associated with congruence in two of the reviewed studies. Finally, one study looked at religious belief and found that being Presbyterian (as opposed to other religions) was negatively associated with congruence (Bannon et al., 2018).

Finally, environmental factors such as place of residence (Escobar Pinzon et al., 2011; Gage et al., 2015; Bannon et al., 2018) and indicators of adequate care by nurses, medical staff, or a caregiver were associated with higher congruence (Ko et al., 2014; Gage et al., 2015; Bannon et al., 2018; Raijmakers et al., 2018; Cai et al., 2020). Other variables associated with higher levels of congruence include the family's role, the caregiver's own preference for place of death (Bannon et al., 2018), being in a high intensity hospice (Lin et al., 2017), using a Rapid Response System (RRS) (Gage et al., 2015), and access to palliative care (Ko et al., 2014). Only the amount of time spent in the study (assessed in number of days) and one of the places of residence in the Gage et al. (2015) study had a negative association with congruence.

DISCUSSION

The present study conducted a systematic review of congruence values between preferred and actual place of death, as well as the main determinants of this congruence in the period of 2010–2021. The results of the present review analysing a total of 30 studies with over 14,000 participants indicate considerable variability in the congruence values identified, in line with results identified in previous reviews (Bell et al., 2010; Billingham and Billingham, 2013).

One of the most important objectives of palliative care is to enhance the quality of life of terminally ill patients and their environment (World Health Organization, 2021). It is therefore necessary to encourage health services to involve patients and their families in the decision-making process about their treatment and end of life care, and it is vitally important to know the patient's preferred place of death and to make it easier for them to die there (Baik et al., 2019). The results of the present review show that, although home remains one of the preferred places of death, previous studies indicate that hospitals are often one of the main places where death occurs (Nilsson et al., 2017).

On this note, a recent systematic review and meta-analysis in cancer patients shows a high degree of variability in the preferred place of death where, while home remained the highest with 55% preference, other participants preferred hospital (17%) and hospice (10%) (Fereidouni et al., 2021). In the data obtained in the present review, home appears as the preferred place of death in all the studies that evaluate this, although the values for the hospital vary greatly between studies, exceeding 30% in several (Gerrard et al., 2011; Janssen et al., 2013).

With regard to congruence, in 21 of the 30 studies the percentage was below 75% for the different locations assessed, which seems to indicate that the level is still insufficient, in line with previous studies (Billingham and Billingham, 2013; Howell et al., 2017; Baik et al., 2019). It is therefore important to consider which factors and variables influence congruence levels, as has been identified in numerous studies in the case of place of death (Cabañero-Martínez et al., 2020).

The factors predicting the congruence rate in this review include those related to the illness itself, and individual and environmental factors. Since Bell et al.'s (2010) systematic review, several new factors influencing congruence have been identified (see **Table 4**). Treatment-related factors associated with physical pain control, marital status, having a non-working relative, age, discussing preferred place of death with a healthcare professional, and caregiver's preference have been associated with higher levels of congruence. Depending on the study, other factors have been associated with higher or lower congruence such as the patient's diagnosis, gender, or place of residence.

In terms of disease-related factors, the use of morphine and other pain-related treatments were positively associated with congruence between the preferred and actual place of death. As such, both variables may refer to the existence of a palliative intervention, which in many cases will be associated with the patient's end of life preferences having been explored, discussed, understood and taken into account (Saugo et al., 2008; Scaccabarozzi et al., 2017). However, the patient's own poorer state of health and unconsciousness at the end of life were negatively associated with congruence in place of death. This could be due to the fact that, in these cases, the doctor and family are the main responsible for the decision-making process and may not be aware of the patient's own preferences, if the issue has not been addressed beforehand, or that these preferences have been put to one side (Medina et al., 2012). In addition, specific diagnoses such as cancer or stroke were negatively associated with congruence in the studies evaluated. In the first case, one possible explanation is that cancer patients in advanced stages of the disease, despite greater knowledge of their prognosis, may still die in hospital due to the wider treatment options available (Fereidouni et al., 2021). Furthermore, this may also indicate an important shortcoming with respect to effective home care plans, since their effectiveness, a priori, should make it possible for many cancer patients to die in their preferred place (Cabañero-Martínez et al., 2020). Previous reviews have highlighted an increased risk of incongruence in non-cancer pathologies, although they also noted that there appeared to be no correlation between overall levels of congruence and the percentage of patients with cancer (Billingham and Billingham, 2013). In the case of stroke, several studies have pointed to the difficulty professionals have in identifying patients' palliative needs, as well as being able to communicate adequately regarding end of life preferences (Eriksson et al., 2016; Cowey et al., 2021).

TABLE 4 | Summary of the factors affecting congruence.

Enhancing congruence	Decreasing congruence
Illness-related	**Illness-related**
Patient's functional status	Patient's functional status
Dementia	Cancer
Treatment-related aspects	Stroke
	Patient's level of consciousness during last days
Individual	**Individual**
Marital status	Preference for dying at home
Non-working relative	Preference for cardiopulmonary resuscitation
Age	Early registering of place of death
Preference for dying in a hospice	Being female
Having spoken to a healthcare professional about the preferred place of death	Religious beliefs
Decision-making capacity	
Being female	
Environmental	**Environmental**
Place of residence	Place of residence
Indicators of adequate care	Time of participation in the study
Family role	
Caregiver's preference	
High intensity hospice	
Having palliative care	

In terms of individual factors, being married or in a relationship, as well as living with a partner, was positively associated with congruence in the preferred and actual place of death. In this regard, it should be noted that the patient's decision is usually respected by their spouse or closest relatives, particularly when there is an advance directives document (Agulles Simó, 2010; Landa and García, 2017; Bejarano Gómez et al., 2019). This document enables patients to exercise their right to plan and decide on their active and palliative treatment guidelines once they are unable to make decisions (Mira et al., 2010). Previous studies suggest that patients who had prepared advance directives received care that was strongly associated with their preferences, increasing the likelihood that these plans would be implemented (Leff et al., 2000; Silveira et al., 2010; Halpern et al., 2020). Therefore, achieving congruence between the preferred place and final place of death should be an aspect that is reflected in the advance directives of those at the end of their lives. However, it is important that healthcare professionals have knowledge, training and experience in the use of advance directives in order to integrate them into end of life decision making (Aguilar-Sánchez et al., 2018).

Other factors related to individual variables refer to end of life preferences, such as the choice not to perform cardiopulmonary resuscitation, and informing healthcare staff of the patient's preferences or decision-making capacity. In this respect, the associations found positively relate these factors to congruence between place of preference and final place of death. These results are in line with previous research where adequate communication with health workers and the patient's decision-making capacity are associated with high levels of congruence (Burge et al., 2015; Cohen et al., 2015; Finucane et al., 2019). Other factors such as gender and age have been shown to be predictors, albeit with positive values in some cases and negative values in others.

Finally, in relation to environmental factors, higher levels of congruence have been found when the family's choice coincided with that of the patient in line with previous studies (Raziee et al., 2017; Cai et al., 2021). In addition, variables directly related to the availability of health services and palliative care have also been positively associated with congruence in many of the studies included in this review. Furthermore, some studies have highlighted the importance of the neighbourhood, area or size of the city in which the patient lived (Escobar Pinzon et al., 2011; Gage et al., 2015). Recent studies have shown how variables related to socio-economic status can have an important influence on the place of death. Nolasco et al. (2020) noted that the probability of dying in hospital, compared to dying at home, is higher as the level of economic deprivation in the urban area of residence increases, both for palliative care-related illnesses and for other pathologies. Future studies should clarify the role that such variables can play in predicting congruence levels in different pathologies.

The review findings suggest that little has changed regarding the congruence percentages identified in the literature. Despite the evolution of palliative care and the importance given to patients' preferences for end of life care, a high percentage do not die where they wish to. Factors such as the provision of palliative care, the role of healthcare professionals, family dynamics, and adequate care are important for improving the level of congruence among palliative care patients. Discussing end of life preferences with both the patient and family members or caregivers may facilitate the process of dying in the preferred place. Future studies analysing the balance between patient and caregiver preferences are also needed to identify their roles in achieving congruence.

The main limitations of the studies included in this review include the use of retrospective observational designs and the lack of prospective designs with which to study congruence. In addition, basic sociodemographic information about participants (such as age, gender, and main diagnosis) is not always included, making it difficult to interpret the results. Research on non-oncological conditions and the role that the diagnosis can play in predicting congruence levels is also an important line of research to be considered (Martí-García et al., 2020). Finally, some of the factors associated with congruence have both a positive and negative association (i.e., place of residence or gender), hence further research is required in order to clarify their role.

The present study has a number of strengths and limitations. In the first instance, a high degree of heterogeneity has been identified in the congruence data whereby, while in many studies this came from the total sample, in other cases only data from the sub-samples were indicated. Secondly, there has also been a high degree of variability in the associated factors across studies, with many being assessed using a single question, or by means of continuous variables in some studies and categorical variables in others. Nonetheless, a methodological assessment of all the research selected in this review was carried out, revealing adequate values. Further studies are required to gain more in-depth knowledge about the factors influencing congruence in order to optimally plan health services and improve the quality of end of life care.

CONCLUSION

In conclusion, the present review shows variability in levels of congruence between preferred and final place of death. The main predictors of congruence include illness-related factors (functional status, treatments, and diagnosis), individual factors (age, gender, marital status, and end of life preferences), and environmental factors (place of residence and availability of health, and palliative care services).

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 authors.

AUTHOR CONTRIBUTIONS

SG-S, MF-A, VC-C, and MC-M: conceptualisation, methodology, software, validation, formal analysis, investigation, resources, data curation, visualisation, and project administration. SG-S, VC-C, CC-C, and NO-B: formal analysis. SG-S, MF-A, VC-C, CC-C, and MC-M: writing original draft preparation, writing review and editing, and funding acquisition. MF-A and MC-M: supervision. All authors contributed to the article and approved the submitted version.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2021.807869/full#supplementary-material

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Bereaved Families: A Qualitative Study of Therapeutic Intervention

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Background: A child's death is the most stressful event and the most complex grief that families face. The process of psychological adaptation to the illness and death of a child is difficult due to a variety of emotional reactions. Parental grief had received the attention of researchers only in recent years when it became clear that this reality differs substantially from the general grief process.

Objective: This work aims to highlight the needs of bereaved parents; increase the specificity and effectiveness of the therapeutic approach to prevent complications in the process of loss-making; and find the recurrent thematic nuclei in the development of bereavement present in a therapeutic group of parents who have lost their child to an onco-hematological disease.

Method: Between 2011 and 2016, five therapeutic groups for the grief elaboration were made. The sample included a total of 50 parents of children who died from cancer between the ages of 0 and 21 years.

Content analysis was carried out as a qualitative analysis method. The SAS® Text Miner software (SAS Institute Inc, 2004) was used to read, interpret, classify and integrate the data from numerous sources.

Results: The development and consecutive interpretation of the 5 clusters have been carried out to analyze the related topics using the node "Topic Analysis" and requesting the subdivision into five topics. Four topics have been well defined. Clear topics are reducible to categories of emotional *relief, tools, legacy, and unfinished business*. The topic analysis provides interesting indications about the different interpretive journeys of the bereavement situation and offers ideas regarding the different types of social responses.

Conclusions: After reviewing the existing bibliography, we have confirmed the lack of specific literature on the problem of grief in parents whose children have died from cancer. Much research has shown that parents who lose a child to cancer want support, and there are still few studies on the most effective interventions for this group.

Keywords: grief, bereaved parents, psycho-oncology, group therapy, pediatric oncology, meaning-making, legacy

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INTRODUCTION

The death of a child is considered taboo, underlining this type of loss's paradoxical and contradictory nature (Barbero and Alameda, 2009). The process of psychological adaptation to the disease, the death process, and the grieving process over the death of a child is a very complex stage for professionals and the family due to the variety of emotional reactions that happen (Die-Trill, 2003).

The specificity of bereavement in parents has been highlighted by Rando (1998) as an explanatory model different from other types of grief. Rando observes that these factors are as follow: premature loss; quality and nature of the relationship with the deceased, role of the deceased, death characteristics, and quality of the supporting social network. They are grouped to form a characteristic constellation, which explains the depth of the wound suffered by the parents. The death moment is necessary to highlight the importance of both the common aspects and the differential variables in mourning a child's death (Field et al., 2013). Studies have shown that bereavement in parents usually persists and often intensifies, even after the first year after death and is associated with higher Prolonged Grief Disorder severity compared to loss of a parent or a sibling (Lichtenthal and Breitbart, 2015; Zhou et al., 2020).

Tizón and Sforza (2008) says that parents feel that an essential part of them dies when a child dies. "The most important bond of love is affected: love for life itself." Parents lose their child and what their child represents, a perspective toward the future, offspring, dreams (Schiff, 1997; Talbot, 2002; Zheng et al., 2017). The death of a child subverts the natural order of events, being experienced as illogical and against nature (Davies, 2004).

Parental grief has received the attention of researchers only in recent years when it became clear that this reality differs substantially from the general grief process. Different studies (Tan et al., 2012; Zhou et al., 2018; Morris et al., 2019) demonstrate the need to develop specific explanatory models for this phenomenon. Families face the difficult job of building a new world of beliefs and values where the reality of loss can take hold, and this process takes months and years to be complete, even knowing that it may never be resolved. This line understands the importance of providing effective interventions to reduce the long-term complications that may manifest in bereavement (Richardson et al., 2011; Wiener et al., 2019; Kochen et al., 2020).

We are working with bereavement parents in a therapeutic group setting. The group becomes an instrument of change by providing members opportunities to work and talk about the problems in their lives. Studies are still very scarce and often inconclusive, demonstrating the need to explore this methodology and this well-defined theme further.

This work aims to highlight the bereaved parent needs; increase the specificity and effectiveness of the therapeutic approach to prevent complications in the process of loss-making. Find the recurrent thematic nuclei in the development of bereavement present in a therapeutic group of parents who have lost their child to an onco-hematological disease.

MATERIALS AND METHODS

Setting and Recruitment

Between 2011 and 2016, five therapeutic groups for grief elaboration were made. The sample included a total of 50 parents of children and adolescents who died from cancer. The population comes from the "Niño Jesús" University Children's Hospital in Madrid and the Gregorio Marañón General University Hospital. Therapeutic intervention is developed by a therapist who is an expert in psycho-oncology and bereavement at the Aladina Foundation Psychological Care Center. This independent organization dedicates its efforts to cancer children and adolescents. Participants are contacted by a therapist approximately 2 months after the death of their child. Before the start of group therapy, the participants carry out several individual or couple interviews with the therapist. The objective is to sign the informed consent to guarantee the privacy and the use for educational purposes of the data collected during the sessions.

The inclusion criteria for group therapy were (1) being the parents of a deceased child in pediatric age (up to 21 years) treated for an onco-hematology disease; (2) being in grief in a time between 2 months and a year; (3) compliance with group norms of acceptable behavior; (4) motivation to do bereavement work; and (5) not participating at the same time in other group psychological therapies.

The exclusion criteria were (1) inability to tolerate the group setting; (2) tendency to assume a deviant role; (3) disagreement with group standards of acceptable conduct; (4) serious incompatibility with one or more members of the group; (5) lack of motivation to work.

Analysis Data

The *Content Analysis* of transcripts of therapeutic group sessions was used for a qualitative study. The objective of content analysis is "to provide knowledge and understanding of the phenomenon under study" (Downe-Wamboldt, 1992). The SAS® Text Miner software (SAS Institute Inc, 2004) was used to read, interpret, classify and integrate the data from numerous sources. The cluster analysis process consists of three steps.

Cluster Analysis

Phase I: Transcript of the Sessions

The basis from which the data were extracted for the discourse analysis was the transcripts of psychotherapeutic sessions. The therapist wrote the phrases of each patient verbatim. At the end of each session, the notes were reviewed, and the transcription expanded. Transcripts of the interventions of mothers and fathers were analyzed in depth. Recording what they said, and how they expressed it, avoiding modifying their language. Consequently, some fragments appear as grammatical forms and others as colloquial forms.

Phase II: Processing Data

The transcripts, 111 files in total, were prepared in table format to be analyzed by the software. The analysis "Text Parsing" allows the analysis of a set of documents to quantify the information about the terms in it, generating a frequency matrix of the words in the papers. In Text Filtering, the Log function has been used to calculate the weight of the frequency to associate for each term. Attributing weights to words is helpful to distinguish important terms from others. Successively two weighting flows have been created, the "inverted frequency"; in the second one the "entropy." The "inverted frequency" method gives more weight to terms that infrequently occur in the sample of documents by placing the number of records that contain the word in the numerator of the formula. While the "entropy" method gives greater weight to terms that infrequently occur in the sample of documents using a derivative of the entropy measure found in information theory. Once the results were obtained, it was decided to consider the second flow (entropy) for the construction of the topics since it reflected better in terms of the categories obtained.

The Text Filtering and Document Analysis nodes must identify the set of valid terms of which we have filtered 214 definitive analysis units. With the 214 units, we have carried out a hierarchical Cluster Analysis to understand what the ideal partition of the categories could be. We have suggested a division of the data into five clusters.

Phase III: Topic Analysis

The realization and consecutive interpretation of the 5 clusters have been carried out to analyze the related topics using the node "Topic Analysis" requesting the subdivision into five topics. Since the results of this analysis, four topics have been well defined while another is too heterogeneous. Clear topics are reducible to categories of emotional *relief, tools, legacy, and unfinished business*.

The first flow of analysis using the inverted frequency had led to the recognition of a single well-defined topic that could be retransmitted to the category "sense." Considering this data, we have used the terms with the most significant weight of this topic to manually define five topic knot Text Analysis of the second flow. To further verify the results, the words with the most important weight of the topic "meaning" have been measured in the other four topics, and it has been seen that they have a low weight. Therefore, the creation of a different category is justified. With the insertion of the topic defined by inverted frequency, the node has been re-executed, and five well-defined and representative topics have been delineated for the research.

RESULTS

The first topic called "meaning" has been defined by 69 words present in 21 documents. The second topic called "emotional relief" has been described by 27 words present in 16 documents. The third topic called "legacy" has been defined by 17 words present in 14 documents. The fourth one called "tools" has been defined by 31 words present in 11 documents. The fifth one called "unfinished business" has been defined by 31 words present in 19 documents (**Table 1**).

TABLE 1 | Topic analysis.

Topic	Topic description	Number of words	Number of files
Birthday, death, fear, sadness, feeling, living	Emotional relief	27	16
Qualities, understanding, desire, support, person	Tools	31	11
Guilt, accept, why, time, understand	Pending issues	31	19
Legacy, love, life, contact, experience	Legacy	17	14
Contribute, happy, spirituality, tribute, channel	Meaning	69	21

The associations, which each word has with the others, are analyzed through concept linking diagrams that show the frequency with which one word appears next to another. We decided to investigate the words that are not repeated in previous connections and underline the words with closer relations with the central concept to give a broader perspective of the topics.

Sense

We will describe the words to define this topic: contribute, happy, spirituality, tribute, and channel. Internal schemas protect against non-sense, and once loss knocks them down, the emptiness remains. If the emptiness appears as deep pain on an emotional level, on a cognitive level, the emptiness seems non-sense. The word meaning refers to the grieving people's attempt to seek meaning in their pain experience. In this process, mourners discover that the loss can lead to personal growth, such as a new perspective or learning something improved about themselves.

Parents feel the need to contribute to people, to turn their experiences into teachings or examples for others. They have learned how to deal with difficult times; they feel proud of their children and themselves for having enriched themselves with this experience and for having collapsed.

> ***"Thanks to my daughter and everything that has happened, I know how to say no, I know how to live without anyone conditioning me, and I do not prepare anyone. I have changed my values; I give importance to what has it, I express my feelings more."***

Happiness is felt again, with the premise that you can no longer be happy as before, and you must build another way of being happy.

> ***"I feel fortunate to have been able to meet you and teach me the purest and most eternal love that will ever exist, mother-child love."***

When death ceases to be scary and is integrated into everyday life, a connection is felt with another dimension, a spiritual dimension that should not be seen only as a call to religion but as a greater goal.

> ***"I want to help others; I want to show more intensity for my own, I want to say I love you more times with more value, I recognize the importance of health, taking care of you, I have learned to say no, I am more spiritual."***

Mourners try to give a new meaning to their lives by living it as a tribute to their children. They want to show that their children have transmitted their courage, the same with which they have faced illness and death. Parents cannot leave this learning in a vacuum since it is already part of them. Thus, they rescue it and teach it.

> ***"This whole situation made me stronger, braver, more fighter, and taught me not to let minor problems destroy me. My daughter V. came into the world to make me a better person and give me a great life lesson."***

Emotional Relief

The words that we will describe to define this topic are birthday, death, fear, sadness, feeling, living.

The word "birthday" synthesizes, on the one hand, memories, death, everyday life, friendship, the other, peace, spirituality, and positivity.

Dates such as birthdays, the anniversary of death, and many others that come out of everyday life sharpen emotions in an ambivalence between a feeling of peace, when contacting the memory of the son, or of agony if it is related to the absence, with death.

> ***"The birthday went well because it is a good day to remember and celebrate, his birth is celebrated, the other anniversary reminds you of his death."***

Fear is a very common feeling, and at this time of therapy, it is related to contact emotions, opening to this world in which pain and sadness predominate, in which an existential emptiness encompasses everything. The fear of developing depression is present, as well as the fear of never leaving this melancholic state and not knowing how to continue living. Fear serves as a sentinel and an emotion block, which is why it is so important to work with it. Furthermore, facing fear serves to be able to start feeling. Another recurring fear is the fear of forgetting; parents feel that if they stop suffering, feeling sorry, crying, being angry, they lose the essence of their children, lose contact, and forget them.

> ***"I am very afraid to forget, fear of not being able to approach this pain in another way, fear of never being able to have a close person and give support to those who suffer because it makes me feel too sorry; fear of rejecting all this so as not to feel again."***

The first emotion that dominates in grief is sadness, huge, deep, portentous, which envelops absolutely everything and never seems to placate. One way of how sadness is expressed is the veto to enjoyment, to happiness. An unconscious treatment is made in which, in memory of the son, any form of well-being is prohibited, and if it occurred, it would be like a betrayal. And it is then that the guilt appears.

> ***"I do not want to condemn myself and my family to a state of permanent sadness."***

"Feel" is the core of emotional release. After beginning to express experiences and memories, leaving aside the inner monolog, and opting for a shared verbalization, fears appear. These are the first defense mechanisms that help us not to feel since we cannot feel when we are not prepared. A dichotomy opens when you outline the possibility of returning to life. The energy of life moves the death, the desire to take advantage of life along with the things that are worth, removed fear.

> ***"Thanks to the fear of losing everything, now I am going with my family instead of doing more hours at work."***

Legacy

It is important to be able to create bonds that connect the deceased person and return the bond, which helps elaborate the loss. The important thing is to realize how we need to put what has been lost in a good place in order to light us up and light up to keep walking, instead of clinging to its memory anchored in the past or trying to bury it. It is important to give a new internal place rather than fear or seek oblivion. The words that define this topic are legacy, love, life, contact, experience.

The legacy they leave is an immense love that teaches them what the true way forward is.

> ***"I want to live from day to day. I have stopped making sense of having things and making plans, now the values are different."***

The parallelism between life and anguish has changed, and the look to life with strength prevails. Parents continue toward a new connection to life, from the love that makes them feel proud and strong.

> ***"Now I see life differently, I take advantage of every moment to follow my needs, there is no future project in the heavy sense of responsibility, I enjoy the day to day."***

Throughout the therapy, new ways of contacting the children other than through grief are learned. We can see now how the fear of letting go of grief.

> ***"Since my daughter died, I have felt a lot of peace this has made me understand that my daughter was going to stay with me, I don't want that thinking about my daughter carrying pain and sadness because she does not deserve it, I will always be happy to have had a daughter and to be able to think of her."***

Tools

The intensity of pain and suffering is enormous, so parents need to defend themselves. Within limits, these defensive maneuvers in grief can be useful. There is a need to defend yourself to "adapt" to the reality of loss, in any way that everyone is able.

The words described to define this topic are qualities, understanding, desire, support, person.

The word "Qualities" emphasizes self-knowledge and knowledge of others, assuming this material as a resource.

> ***"I have discovered these qualities. to be understanding and able to listen and talk about pain, love, patience."***

Mourners face new thoughts and sensations, some heartbreaking, others embarrassing. In group therapy, there is a climate of respect and trust that favors an expression without judgment of which understanding is the fundamental axis.

> ***"I just want to share how I feel about who understands him like you."***

Discovering denied parts, validating internal resources, sharing with the group doubts, questions, blocks and achievements is necessary to regain the desire to face life, to seek other ways of living and to recognize and face problems and emptiness.

Emptiness is something that everyone feels in front of the loss of a son or daughter. It is also something that must be defended while it cannot be sustained.

> ***"I have also found many people who love me; love for others is where I want to start, now I am looking for the good. I have to move forward. And the group helps me because I share, because I know other ways to carry out grief. I feel good."***

Parents feel support, so they can move forward in their ways and find themselves as people, not only as mourners.

It is now when the mourners can see the defenses that no longer serve as avoidance, rationalization, rumination, and they can advance on the path of grief.

> ***"It distressed me a lot, and these days I feel that I have moved toward acceptance; we have all worked hard. I have learned from myself that I can express more."***

Unfinished Business

Unfinished business with whom we have lost makes separation difficult. Guilt, resentment, doubts act as invisible threads that bind parents and prevent farewell. As a result, energy is dispersed in the past, and parents look back and do not face the present in full potential.

The words that define this topic are guilt, accept, why, time, understand.

Guilt is one of the emotions that are most associated with grief and sadness. It often expresses itself in different ways. It can be felt toward oneself or someone else. In this case, it is closely connected to impotence since parents feel that they have not been able to defend their children. They also feel that they have failed to fulfill the primary function of the parent: protection.

> ***"I often feel guilty because I want her here with me, I want to touch her, and I can't and impotence kills me and I feel much anger. Then I think about how bad she was, the suffering caused by the disease, and I feel very selfish to love her like that with me."***

Accepting is the end that everyone wants to reach and feels that they cannot.

For each parent, the word "acceptance" has a different nuance but a common goal: to learn to live again.

> ***"Acceptance is to understand and believe that you can live with the absence you have left."***

Death in children is seen as an event against nature, since it is not possible to fit in a logical order, and therefore, it is not understood.

An infinite range of "whys" and "what ifs" opens up until it becomes a labyrinth that traps parents, preventing progress in bereavement.

> ***"The more time passes, the more I feel bad, the bad thoughts do not go away and only the traumatic images come to me."***

The concept of time is something complicated to explain for the mourners because the future cannot be thought of and the past is a continuous remembering and recalling trying to understand and retain. When parents feel that the months go by and instead of mitigating the pain increases, the fear of being "sick" grows, of never getting better, of being condemned to a life of suffering.

All the systems of certainties and logic that sustained each one's life collapse during grief and "understanding" requires much effort. They work to understand all these answers so that they can close back on unfinished business and focus on new tasks.

> ***"I want to isolate myself because nobody understands it and people say non-sense, and for this reason, I prefer not to speak. In the group, I feel comfortable and understood."***

DISCUSSION

The topic analysis provides interesting indications about the different interpretive journeys of the bereavement situation and offers ideas regarding the different types of social responses. The study and analysis of the speech of bereaved parents help us to understand the strategies that parents use to go through the grieving process, which goals and objectives are proposed in both the long and short term, and which tools they use in order to face the grief. This analysis helps both professionals and mourners to understand what the new construction of identity is. How the meaning resulting from the bereavement process and what are the categories that sustain it, some being linked to others, some independent, and some that end when others appear.

One of the most recognized benefits of group therapy is the creation of a space for the expression of emotions (Heiney et al., 1995). At this time, it is appreciated that experiencing pain involves allowing yourself to feel the full range of feelings and thoughts that accompany a loss, including sadness, anger, fear, anxiety, shame, guilt, and even relief. Although emotions are indeed forces that push us, shake us and wobble, it is also true that we can regulate them: feed them, understand them, express them, avoid them, and go through them. On the other hand, emotions also have an essential informative function. They tell us if our needs are being met or not, if we are respecting our values, if our ties are in danger.

As it has been possible to verify in this study, the topics that we mainly faced in the initial phase of the group have been remembering the most important events in the children's lives; reconstructing the circumstances of death and illness; sharing emotions and coping mechanisms.

The emotional bereavement state is not a linear progression, it is instead a constant adaptation to an emotional roller coaster that has no logic or temporality. In addition to the value of emotional awareness as a source of information, symbolizing emotion in awareness promotes reflection on the experience to create new meaning, which helps patients develop new narratives to explain their experience (Greenberg, 2012).

Bereaved families often struggle against double isolation: one that is voluntary, since they are afraid of burdening their support network with their persistent sadness; and the other that is forced, since they perceive that social support diminishes over time (O'Connor and Barrera, 2014; Ljungman et al., 2015). Throughout group psychotherapy, parents once again trust people, open up again with others, share because they no longer feel "out of place."

The therapeutic process begins with becoming aware of the defense mechanisms that are used against the suffering caused by loss. These are unconscious behaviors that allow us to defend ourselves from the threats that we perceive in our integrity as people, in our values and serve to avoid facing something that cannot be integrated or that feels intolerable. During therapy, tools are learned to know and manage the defensive mechanisms, to normalize them if they are adaptive and to change them if they are not helping mourners.

A relatively common source of discomfort and anxiety for grieving individuals is represented by unfinished business, as highlighted by the few empirical research on this construct (Steinhauser et al., 2015). In addition, different studies have shown the correlation between the presence of unfinished business with prolonged grief and other psychological and psychiatric disorders (Horowitz et al., 1993; Klingspon et al., 2015; Holland et al., 2020). Therefore, one of the most important tasks that can be done in bereavement is to help patients deal with their unfinished business, on which they are stuck or blocked.

Possible emotional responses to unfinished business can include various reactions, such as regret, anger, guilt, or remorse (Klingspon et al., 2015). Guilt deserves special mention since it is one of those responsible for the failure to close the bereavement and one of the most studied in this area Grinberg (1971). An important aspect of directing the work is to identify if there is regret behind the guilt. It represents a strong link with unfinished business (Klingspon et al., 2015) and it is emotionally related to unfavorable responses to one or different decisions made during treatments. Understanding the cause of death, talking about the course of illness and treatment, rebuilding the last hours of one's child's life, has been denoted as facilitating aspects of bereavement (Meyer et al., 2006; Meert et al., 2007; Eggly et al., 2011).

Studies identified the presence of unfinished business as an unfavorable cause for adaptive grief. Klingspon et al. (2015) emphasize that the presence of unfinished business shows worse results in bereavement, as indicated by more severe prolonged grief symptoms, significant psychiatric symptoms, and lack of construction of meaning regarding loss. In this study, we have seen how unfinished business are kept in a last and desperate attempt not to face the emptiness of loss, to remain stuck where we are, even if it is a place full of pain from the fear of letting go.

Another aspect that we have to pay attention to is what we call "legacy." The use of legacy, of the continuous bonds with the deceased as a mechanism of adaptation to grief, is a new aspect and still little studied. The most influential authors (Klass et al., 1996; Currier et al., 2006; Neimeyer et al., 2006) do not reach the same conclusions about the usefulness of this construct for an adaptive bereavement process.

Klass (1997) confirm that the persistence of a strong bond with the deceased relative helps a reasonable resolution of the mourning; while authors such as Lehman et al. (1987) and Stroebe et al. (2012) question that a continuous bond may be an aspect that favors bereavement, and they believe that in some cases it may even make mourning work difficult. Currier et al. (2006) and Neimeyer et al. (2006) refer that the adaptive or maladaptive nature of the legacy depends on the way it is used by the bereaved. The disparity of opinions is explained in the light of the two meanings that can be attributed to the concept of a continuous bond. Connections with the loved one may be physical, such as having an object that serves as a memory. These rituals are defined as external links, prevalent in an immature and initial phase of bereavement and they are characterized by the inability to accept the physical absence of the loved one (Field et al., 2013; Zhang and Jia, 2018). While internalized forms can be sharing anecdotes, having lessons to follow, or feeling support in vital moments. In this sense, we refer to legacy, and we interpret it as an adaptive way of coping with grief (Field et al., 2005; Schaefer et al., 2020).

In this study, we showed how the individuation of an evident legacy with the child turns out to be adaptive and favorable for the parents. Many parents claim that their lives are guided by what was essential to their children. They describe living as the way to do things that their children valued or that their children had not been able to do. If parents manage to find the legacy that unites them with their children, they will be able to access a new sense of meaning.

It is important to note that parents struggle to make sense of their child's loss, and those who are unable to do so are at greater risk for symptoms of prolonged grief; while the creation of meaning is associated with a better adjustment (Keesee et al., 2008; Lichtenthal et al., 2019; Grassi et al., 2021).

Gillies and Neimeyer (2006) propose a specific model for the reconstruction of meaning. The creating meaning process happens through three specific ways: the search for meaning, the search for benefits, and the identity change. Some families use religious and spiritual beliefs as a framework to make sense of their loss and find meaning, both in their lives and in those of the deceased (Folkman, 2001). In this study, we have seen how spirituality for some, covers an important role in achieving a new meaning to loss.

Various processes that make up the reconstruction of meaning seem to mitigate the impact of other risk factors focused on the characteristics of the individual grieving, the relationship with the deceased, and the death itself (Neimeyer et al., 2006).

Keesee et al. (2008) found that 47% of grieving parents were unable to create any meaningful sense of their loss for an average of 6 years. In other studies, many parents emphatically express that the loss of a child does not have a sense (Lichtenthal and Breitbart, 2015). In accordance with our study, these results would explain why the construction of meaning, even being helpful to the bereaved, is not easily achieved or accepted and explain why we had to work on the topic of "sense" the "reconstruction of meaning."

In this area, it should be noted that this is a delicate issue to work with parents who, understandably, may have a difficult time identifying any positive outcomes that are associated with their loss.

As it was to be expected in this study, we had also encountered resistance when it was questioned whether we could find meaning in the death of children or could cause a positive movement in their lives. When explaining the topic to parents, they have recognized their work within this category and affirm that it is a complicated and laborious step, since it confronts them with the enormous turn their lives have taken; and in some cases, they are reluctant to get rid of the pain and of a new form of happiness.

Limitations

We should pay attention to some limitations of this study. Firstly, it has intrinsic limitations to transcriptions there need to be very detailed and reflect tones of voice, inclinations, pauses that are sometimes difficult to write down. The results were triangulated to face this limitation, confirming them with the existing bibliography and carrying out different Focus Groups (Moriconi and Cantero-García, 2022) with the parents who participated in the therapeutic groups to discuss the findings. Second, it is important to note that the distortion factor can be given by the interpretation and subjectivity of the same researcher. Finally, it is essential to point out the limitations regarding the sample in terms of number and homogeneity.

Conclusion

After reviewing the existing bibliography, we have confirmed the lack of specific literature on the problem of grief in parents whose children have died from cancer. Much research has shown that parents who lose a child to cancer want support, and there are still few studies on the most effective interventions for this group. Because of this scarcity, this work turns out to be a starting point and an incentive for future research.

Once we have analyzed the data related to the therapeutic sessions of the bereavement groups, we have been able to show five well-defined thematic nuclei that are well defined and not confluent with each other. We have named the topics Sense, Emotional Relief, Tools, Unfinished Business, and Legacy.

This study aims to help us understand the difficult mechanism that is set in motion in parents when a child dies, what they need and how it can be provided with effective and timely interventions. Health professionals who work with this group must bear in mind that each member of the family can experience and cope with loss in qualitatively different ways. Therefore, parents and close relatives need a well-defined and expertly guided space to be able to grieve. In addition, emphasis should be placed on the idea that there is not a "right" way to grieve, and the support must be built around individuals and what is useful during the specific grieving process.

The implications that this study has in clinical practice are useful not only in inherent bereavement in death, but also in the work of bereavement understood as a loss. Therefore, knowing the thematic nuclei on which the psychic elaboration of the trauma moves is relevant in all phases of the disease. Knowing the mechanisms underlying the development of loss prevents not only psychological suffering but also an increase in the human quality of care for patients.

DATA AVAILABILITY STATEMENT

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

ETHICS STATEMENT

Ethical review and approval was not required for the study on human participants in accordance with the local legislation and institutional requirements. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

VM: conceptualization, methodology, investigation, software, data curation, and writing—original draft preparation. MC-G: visualization, supervision, and writing—reviewing and editing. Both authors have read and agreed to the published version of the manuscript.

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BRIEF RESEARCH REPORT
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The Intervention Areas of the Psychologist in Pediatric Palliative Care: A Retrospective Analysis

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Infants, children and adolescents with life-limiting and life-threatening disease need long-term care that may change according to disease's natural history. With the primary goal of quality of life, the psychologist of pediatric palliative care (PPC) network deals with a large variety of issues. Little consideration has been given to the variety of intervention areas of psychology in PPC that concern the whole life span of the patient and family. The PPC network is composed by a multidisciplinary team of palliative care specialists that intervenes at home, in the hospital and in every place where the patient is living. The network coordinates different public health services to respond to clinical, psychosocial and spiritual needs. In these scenarios, the psychological need is not a single event but a moment inserted in the complexity of the child's needs. This retrospective monocentric project consists of an analysis of characteristics of psychological interventions in our PPC service. The time frame taken into consideration is 2019–2020, analyzing the clinical records of 186 patients of Pediatric Palliative Care and Pain Service of Veneto Region (Italy). The areas that emerged in the analysis show how the intervention of the psychologist in PPC does not concern only end-of-life, but a series of topics that are significant for the family to guarantee psycho-social wellbeing oriented toward the best quality of life. In conclusion, these different topics highlight the complexity of the child and family experience. This variety must be taken into consideration, the psychologist must increase holistic support with a dedicated skills curriculum.

Keywords: pediatric palliative care (PPC), retrospective analysis, biopsychosocial model, intervention areas, psychologist

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INTRODUCTION

Palliative care for children and adults is defined by the World Health Organization as the active and global care of the child with life limiting and life threatening diseases and his family. This care involves managing the child and families' physical, psychosocial and spiritual needs (World Health Organization [WHO], 2018). The criteria for eligibility for pediatric palliative care (PPC) primarily include the presence of a diagnosis of life limiting and life threatening diseases and a highly complex care situation (Benini et al., 2008). PPC are provided by a multidisciplinary specialist team (Fitchett et al., 2011; O'Connor and Fisher, 2011; Verberne et al., 2017; Cervantes Blanco and Jones, 2018) and interface with the professionals present in the other health centers. The network is a functional and integrated aggregation of PPC activities delivered in hospital, pediatric hospice and at home. It is made up of all the institutions necessary for the child care management (hospital and territorial health, socio-health, social-assistance and educational services) and integrates and collaborates with the networks, pediatric and not, existing in the geographic area (Bergstraesser et al., 2013). The psychologist is part of this specialist team which aims to respond in a synergistic way to global needs that are taken care of in a biopsychosocial paradigm (Quill et al., 2003; Brown, 2006; Moore, 2019; Ribbers et al., 2020; Benini et al., 2022).

The experience of the caregivers of young patients is widely taken into consideration. The researchers underline patients' family members' fatigue and different psychosocial needs (Jünger et al., 2010a; Collins et al., 2016; Perpiñá-Galvañ et al., 2019) who often live with a strong sense of guilt (Gonyea et al., 2008). The impact of being a caregiver involves social isolation and work difficulties (Wakabayashi and Donato, 2005; Knapp et al., 2010; Price et al., 2012; Collins et al., 2016; Harputluoğlu et al., 2021). Stress, anxiety and depression are common in parents (Collins et al., 2016; Wightman, 2020) and can generate traumatic experiences (Mitchell et al., 2006; Kars et al., 2011). Such emotions are present not only in adult caregivers but also in pediatric patients and their siblings (Barlow and Ellard, 2006; Collins et al., 2016; Weaver et al., 2019).

Family's reaction mechanisms are a promoter of the child's health (Cipolletta et al., 2015) and this allows them to develop coping and emotional management strategies (Sloper, 2000; Goldbeck, 2006; Felipe Silva et al., 2021). Psychological needs can be traced throughout the disease's history, which can be very long and can have different critical moments (Mitchell et al., 2006; Liben et al., 2008). Caregivers often feel they are in a battle and use the metaphor of "combat" to describe their story (Knapp et al., 2010), so continuous monitoring of psychosocial needs is crucial (Goldbeck, 2006; Mitchell et al., 2006; Liben et al., 2008; Weaver et al., 2016). The PPC psychologist takes care not only of the patient, but of the entire family (Kazak and Noll, 2015).

Pediatric palliative care is not limited to end-of-life support, but involves the whole natural history of child's disease, even where specific treatments for the disease are pursued (Nilsson et al., 2020).

Despite this, PPC is often associated only with end-of-life care and this leads many professionals to not use the service adequately or refer patients late (Thompson et al., 2009; Twamley et al., 2014). Professionals show a certain reluctance to send families to CPP (Davies et al., 2008; Knapp and Thompson, 2012) both because they struggle to use certain terms with their parents (Davies et al., 2008; Durall et al., 2012) and because families can perceive this as a giving up by lead physicians (Thompson et al., 2009).

Furthermore, there is a lack of knowledge on the subject of PPC and this leads to little knowledge of temporal indicators, protocol and strategies (Thompson et al., 2009). In evaluating the specific challenges that differentiate CPP from adult CP, the following are pointed out: the need to offer family support to the whole family and not just to the patient; the presence of pathologies that can have very long life expectancies; the difficulty of having a health system that meets the special needs and priorities of children; the variety of care needs of different clinical conditions; the difficulty in relating not with the patient but often with family members and the tensions that arise; the priority of managing communications taking into account the parental authority and the communicative possibilities of the young patient; the need for highly specialized skills and knowledge (Jünger et al., 2010b). However, some works are also focusing more professional attention on the PPC issue and more recognition of PPC services in ensuring the patient's quality of life (Jünger et al., 2010b; Bergstraesser et al., 2013).

The purpose of this work is to show, through the retrospective study of clinical diaries, the complexity of the psychologist's work within the CPP network, in order to increase knowledge of a holistic and global care model.

METHODS

The sample examined was 225 patients, of which 57 were excluded due to private psychological counseling or absence of psychological sessions records at the time of the study. Therefore the sample of the clinical reports analyzed was 168 patients, of which 49 died. The time range taken into consideration runs from 01/01/2019 to 12/31/2020. The service had a number of variable psychologists (3 between 01/01/2019 to 01/07/2019, 2 between 02/07/2019 to 01/10/2020, and 1 between 02/10/2020 and 31/12/2020). In addition, this period also includes the beginning of the COVID-19 emergency, for which the activity of our service has seen a change since 07.03.2020.

The analysis objectives were to identify: the different teams with which the psychologist works during communications, the settings and target of psychological interviews, the interviews' topics.

The material examined includes clinical reports, consultations and clinical charts for a total of 1292 interventions: 511 interventions by the PPC team and 781 psychological sessions. The psychological work has been divided on the basis of a thematic analysis (Vaismoradi et al., 2013; Nowell et al., 2017) which has made it possible to identify homogeneous thematic areas by topic. This process took place following the indications of Nowell et al. (2017), using the clinical material produced by psychologists. A deductive approach was used, the writings were analyzed using *a priori* categories relating to the psychological issues found in the literature and compared with the clinical experience of the PPC psychologists.

These categories have been discussed numerous times on the basis of the readings of clinical material, two different operators have worked on databases (authors AM, AS) and all discrepancies and uncertainties have been addressed through a peer-debriefing.

The thematic areas identified are:

- "Medical stress and symptoms management": needs to build, modify or emotionally manage issues related to the disease and its symptoms.
- "Functional loss or device introduction": critical issues and strategies of intervention focused on adaptation to a disability increase resulting from a function's loss (e.g., mobility, sight, urination) or to a new medical device introduction (e.g., ventilator, wheelchair, tracheo).

- "Parenting and relationship": issues related to practical and emotional management of significant family relationships but also formal (e.g., school, work) and informal network (e.g., friends).
- "Emotional displays": manage expressions of emotions (e.g., anxiety, fear, depression, joy).
- "Behavioral manifestations": behavioral expressions that are considered critical for some reason (e.g., hyperactivity, impulse management difficulties).
- "Existential concerns": thoughts related to the signification of events, the purpose of existence and anticipations of the future.
- "End of Life": issues related to the critical moment that has been communicated at that time or that is glimpsed in the near future.
- "Advance care planning": thoughts and experiences related to a decision on the proportionality of care for the patient's disease.
- "Management covid pandemic period": thoughts and experiences related to the management covid emergency.

RESULTS

Socio-demographic variables (**Table 1**) showed a patient average age of 9.5 years, mostly male (54.1%). The most frequent disease was non-oncological one (79.76%). Parents spoke fluent Italian for 88.10% of the sample, most of them were also a married couple (90.48%). In these families, another psychologist was rarely already present before accessing PPC service (6.19%).

As interventions with psychologist presence were all communications that involve psychologists with other members of the PPC team (**Figure 1**). The highest number of communications were in Hospice with members of the CPP team (31.3%) such as communications relating to treatment, worsening clinical conditions, end-of-life communications, etc. Other interventions included discussions with public and private territorial structures (27.2%) with the aim of sharing family needs and coordinating resources. The PPC team also carried out consultations in local hospitals (18.0%). A structured and formalized intervention was carried by a multidimensional assessment unit to define an individualized care project. Furthermore, the PPC psychologist carried out information and training meetings for school staff (8.8%).

TABLE 1 | Socio-demographic variables.

Socio-demographic variables (tot = 168)	
Mean age, years (st. deviation)	9,49 (± 6)
Male, number (%)	91 (54,17%)
Type of disease, number (%)	
Oncological	27 (16,07%)
Non-oncological	134 (79,76%)
Undefined diagnosis*	7 (4,17%)
Fluent italian, number (%)	148 (88,10%)
Parents couple status, number (%)	
Unite	152 (90,48%)
divorced	13 (7,74%)
Widow/-er	1 (0,60%)
Foster family	2 (1,19%)
Presence of other psychologist, number (%)	44 (6,19%)

A total of 781 psychological sessions were carried out and divided on the basis of target (**Table 2**). Most of the interventions were carried out with caregivers (63.5%) and then with patients (18.4%). Less frequently, psychological sessions were carried out with the couple (8.3%), siblings (4.7%), entire family unit (2.7%), and extended family (2.3%). In general, the most addressed issues were relationship management (26.50%) and emotional manifestations (25.22%). Specifically for the target, the parent's sessions concerned manifestations management (43.75%) and relationships management (20.83%). Caregivers, on the other hand, mostly dealt with parenting and relationships (31.45%) and emotional variables (19.56%) management. Couple sessions mainly concerned end-of-life (47.69%). The end-of-life topic latter issue was more commonly addressed in interviews with the extended family (38.89%), while the family in the strict sense mainly managed interpersonal relationships (33.33%). Siblings, instead, dealt more with emotional management (51.35%).

In general (**Table 3**), the most common setting was hospice (52.5%), followed by sessions conducted online (27.7%) and by those at home (14.6%). This particular covid-emergency period may had led to favoring online sessions. The least used setting was the hospital (5.12%). Although the hospice setting was the most common, the out-of-hospital setting (home and online) was used for 43.75% by patients, for caregivers for 46.37%, for couples 20%, for siblings 37.84%, for the family 47.62% and for the extended family it was only 5.56%.

DISCUSSION

Teams

The psychologist is one part of a PPC professional's network, because of this a large number of communications is given in the presence of some team members based on the purposes (31.31%, n = 160). In addition, the psychologist as part of PPC is present in the communications that are given both with hospital and territorial professionals networks (68.69, n = 351). The psychologist is inserted during communications with the aim of being a facilitator of communication and witness to continue interviews later with the family and the child. The PPC psychologist participates in communications with the aim of being communication facilitator and witness to continue the psychological work later.

Settings

Our results show a flexibility of the psychologist's work with regard to the setting that could be home or online (42.38%) even if a higher percentage of interviews is conducted in Hospice (52.5%). Hospice, as a clinical setting for follow-up, is where evaluation and diagnostic assessment take place.

However, when the family returns home the psychologist enters the family routine. The many sessions conducted

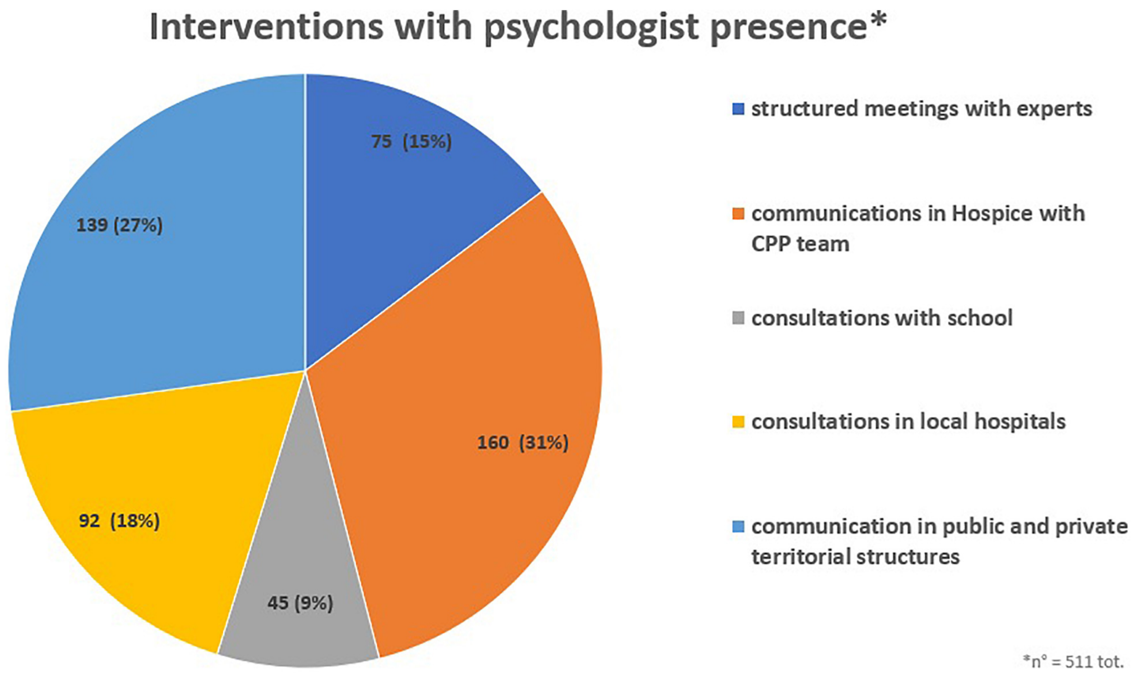

FIGURE 1 | Interventions with psychologist.

TABLE 2 | Thematic areas.

Thematic areas							
	Patient, *n* (%)	**Caregiver, *n* (%)**	**Parents, *n* (%)**	**Siblings, *n* (%)**	**Family, *n* (%)**	**Extended family, *n* (%)**	**Total, *n* (%)**
Medical stress and symptoms management	27 (18,75%)	72 (14,52%)	7 (10,77%)	1 (2,70%)	2 (9,52%)	1 (5,56%)	110 (14,08%)
Functional loss or device introduction	7 (4,86%)	29 (5,85%)	1 (1,54%)	0 (0%)	0 (0%)	0 (0,00%)	37 (4,74%)
Parenting and relationship	30 (20,83%)	156 (31,45%)	8 (12,31%)	4 (10,81%)	7 (33,33%)	2 (11,11%)	207 (26,50%)
Emotional displays	63 (43,75%)	97 (19,56%)	9 (13,85%)	19 (51,35%)	3 (14,29%)	6 (33,33%)	197 (25,22%)
Behavioral manifestations	4 (2,78%)	3 (0,60%)	1 (1,54%)	2 (5,41%)	4 (19,05%)	0 (0,00%)	14 (1,79%)
Existential concerns	8 (5,56%)	42 (8,47%)	5 (7,69%)	2 (5,41%)	0 (0,00%)	2 (11,11%)	59 (7,55%)
EoL	0 (0%)	55 (11,09%)	31 (47,69%)	9 (24,32%)	4 (19,05%)	7 (38,89%)	106 (13,57%)
Advance care planning	0 (0%)	7 (1,41%)	3 (4,62%)	0 (0%)	0 (0,00%)	0 (0,00%)	10 (1,28%)
Management covid	5 (3,47%)	35 (7,06%)	0 (0%)	0 (0%)	1 (4,76%)	0 (0,00%)	41 (5,25%)
Total	144 (18,44%)	496 (63,51%)	65 (8,32%)	37 (4,74%)	21 (2,69%)	18 (2,30%)	781

TABLE 3 | Psychological sessions setting.

Setting							
	Patient, *n* (%)	**Caregiver, *n* (%)**	**Parents, *n* (%)**	**Siblings, *n* (%)**	**Family, *n* (%)**	**Extended family, *n* (%)**	**Total, *n* (%)**
Home	34 (23,61%)	45 (9,07%)	10 (15,38%)	14 (37,84%)	10 (47,62%)	1 (5,56%)	114 (14,60%)
Hospital	5 (3,47%)	25 (5,04%)	6 (9,23%)	1 (2,70%)	1 (4,76%)	2 (11,11%)	40 (5,12%)
Hospice	76 (52,78%)	241 (48,59%)	46 (70,77%)	22 (59,46%)	10 (47,62%)	15 (83,33%)	410 (52,50%)
Online	29 (20,14%)	185 (37,30%)	3 (4,62%)	0 (0,00%)	0 (0,00%)	0 (0,00%)	217 (27,78%)
Total	144 (18,44%)	496 (63,51%)	65 (8,32%)	37 (4,74%)	21 (2,69%)	18 (2,30%)	781

online are probably increased due to the historical moment of the covid pandemic, even so these are effective means of structuring the therapeutic alliance (Lai et al., 2020).

Target and Content Analysis

These results show that psychological interviews targets are mainly caregivers (63.51%) and it is less frequent that they are patients (18.44%). This data can be explained by the fact that children in PPC may have various cognitive disabilities that do not allow them to have a proper psychological session. Furthermore, the average age of the sample is 9 and a half years, with a standard deviation of 6, which is why not all patients are currently able to have a psychological session. However, parents are a common target of psychological sessions (Strada and Sourkes, 2009), but it is crucial to analyze a child's cognitive and understanding abilities. The child always has the right to know his situation and express his wishes (Benini et al., 2014b).

Overall, the prevailing themes are management relationships (26.5%), emotions (25.22%) and medical stress (14.08%). These results are in line with some research in the literature that shows that negative emotions such as anxiety, fear, depression are very present in patients and families (Mitchell et al., 2006; Collins et al., 2016).

The management of uncertainty becomes the norm and allows us to create coping mechanisms to deal with new problems and critical issues (Santini et al., 2020). Part of the psychologist's work is to stimulate family resilience. The changes also affect the quality and quantity of relationships in different environments (e.g., work for parents and school for children). Children in CPP have a very variable life expectancy and unpredictable disease course, which is why they face many psychological and social challenges.

If patients need to talk mainly about their emotions (43.75%), parents are more focused on how to relate to their children and to the formal and informal network. From a systemic perspective, the meetings with both parents focused on end-of-life management and the sessions with extended family also had this topic. The importance of being able to discuss the future and the difficult moment of the death of a child emerges, the role of the psychologist here is to allow the sharing of thoughts, emotions, memories and wishes. Managing everyday family life can become difficult after bad news, it can be equally difficult to enjoy the present moments without thinking about death (Lou et al., 2015; Verberne et al., 2019).

It is interesting that siblings have dealt above all with the emotional part, perhaps because brothers are often put in a position to witness complex situations that are not always explained to them and at the same time they develop greater maturity and independence (Gaab et al., 2014). However, greater attention and space should be given to siblings' personal experiences.

LIMITATION

This work shows the variety of the psychologist's skills in CPP, but the main research limitations concerning database. The clinical records are certainly not as punctual as verbatim transcripts and not all contacts between the family and the psychologist have been officially documented. Interviews cover the history of the disease, but it is interesting for future work to analyze the psychologist role after patient death. The data collected can be a first exploration, it would be interesting to be able to evaluate also the effectiveness of the interventions or analyze drop-outs. It might be interesting to consider the perspective of the patient and the family through a qualitative analysis of their point of view. Other future interventions could be aimed at understanding which interventions were useful and why.

CONCLUSION

The management of complexity is the mission of PPC professionals, the psychologist is called to improve transversal skills and specific intervention methods. The psychologist must first of all show flexibility, combined with the ability to 1dialog with all professionals and be able to deal with different topics with changing targets. Health is promoted in different places of care, which are not only the hospital environment.

In this regard, a dedicated skills curriculum has been developed in the Italian PPC (Benini et al., 2014a). The main characteristics that are required of the psychologist working in this field are:

- ability to analyze psychosocial characteristics and potential issues of clinical situation;
- ability to structure an emotionally meaningful relationship with the child and his/her family members aimed at orienting the team's work to the most appropriate communication methods;
- ability to carry out a clinical evaluation and psychological intervention projects aimed at children, couples and families in the developmental phase of the disease, in situations of chronic pain and in the proximity of death and bereavement;
- ability to integrate one's specific professional contribution within the team work in order to promote awareness of the emotional and relational dimension in the working group;
- awareness of one's own emotional sphere and personal mechanisms for constant self-observation;
- organizational and administrative skills necessary to contribute to the development, institutional/territorial rooting and management of specialisti PPC networks;
- continuous training on methods and intervention techniques and ability to manage research projects.

In general, it is necessary to think of a clear training path for the psychologist in PPC because this expert is called upon to operate in extremely complex areas.

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

AM, AS, and VD were involved in the data recollection and thematic analysis. IA and AS drafted the manuscript. PL, GP, and FB were involved in the critical revision of the manuscript and approved the final version of the manuscript. All authors contributed to the article and approved the submitted version.

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Developing a Model for the Establishment of the Hospice Care Delivery System for Iranian Adult Patients With Cancer

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Introduction: Making appropriate plans for the provision of hospice care is considered a perceived need in the Iranian health system. The current study aimed to develop a model for establishing hospice care delivery system for the adult patients with cancer.

Materials and Methods: This study is part (data of the third and fourth phases) of a larger study that has been done in four phases. This Health System Policy Research (HSPR) utilized a mixed qualitative-quantitative approach. At the first phase, a qualitative study was conducted which explained the care needs and the requirements for establishing this system from the stakeholders' perspective (directed content analysis). The second phase aimed to examine the current situation of hospice care delivery in Iran and identify and determine the similarities and differences among them in the selected countries (comparative study). At the third phase, the main areas of the model and the related indicators were extracted and prioritized by consulting with experts (Delphi survey). Then the model was formulated. At the fourth phase, the proposed model was validated and finalized in terms of importance, scientific acceptability, and feasibility.

Results: Based on the developed model the first and the most important step in establishing the hospice care delivery system is laying the groundwork in the health system which requires focusing on policymaking. It is necessary to establish hospice centers and implement public awareness raising programs, train, and supply expert manpower, strengthen family physician and referral process, formulate clinical guidelines, encourage the private sector and NGOs(Non-Governmental Organizations (NGOs).) to invest, develop end-of-life and hospice care service packages, and create quality care indicators. The proposed model had moderate feasibility.

Conclusion: This model was developed based on the current Iranian healthcare structure and the needs of terminally ill cancer patients. It can be used as a model tailored to the current state of the health system and community in Iran. It is suggested to use this model as a pilot at the regional level.

Keywords: cancer, hospice, palliative care, health system, Iran

INTRODUCTION

Cancer is a chronic disease with a rising prevalence. According to the WHO reports in 2018, the incidence rate of cancer has reached 18.1 million cases globally. Furthermore, according to this report, the latest cancer-related mortality rate is almost 9.6 million cases worldwide (World Health Organization, 2018b). In Iran, the prevalence of cancer is increasing progressively and is known as the second leading cause of death. In 2016, 82% of deaths in Iran were related to chronic diseases, with cancer accounting for 16% of them (Bray et al., 2018).

Advancements in technology and pharmacotherapy has often led to an increased number of patients with chronic diseases including cancer, and the long-term survival of these patients, which will increase the need for end-of-life and death care (Kumar et al., 2017). More than half of cancer patients are hospitalized at the time of death and undergo highly invasive procedures that do not necessarily lead to better outcomes for the patient and family and may not even be consistent with their preferences, while these patients only need end-of-life care (Wright et al., 2016).

The lack of a formulated system for addressing the primary needs of these patients will result in unnecessary and frequent visits to the ERs of specialty and subspecialty centers, hospitalization in ICUs[1], burdening patients with expensive examinations and tests and, in some cases, taking unnecessary treatment measures (Amiresmaeili et al., 2015; Angus and Truog, 2016).

In addition, the lack of formal and structured training, insufficient knowledge, and experience in providing specific end-of-life care, the sense of providing futile care, mental pressures, and ethical problems and issues is among the challenges nurses and other members of the care team face while providing care for these patients in hospitals' Ers[2] and ICUs (Valiee et al., 2012; Borhani et al., 2014; Rassouli and Sajjadi, 2016). Family caregivers encounter many problems and are concerned about safe care, too (Nemati et al., 2018). Therefore, the hospital is not considered a proper place to provide end-of-life services.

In many communities, this type of care is provided by hospice centers and specialty end-of-life care teams (Clark et al., 2014). Hospice centers are defined as settings for providing out of hospital palliative care services or, in other words, at the community level, which are considered to be essential in some countries for addressing the needs of the people with reduced capacity who are at the end-of-life stage (Hui, 2014). Care services at hospice centers focus on the provision of the best possible quality of life for end-of-life patients and their families by preventing and relieving their physical, emotional, social, and spiritual suffering (National Hospice Palliative Care Organization, 2020). Despite this necessity, the available data shows large deficiencies in the provision of and access to such services in many low- and middle-income countries (World Health Organization, 2018a). The results of the studies conducted in Iran, as in other countries, show that critically ill and end-of-life patients are generally hospitalized in ICUs and extremely large sums are spent for them (Ziloochi et al., 2012; Amiresmaeili et al., 2015). There are no hospice centers in Iran, and the conducted research have focused on setting up and establishing centers specifically for providing end-of-life care and hospice services (Valiee et al., 2012; Borhani et al., 2014; Azami-Aghdash et al., 2015; Ansari et al., 2019).

According to National Action Plan for Prevention and Control of Non-Communicable Diseases, the plans developed for the next two decades should focus on the specific needs of those with the most common chronic diseases, especially cancer. As the demand for end-of-life care increases, patients and their families seek optimal care (Borhani et al., 2014). At the same time, considering the increased rate of chronic diseases, especially cancer, and the aging of the population, and based on the successful experiences of the developed countries in this aera, the designing and development of hospice services in Iran seems necessary. However, the provision of these services depends on various factors, including the cultural and social status of community (Azami-Aghdash et al., 2015). In this regard, a study, titled as *Developing a Hospice Care Delivery System for Iranian adult patients with cancer*, was conducted using a mixed qualitative-quantitative research approach. By adopting a mixed approach, the research team tried to design and propose a local model of hospice care provision system for the adult patients with cancer across the country. So, the present study Contains results of the third and fourth phases of the main study that was done with the following aims.

1. extracting and prioritizing the main areas of the model and the related indicators by consulting with experts (phase 3).
2. formulating the model and validating it in terms of importance, scientific acceptability, and feasibility.

CONCEPTUAL FRAMEWORK OF THE RESEARCH

The present study was designed and conducted based on the Hospice Palliative Care System Design Framework (2010). This framework is specifically developed by Ontario's Regional

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[1] Intensive Care Units (ICUs).

[2] Emergency Rooms (ERs).

End of Life Care Networks (EOLCN) and Hospice Palliative Care Networks (HPCN) for designing regional hospice care systems and evaluates 6 domains: (1) care settings and service, (2) programs within care settings and services, (3) integration/linkages, (4) human resources, (5) accountability, and (6) policies, guidelines, and funding (Seniors Health Research Transfer Network, 2012). In each domain, the requirements, and the standards for designing a hospice palliative care delivery system are discussed in detail. This framework describes and classifies the essential domains and elements of an integrated hospice palliative care system and shows the essential and favorable parts of each domain separately.

METHODOLOGY

This is a Health System Policy Research (HSPR) (Hojjat-Assari et al., 2021) conducted using a sequential mixed method in 4 phases from February 2018 to July 2020. **Figure 1** displays the phases of the study.

The First Phase

In this phase (from February to November 2019), a qualitative study was done using the directed content analysis to explain patients, families, caregivers, specialists, experts, and policymakers' perceptions of the care needs of cancer patients in the end-of-life stages, and the requirements for establishing a hospice care delivery system. The participants were selected through purposive sampling which continued until data saturation was achieved. Some of the samples were selected using snowball sampling. Data was collected using semi-structured interviews with open-ended questions. The results of this phase are also accepted for publication in the International Journal of Palliative Nursing. But not yet online.

The Second Phase

This phase (from December 2019 to April 2020) aimed to review and analyze the background and the *status quo* of hospice care delivery in Iran and tried to identify and compare the existing models of hospice care delivery for the adult patients with cancer across the world in the form of a comparative study. The hospice care systems of the United Kingdom, Canada, Australia, Japan, India, Jordan, and Iran were chosen for further investigation as target samples based on the reports by Economist Intelligence Unit (EIU). The search was conducted using keywords extracted from Medical Sub Headin (MeSH) in databases both national (SID, Magiran and Iran Medex) and international (Scopus, PubMed, Web of sciences, ProQuest, CINAHL, MedlinePlus, EMBASE, Cochrane Library, and Google Scholar), and prominent specialty journals in the field of hospice and palliative care, scientific and administrative documents, WHO website and reports, governmental websites and other official sites, important national and regional websites related to organizations active in the field of cancer, hospice and palliative care and the data provided by the websites of at least three hospice centers in each country were all reviewed in detail (Zarea et al., 2020).

The Third Phase

The purpose of this phase (from May to August 2020) is to combine the findings of the previous phases of the study, to extract the main components related to each domain for designing and developing a regional model of hospice care delivery system for adults with cancer in Iran, and to prioritize them. Therefore, after combining the results of the first and the second phases of the study, and identifying the main domains for model development, a questionnaire consisting of 127 items (components based on the main domains and subdomains of the model) was designed in seven parts. In fact, each part of the questionnaire was designed for each extracted domain from the results of qualitative and quantitative studies in the first and second phases. Each section contained several questions which were also based on the most important components of the model related to each domain. Given that seven domains were extracted from the first and second stages of the study, therefore, the questionnaire consisted of seven sections. Likert scale ranging from *very high* (4), *to low* (1) was used. The questionnaire provided to a group of experts for prioritization based on Delphi survey. The Delphi panelists were selected in this phase of the research, through purposive sampling and the snowball sampling, from among the health system policymakers and the physicians with competences in the field of cancer and palliative care, university faculty members and nursing professors with competences of teaching theoretical and practical courses related to cancer, and palliative care and other health care workers such as nurses, psychologists, occupational therapists, and nutritionists with more than 1 year experience of working with end-stage cancer patients. It should be noted that some of the specialists and experts were selected from among charity donors and the officials of NGOs, charitable organizations, and the Health Insurance Organization. In this study, according to the vast scope of the subject and the available resources as well as similar previous studies (Habibi et al., 2014), after the consent of 21 experts was obtained for participation in the research, the questionnaire *A Survey on the Importance of the Domains of Designing Hospice Care Delivery System for adult patients with cancer* were provided for them through in-person visits or *via* e-mail. The participants were asked to prioritize the importance of each item of the questionnaire based on their own perspective and competences, ranging from *very high* (4), *high* (3), *medium* (2), and *low* (1). Furthermore, there was an open-ended question at the end of the questionnaire, where they could express their other opinions in regard with the different parts of the questionnaire. The Decision Delphi was used in this study with a quantitative approach as well as the 10 steps proposed by Fawle (Rauch, 1979) to implement the Delphi survey. To this end, the data was analyzed in this phase using descriptive statistics through SPSS software, and the mean value and the standard deviation were calculated for the score of each item. Then, based on the approaches of similar studies (Ansari, 2018), items with a mean score of 3 and above were extracted. Also, to determine the degree of importance of each dimension, the standardized score of each dimension was calculated using the minimum and maximum scores of that dimension. By the end of this phase, a draft of

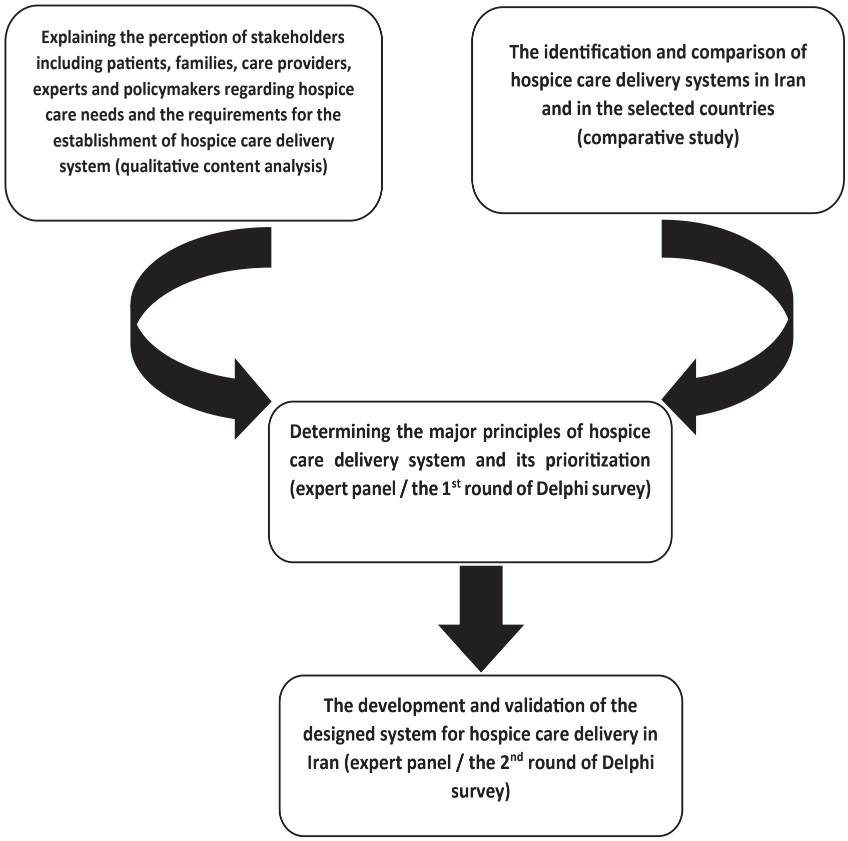

FIGURE 1 | Phases of the research.

the hospice care delivery system model for cancer patients was developed

The Fourth Phase

Finally, the Phase 4 (from September to December 2020)of the study was conducted again with the aim of assessing the credibility of the developed model in term of feasibility using the Delphi survey. It should be noted that this phase of the research is the 2nd round of Delphi survey. As a result, due to the participation of the same subjects in both stages, some previously explained characteristics of the panelists are not mentioned in this phase. For this purpose, after removing some items with a mean score below 3 in the previous phase, a 109-item questionnaire containing the 7 domains were developed and offered to the experts. The participants were asked to separately rate the feasibility of each of the components as *low* (1), *medium* (2) and *high* (3). The model of hospice care delivery system for cancer patients was then finalized, after receiving the comments and the suggestions and applying them. In this phase, SPSS and descriptive statistics were used to investigate how to answer the questionnaire items.

Ethical Consideration

This paper was a part of a PhD dissertation in nursing at the Chronic Disease Care Research Center affiliated to the Ahvaz Jundishapur University of Medical Sciences (grant No. NCRCCD9709). The ethics approval for the research was obtained from the Ahvaz Jundishapur University of Medical Sciences under the code of IR.AJUMS.REC.1397. 306.The authors have no conflicts of interest.

FINDINGS

The Results of Phase 1

At this stage, according to the research objectives and the inclusion criteria, 21 key experts including 3 cancer patients, 6 family caregivers, 8 caregivers, and 4 policymakers were interviewed. Finally, 7 main domains, 18 sub-domains, and 55 sub-subdomains were extracted from the analysis of the qualitative data. The results along with some more details are accepted to the International Journal of Palliative Nursing as a paper, and it will be published soon.

The Results of Phase 2

Based on the results of the comparative study, it was discovered that Iran does not have any integrated and formal structure or program for the provision of hospice services, and even end-of-life services. Of course, in recent years, few centers have provided sporadic palliative care services to patients with life-threatening conditions, especially those with cancer, in inpatient wards and homes, mainly in large cities, which contained end-of-life care. These services are provided without referring to clinical guidelines and solely based on caregivers' experiences and knowledge and are far from global standards. In addition, based on the comparison of hospice care system in the selected countries (United Kingdom, Canada, Australia, Japan, India, and Jordan), it can be said that successful and leading countries, despite the establishment of hospice centers, try to expand this type of service into all areas of the health system. In order to establish and develop hospice services in the above countries, important measures have been taken including having a strong and effective policymaking framework and comprehensive national programs, the integration of services into the palliative care system and, at a higher level, in the health system, government commitment to funding services, training specialized manpower, financial plans to help families and caregivers, adopting essential laws and regulations, the development of clinical protocols and guidelines, providing access to essential drugs and narcotics, increased public awareness and expanding research. The results of the first phase are already published in detail (Rassouli and Sajjadi, 2014, 2016; Mojen et al., 2017; Zarea et al., 2020).

The Results of Phase 3

The preliminary draft of the model for establishing hospice care delivery system was developed as a questionnaire. Out of 21 questionnaires distributed among the participants in Phase 3 (1st round of Delphi), 18 were fully completed and were then analyzed. The results showed that 10 men and 8 women with the mean age of 44.33 ± 15.67 years and 12.11 ± 9.89 years of work experience participated in the study. **Table 1** shows the personal characteristics of the participants in this phase.

After data analysis, the subjects obtained scores from 2.15 ± 0.87 to 4. Of these, the score of 18 questions were between 2.15 ± 0.87 and 2.91 ± 0.9, which were then excluded. Of these 18 questions, 4 questions were in the need to hospice center in home, hospital, outpatient clinics, and long -term care centers, 1 question was in the level of referral, 6 questions were about the specialist interdisciplinary team, 3 question about the skill and academic training/expected competencies, 1 question about the transparent decision making and 3 questions about the research development. Since the questionnaire contained a lot of details and was almost complete, most panelist did not have a specific comment for this part. In **Table 2** shows how the findings of the first phase (the comparative study) and the second phase (the qualitative study) are combined. Some components are only obtained from the findings of the comparative study; some, from the results of the qualitative study; and most of them, from both. The components were placed in a questionnaire. As well as the mean and standard deviation of the scores assigned by the experts has been reported. After removing the items with the mean scores below 3, 102 indicators remained. According to experts, "*Organizing Accountability,*" "*Integration into The Health System,*" "laying the groundwork *in the health system*" with a degree of importance of 93%, 88%, 85% respectively, are the most important domains in the design and deployment of the model. " *comprehensive care plan*" (83%), "*capacity building in the community*" (79.5%), "*the need to provide a variety of settings and services*" (79.2%), and "*specialized manpower*" (76%) were respectively in the next degree of importance.

The Model for Establishing Hospice Care Delivery System

Figure 2 show the proposed model for hospice care delivery system for adult patients with cancer and its dimensions that were adjusted to the upstream rules of Iranian health system.

TABLE 1 | The demographic characteristics of participants in Phase 3.

No	Gender	Age (years)	Working experience (years)	Specialty	Education
1	Male	45	12	Faculty member	PhD in nursing
2	Male	39	10	Instructor	BSN
3	Female	45	16	Faculty member	PhD in nursing
4	Male	30	7	Oncology nurse	MSc in nursing
5	Male	38	5	Palliative nurse	BSc in nursing
6	Female	35	3	Oncology psychologist	PhD in clinical psychology
7	Male	60	24	Faculty member	PhD in health policy
8	Male	50	15	Surgical oncologist	Specialist
9	Male	55	20	Oncologist	Specialist
10	Female	42	13	Faculty member	PhD in nursing
11	Female	50	18	General practitioner	MD
12	Male	46	10	General practitioner	MD
13	Female	48	13	Public relations postgraduate	Social worker
14	Female	28	6	Spiritual therapist	PhD in theology
15	Male	55	23	Faculty member	PhD in health economics
16	Male	41	10	Epidemiologist	MD
17	Female	48	5	General practitioner	MD
18	Female	38	8	Lymphatic therapist	MD

TABLE 2 | The main domains and components extracted from the findings of Phase 1 (comparative study) and Phase 2 (qualitative study) and the mean, minimum and maximum score of components in Phase 3 (minimum mean score = 1, maximum mean score = 4).

Domains	Subdomains	Components	Based on		Mean ± SD
			Comparative study	Qualitative study	
The need to provide a variety of settings and services	The need for hospice centers	The establishment of hospice centers	*	*	3.9 ± 0.85
		Services provision by hospice centers at home	*		2.91 ± 0.9
		Services provision by hospice centers at hospital	*		2.15 ± 0.87
		Services provision by hospice centers at outpatient clinics	*		2.91 ± 0.9
		Services provision by hospice centers at long-term care centers	*		2.15 ± 0.87
	Comprehensive patient/family centered services	Managing physical symptoms of the end stage patient	*	*	4
		Managing mental symptoms of the end stage patient	*	*	4
		Providing spiritual care to the end-stage patient and family caregivers	*	*	3.3 ± 0.75
		Addressing the mental-spiritual needs of the family until the end-stage patient passes away	*	*	3.8 ± 0.75
		Addressing the social needs of the patient and family until the end-stage patient passes away	*	*	3.3 ± 0.75
		Providing bereavement care to the family after patient's death	*	*	3.5 ± 0.7
		The training of end-stage patient and family caregivers	*	*	3.9 ± 0.87
		Offering the necessary information to the end-stage patient and family caregivers	*	*	4
		Considering the care ability of family caregivers while entrusting the end-stage patient to them		*	3.6 ± 0.85
Comprehensive care program	Participatory decision-making	End-stage patient's involvement in decision making regarding the place of death and care receiving	*		3.2 ± 0.64
		Family's involvement in decision making regarding the place of death and end-stage patient's care receiving	*		3.3 ± 0.81
		Respecting end-stage patient's right to know his/her own state	*	*	3.8 ± 0.78
		Respecting family's right to know the state of the end-stage patient	*	*	4
		Supporting end-stage patient in making his/her decision regarding self	*	*	4
		Supporting family in making decision regarding the end-stage patient	*	*	4
		Attention to physician's decision at patient's end-of-life stage	*	*	3.3 ± 0.75
		Attention to the end-stage patient's religious-cultural issues		*	3.7 ± 0.81
		Attention to the religious-cultural issues of the family caregivers of end-stage patient		*	3.7 ± 0.81
	Consensus on the definition of the procedure	The need to define hospice care according to the WHO's definition		*	3.3 ± 0.75
		The need to set a standard referral time for receiving hospice care	*	*	3.9 ± 0.8
		Setting the duration of hospice service delivery	*		3.8 ± 0.78
		Setting precise admission criteria for receiving hospice services	*	*	3.8 ± 0.78
		Comprehensive assessment of end-stage patient's needs	*	*	3.7 ± 0.78
		Comprehensive assessment of the needs of the family with end-stage patient	*	*	3.6 ± 0.80
		Developing a care plan based on the end-stage patients and family	*	*	3.5 ± 0.78
		Precise explanation of hospice care delivery procedure (assessment, follow up, visits, etc.)	*	*	4
		24/7 phone consultation to the family of end stage patient regarding care	*		3.5 ± 0.76
		Establishing communication between family and hospice care team during patient's life and after death	*	*	3.6 ± 0.85
		Formulating guidelines for referral and transfer between different settings (hospital, home, etc.) if necessary	*	*	4
		Formulating visitation guidelines in the care plan at various settings	*	*	3.5 ± 0.78
		Formulating standard diagnostic and treatment guidelines	*	*	3.5 ± 0.78

(Continued)

TABLE 2 | (Continued)

Domains	Subdomains	Components	Based on		Mean ± SD
			Comparative study	**Qualitative study**	
		Formulating standard guideline for pain management in end-stage patient	*	*	4
		Formulating standard guideline for symptom management in end-stage patient	*	*	4
		Assessing the effectiveness of the training of family caregivers	*		3.30 ± 0.80
		Care quality assessment	*		3.5 ± 0.78
Integration into the health system	Integration of services	The integration of hospice care services at the primary level (outpatient clinics, urban health centers and comprehensive health centers)	*	*	3.5 ± 0.75
		The integration of hospice care services at the secondary level (general and specialty hospitals)	*	*	3.8 ± 0.81
		The integration of hospice care services at the tertiary level (home, long-term care centers)	*	*	3.4 ± 0.75
	Level of referral	Patient's referral by a family physician for receiving hospice services	*	*	3.5 ± 0.75
		Patient's referral by a nurse for receiving hospice services	*	*	3.8 ± 0.81
		Patient's referral by a general practitioner for receiving hospice services	*		3.7 ± 0.78
		Patient's referral by an oncologist for receiving hospice services	*	*	4
		Patient's referral by other member of the care team for receiving hospice services	*		2.9 ± 0.9
	Connection with other care settings	Establishing connection between hospice centers and other care settings (outpatient clinics and urban health centers, hospitals, and home)	*	*	3.8 ± 0.81
		Service coordination among different levels	*	*	3.9 ± 0.8
		Providing hospice centers with access to care and treatment documents	*	*	4
		Creating a national hospice care network to record the data of patients receiving care	*		3.5 ± 0.75
		The connection of hospice care national network to the electronic health record system (SEPAS)		*	3.9 ± 0.8
Specialist manpower	Specialist interdisciplinary team	Providing interdisciplinary and team care	*	*	4
		Training specialist hospice care delivery teams	*	*	4
		The presence of a pain specialist in the hospice care delivery team	*		2.9 ± 0.68
		The presence of a specialist nurse in the hospice care delivery team	*	*	4
		The presence of a nurse as a coordinator of team care	*		4
		The presence of a psychologist in the hospice care delivery team	*	*	4
		The presence of a social worker in the hospice care delivery team	*	*	4
		The presence of a nutritionist in the hospice care delivery team	*	*	2.9 ± 0.63
		The presence of a spiritual caregiver/cleric in the hospice care delivery team	*	*	4
		The presence of a physiotherapist in the hospice care delivery team	*	*	2.9 ± 0.63
		The presence of an occupational therapist in the hospice care delivery team	*		2.3 ± 0.79
		The presence of a general practitioner in the hospice care delivery team	*	*	3.9 ± 0.81
		The presence of an oncologist in the hospice care delivery team	*	*	2.9 ± 0.60
		The presence of volunteer workforce in the hospice care delivery team	*		2.9 ± 0.60
	skill and academic training/Expected competencies	Including the concept of hospice care in the curriculums of undergraduate nursing program and general practice	*	*	3.9 ± 0.81
		Including the concept of hospice care in the curriculums of the undergraduate programs in other related disciplines	*	*	3.9 ± 0.78
		The need to develop hospice care specialized discipline	*	*	2.9 ± 0.63
		The need to develop hospice care fellowship course	*	*	2.9 ± 0.63
		The need to develop a master program for hospice care	*	*	2.7 ± 0.68
		Holding hospice care short-term skill training courses	*	*	4
		Holding ongoing training courses for the staff	*	*	3.9 ± 0.81
		Holding periodic hospice care workshops	*	*	4
		Providing the necessary educational settings for training specialist manpower	*	*	4
		Providing standard educational content for training specialist manpower	*	*	4

(Continued)

TABLE 2 | (Continued)

Domains	Subdomains	Components	Based on		Mean ± SD
			Comparative study	Qualitative study	
		The involvement of related organizations in hospice care training	*	*	3.8 ± 0.78
		Defining credible specialized licenses and certificates for the trained manpower	*	*	3.9 ± 0.81
		Defining the competencies expected from the members of the hospice care team	*	*	3.8 ± 0.78
		Educational planning based on the competencies expected from the members of the hospice care team	*		3.8 ± 0.78
		Considering the topics related to hospice care in professional competence exams of nurses and physicians and other disciplines	*		3.2 ± 0.64
Organizing the accountability system	Transparent decision-making	Developing the hospice care delivery system for various types of adult cancer	*		2.9 ± 0.63
		Creating a joint taskforce for hospice care policymaking at the level of the Ministry of Health	*	*	3.9 ± 0.81
		Creating a joint taskforce for hospice care at the university level	*	*	3.9 ± 0.81
		Creating a joint taskforce for hospice care at the level of specialty hospitals	*	*	3.7 ± 0.68
		The participation of all sectors involved in the development of hospice care system	*	*	4
		The participation of governmental organizations in the development of hospice care system	*	*	4
		The participation of private organizations (charities, NGOs, etc.) in the development of hospice care system	*	*	3.9 ± 0.81
		The formulation of job descriptions, roles, responsibilities, and jurisdiction of each sector	*	*	3.8 ± 0.78
	Accountable management	The presence of a system for monitoring, assessment, and supervision	*	*	3.8 ± 0.78
		Developing indicators for monitoring, supervision, and accreditation	*	*	3.9 ± 0.81
		Launching the electronic document system and the access to database to make decisions in the field of policymaking	*	*	3.9 ± 0.81
Laying the groundwork *in the health system*	Optimal policymaking	Providing documents related to the importance of hospice care for health policymakers	*	*	3.9 ± 0.81
		The obligation of government and the Ministry of Health to implement hospice care plan	*	*	3.4 ± 0.75
		Developing the national hospice care plan	*		3.8 ± 0.78
	Financial prerequisites	Government funding	*	*	4
		Funding by private organizations	*	*	3.9 ± 0.81
		Developing insurance service packages	*	*	4
		The involvement of charitable organizations in funding hospice services	*	*	3.9 ± 0.81
		Cost determination for secondary insurance services	*	*	3.7 ± 0.68
		Cost determination for governmental insurance services	*	*	4
		Cost determination for services separately for each discipline	*	*	3.8 ± 0.81
		Supporting private sector in service provision	*	*	3.9 ± 0.80
		Insurance supportive policies for the proper insurance coverage of medications	*	*	4
	Structural prerequisites	Providing infrastructure (resources, space, etc.)	*	*	3.9 ± 0.81
		Providing care equipment (ventilator, suction machine, oxygen, etc.)	*	*	4
		Developing palliative care network (service delivery at outpatient clinics, home-care centers, hospital, etc.)	*	*	3.8 ± 0.78
		Access to center data registry	*	*	3.4 ± 0.75
	Legal prerequisites	Access to a variety of necessary narcotic drugs	*	*	4
		Formulating guidelines for access to necessary narcotic drugs	*	*	4
		Formulating drug packages at hospice centers	*	*	3.9 ± 0.81
		The possibility for other disciplines such as nursing, pharmacy, etc. to prescribe drugs	*		3.4 ± 0.75
		Solving legal issues surrounding drug prescription (which drugs by whom?) at hospice centers	*	*	4

(Continued)

TABLE 2 | (Continued)

Domains	Subdomains	Components	Based on		Mean ± SD
			Comparative study	Qualitative study	
		Developing monitoring and supervisory indicators for supervision on and monitoring of pharmacotherapy at hospice centers	*		3.6 ± 0.73
		Formulating guidelines and instructions in accordance with the law, ethics, and scientific references in regard with continuing or stopping CPR	*	*	3.8 ± 0.81
		Determining the legal procedure related to hospice care	*	*	4
		Clarifying legal issues regarding hospice care	*	*	4
		Formulating supervisory rules for care at hospice centers	*	*	4
		Formulating supportive rule for care providers at hospice centers	*	*	4
Capacity building at the community	Raising public awareness	Holding training courses at the community level	*	*	4
		Providing the cultural and social infrastructure with the aim of increasing hospice care acceptability	*	*	4
	Research development	Launching interdisciplinary research centers related to hospice care	*		2.9 ± 0.63
		Publishing specific journals in the field of cancer hospice care	*		2.9 ± 0.63
		The participation of other organizations in conducting research on hospice care	*		2.7 ± 0.68
		Holding annual hospice care seminars and conferences	*		3.2 ± 0.63
		Determining the cost-effectiveness of hospice services	*	*	4

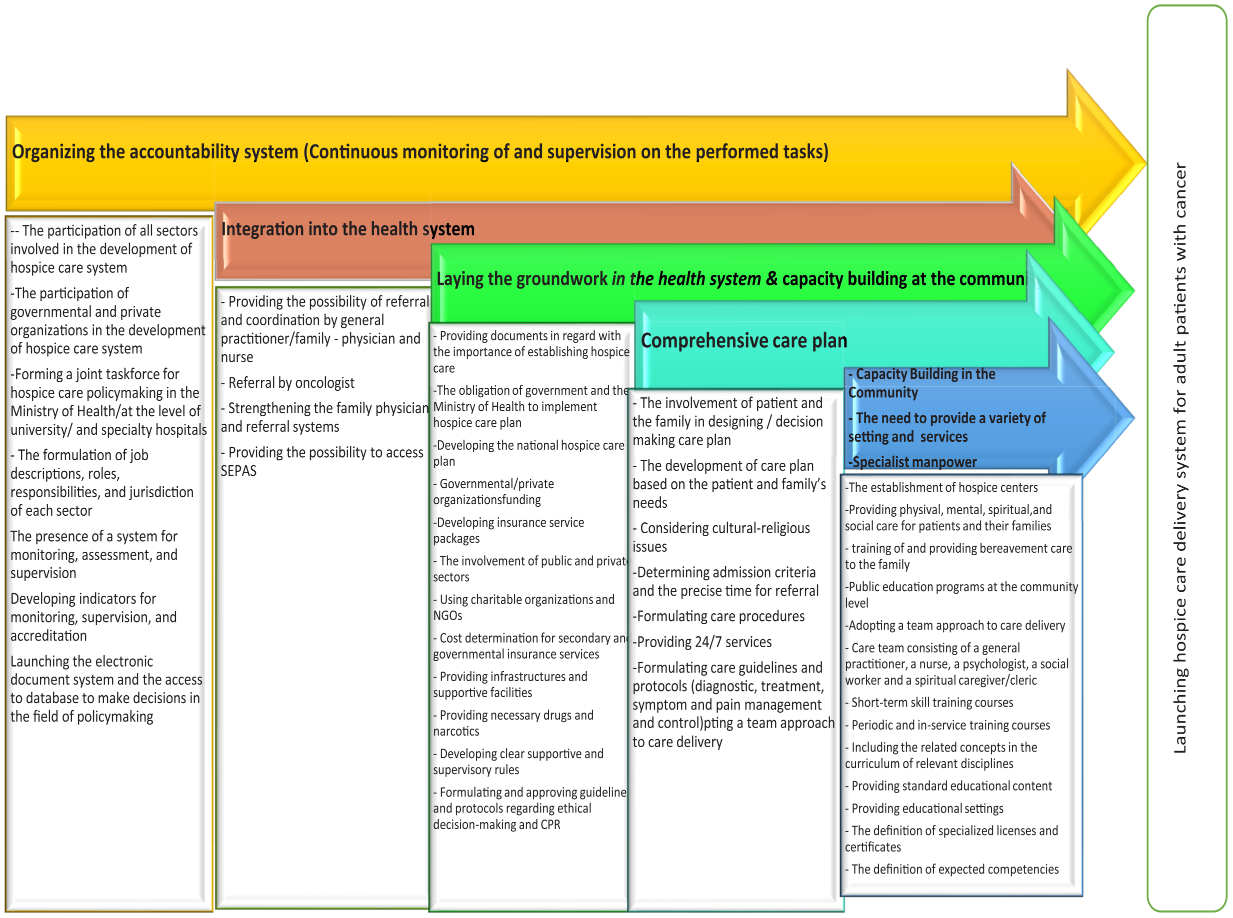

FIGURE 2 | The model for the establishment of hospice care delivery system for adult patients with cancer.

TABLE 3 | The demographic characteristics of participants in Phase 4.

No	Gender	Age (years)	Working experience (years)	Specialty	Education
1	Male	45	12	Faculty member	PhD in nursing
2	Male	39	10	Instructor	BSN
3	Female	45	16	Faculty member	PhD in nursing
4	Female	30	7	Oncology nurse	MSc in nursing
5	Female	38	5	Palliative nurse	MSc in nursing
6	Male	55	23	Faculty member	PhD in health policy
7	Male	50	15	Surgical oncologist	Specialist
8	Male	55	20	Oncologist	Specialist
9	Female	42	13	Faculty member	PhD in nursing
10	Female	50	18	General practitioner	MD
11	Male	46	10	General practitioner	MD
12	Male	55	23	Faculty member	PhD in health economics

Dimensions are arranged according to the degree of importance in the opinion of experts. In addition, inside each box, there are components (activities) that are related to that dimension and are necessary for the establishment of a hospice care system.

The Results of Phase 4

Out of the 18 questionnaires submitted in the 2nd round of Delphi, only 12 complete questionnaires were received. **Table 3** shows the demographic characteristics of the participants.

The statistical results of the components in this phase of the research are shown in **Table 4**. Panelists did not have a specific comment for this part too. In general, the results show only the low feasibility of model in some components.

Accordingly, the practical process of establishing a hospice care delivery system for adult cancer patients were designed and approved by the research team (**Table 5**).

DISCUSSION

In this study a model of hospice care delivery system for Iranian adult patients with cancer was developed and validated. According to the proposed model, only the establishment of hospice centers and care provision in these centers was prioritized. In regard with services provision, the management of patients' physical and mental symptoms and offering the necessary information to the patient and the family are important and have high feasibility.

Consistent with the finding of this study, The Medicare Hospice Benefit program was established in 1982 and covers hospice care at home or inpatient facilities. According to the Dartmouth Atlas Project, 63% of patients with cancer enrolled in hospice before death in 2012 Dartmouth (Goodman et al., 2013). It should be noted that in Iran, even in the field of palliative care, services are more hospital-based, and home care centers are less developed due to the existing challenges (Mojen et al., 2017). On the other hand, given the current cultural background, most families seek care in inpatient centers (Valiee et al., 2012; Borhani et al., 2014; Heidari et al., 2018). In addition, recent Iranian studies on palliative care have focused on the need to launch various structures for providing care services, including hospice centers (Ansari et al., 2018; Khanali Mojen, 2018).

Based on our model, it is important in addition to manage physical, mental, spiritual, and social symptoms of the patient, provide mental, spiritual and social support for the family, and offer bereavement care. Like the model designed in this study, based a designed Season's Community-Based Palliative care model In Western North Carolina, an inter-disciplinary team provided care to patients in both outpatient and inpatient settings, including psychosocial/spiritual care, advanced care planning, symptom management, and patient/family education. According to developed model, spiritual support is also one of the domains of comprehensive care in hospice centers and has a high feasibility. Kang (2018) stated based on the study of different models, spiritual care is one of the core components of hospice palliative care. The literature has supplied numerous evidence-based models (such as quality of life, coping, the spiritual-relational view, . . .) that focus on spiritual care and can be used as models for providing spiritual care in these centers. The above results confirm the results of the present study (Kang, 2018).

Obviously, the need for bereavement care is a priority which has high feasibility in this model, too. Consistent with this finding, bereavement care is an essential component of hospice care that includes anticipating grief reactions and providing ongoing support for the bereaved over a period of 13 months. While the terms are often used interchangeably, bereavement refers to the state of loss, and grief is the reaction to loss (Grant et al., 2020). The results of the previous Iranian studies on palliative care also confirm this finding (Khanali Mojen, 2018; Pakseresht et al., 2018).Based on the designed model in the domain *comprehensive care plan*, participatory decision-making and consensus in the definition, have priority. These patients need to receive comprehensive and standard care for their quality of life to improve during their remaining days. Providing this type of care requires the development of guidelines and instructions that are part of the palliative care delivery process. Highlighting the structures and the process of care is very important. In addition, the development of care guidelines and standards, and service packages tailored to the needs of the family is of great significance (Eshaghian-dorcheh et al., 2020).

TABLE 4 | The mean, minimum and maximum score of the Feasibility of components in Phase 4 (minimum mean score = 1, maximum mean score = 4).

Domains	Subdomains	Components	Feasibility (Mean ± SD)
The need to provide a variety of settings and services	The need for hospice centers	The establishment of hospice centers	2.91 ± 0.31
	Comprehensive patient/family centered services	Managing physical symptoms of the end stage patient	2.91 ± 0.31
		Managing mental symptoms of the end stage patient	2.91 ± 0.31
		Providing spiritual care to the end-stage patient and family caregivers	2.91 ± 0.31
		Addressing the mental-spiritual needs of the family until the end-stage patient passes away	2.85 ± 0.60
		Addressing the social needs of the patient and family until the end-stage patient passes away	2.42 ± 0.40
		Providing bereavement care to the family after patient's death	2.85 ± 0.63
		The training of end-stage patient and family caregivers	2.85 ± 0.63
		Offering the necessary information to the end-stage patient and family caregivers	2.89 ± 0.70
		Considering the care ability of family caregivers while entrusting the end-stage patient to them	2.89 ± 0.70
Comprehensive care program	Participatory decision-making	End-stage patient's involvement in decision making	2.73 ± 0.50
		Family's involvement in decision making	2.85 ± 0.60
		Developing care based on patient's needs	2.85 ± 0.80
		Developing care based on the family's needs	2.89 ± 0.70
		Considering patient and family's cultural issues	2.81 ± 0.78
	Consensus on the definition of the procedure	The definition of hospice care according to the WHO's definition	2.55 ± 0.60
		Setting a standard referral time for receiving hospice care	2.89 ± 0.70
		Setting the duration of hospice service delivery	2.73 ± 0.50
		Setting precise admission criteria for receiving hospice services	3
		Comprehensive assessment of the patient and family's needs	2.85 ± 0.60
		Developing a care plan based on the needs of end-stage patients and family	2.58 ± 0.80
		Precise explanation of hospice care delivery procedure (assessment, follow up, visits, etc.)	2.89 ± 0.70
		24/7 phone consultation and services	2.35 ± 0.40
		Formulating visitation guidelines in the care plan	3
		Formulating guidelines for admission, referral, and transfer between different settings (hospital, home, etc.)	3
		Formulating standard diagnostic and treatment guidelines	3
		Formulating standard guideline for pain management	3
		Formulating standard guideline for symptom management	3
		Establishing communication between family and hospice care team during patient's life and after death	2.89 ± 0.70
		Identifying the stages of family bereavement and providing a care plan	2.73 ± 0.50
		Assessing the effectiveness of the training of family caregivers	2.58 ± 0.80
		Care quality assessment	2.42 ± 0.80
Integration into the health system	Integration of services	The integration of hospice care services at the primary level (outpatient clinics, urban health centers and comprehensive health centers)	2.34 ± 0.63
		The integration of hospice care services at the secondary level (general and specialty hospitals)	2.91 ± 0.31
		The integration of hospice care services at the tertiary level (home, long-term care centers)	2.91 ± 0.31
	Level of referral	Patient's referral by a family physician/general practitioner for receiving hospice services	2.64 ± 0.84
		Patient's referral by a nurse for receiving hospice services	2.56 ± 0.84

(Continued)

TABLE 4 | (Continued)

Domains	Subdomains	Components	Feasibility (Mean ± SD)
		Patient's referral by an oncologist for receiving hospice services	2.58 ± 0.60
	Connection with other care settings	Establishing connection between hospice centers and other care settings (outpatient clinics and urban health centers, hospitals, and home)	2.73 ± 0.50
		Service coordination among different levels	2.70 ± 0.63
		Providing hospice centers with access to care and treatment documents	2.81 ± 0.78
		Creating a national hospice care network to record the data of patients receiving care	2.56 ± 0.84
		The connection of hospice care national network to the electronic health record system (SEPAS)	2.48 ± 0.78
Specialist manpower	Specialist interdisciplinary team	Providing interdisciplinary and team care	2.73 ± 0.50
		Training specialist hospice care delivery teams	2.58 ± 0.60
		The presence of a pain specialist in the hospice care delivery ream	2.73 ± 0.50
		The presence of a specialist nurse in the hospice care delivery team	2.58 ± 0.60
		The presence of a nurse as a coordinator of team care	2.91 ± 0.31
		The presence of a psychologist in the hospice care delivery team	2.73 ± 0.50
		The presence of a social worker in the hospice care delivery team	2.56 ± 0.84
		The presence of a spiritual caregiver/cleric in the hospice care delivery team	2.81 ± 0.78
		The presence of a general practitioner in the hospice care delivery team	2.91 ± 0.31
	skill and academic training/Expected competencies	Including the concept of hospice care in the curriculums of undergraduate nursing program and general practice	2.91 ± 0.31
		Including the concept of hospice care in the curriculums of the undergraduate programs in other related disciplines	2.56 ± 0.84
		Holding hospice care short-term skill training courses	3
		Holding ongoing training courses for the staff	2.73 ± 0.50
		Holding periodic hospice care workshops	2.46 ± 0.38
		Providing the necessary educational settings for training specialist manpower	2.64 ± 0.84
		Providing standard educational content for training specialist manpower	2.58 ± 0.60
		The involvement of related organizations in hospice care training	2.56 ± 0.84
		Defining credible specialized licenses and certificates for the trained manpower	2.58 ± 0.60
		Defining the competencies expected from the members of the hospice care team	2.81 ± 0.78
		Educational planning based on the competencies expected from the members of the hospice care team	2.58 ± 0.60
		Considering the topics related to hospice care in professional competence exams of nurses and physicians and other disciplines	2.56 ± 0.84
Organizing the accountability system	Transparent decision-making	Creating a joint taskforce for hospice care policymaking at the level of the Ministry of Health	2.73 ± 0.50
		Creating a joint taskforce for hospice care at the university level	2.91 ± 0.31
		Creating a joint taskforce for hospice care at the level of specialty hospitals	2.58 ± 0.60
		The participation of all sectors involved in the development of hospice care system	3
		The participation of governmental organizations in the development of hospice care system	2.73 ± 0.50
		The participation of private organizations (charities, NGOs, etc.) in the development of hospice care system	2.81 ± 0.78
		The formulation of job descriptions, roles, responsibilities and jurisdiction of each sector	3
	Accountable management	The presence of a system for monitoring, assessment and supervision	2.46 ± 0.38
		Developing indicators for monitoring, supervision and accreditation	2.46 ± 0.38
		Launching the electronic document system and the access to database to make decisions in the field of policymaking	2.56 ± 0.84

(Continued)

TABLE 4 | (Continued)

Domains	Subdomains	Components	Feasibility (Mean ± SD)
Laying the groundwork *in the health system*	Optimal policymaking	Providing documents related to the importance of hospice care for health policymakers	2.91 ± 0.31
		Developing the national hospice care plan	2.85 ± 0.60
	Financial prerequisites	Government funding	2.68 ± 0.51
		Funding by private organizations	2.73 ± 0.50
		Developing insurance service packages	2.68 ± 0.51
		The involvement of charitable organizations in funding hospice services	2.73 ± 0.50
		Cost determination for secondary insurance services	2.34 ± 0.61
		Cost determination for governmental insurance services	2.68 ± 0.51
		Cost determination for services separately for each discipline	2.15 ± 0.43
		Supporting private sector in service provision	2.56 ± 0.84
		Insurance supportive policies for the proper insurance coverage of medications	2.73 ± 0.50
		Determining the cost-effectiveness of hospice services	2.91 ± 0.31
	Structural prerequisites	Providing infrastructure (resources, space, etc.)	2.83 ± 0.41
		Providing care equipment (ventilator, suction machine, oxygen, etc.)	2.85 ± 0.60
		Making structural changes in the health system	2.34 ± 0.67
		Access to cancer data registry	2.46 ± 0.38
	Legal prerequisites	Access to a variety of necessary narcotic drugs	2.68 ± 0.51
		Formulating guidelines for access to narcotics and necessary drugs	3
		The possibility for other disciplines such as nursing, pharmacy, etc. to prescribe drugs	2.34 ± 0.67
		Clarifying legal issues regarding hospice care	3
		Determining the legal steps regarding death at hospice centers	3
		Formulating guidelines and instructions in accordance with the law, ethics and scientific references in regard with continuing or stopping CPR	3
		Solving legal issues surrounding drug prescription (which drugs by whom?)	3
Capacity building at the community	Raising public awareness	Holding training courses at the community level	2.68 ± 0.51
		Providing the cultural and social infrastructure with the aim of increasing hospice care acceptability	3
	Research development	Holding annual hospice care seminars and conferences	2.85 ± 0.60

In the developed model, the integration of services at the primary, secondary and the tertiary levels of the health system was feasible. In addition, according to the designed model, hospice centers will be connected with other care settings. In line with the above-mentioned, it should be noted that the studies have shown that the palliative care and hospice services are provided mainly at the primary health care (PHC) (Zarea et al., 2020). As suggested by WHO, providing palliative care at the PHC level is mandatory, in line with UHC goals (World Health Organization, 2018a). Therefore, the integration of services into this level of care is essential in developing a hospice care delivery system. Consistent with this study, Hojjat-Assari's model, palliative care is provided at various levels of the health system (Hojjat-Assari et al., 2021).

Furthermore, in this model patients will be referred by oncologists, general practitioners, family physicians, and nurses. As you see, the patient referral process in this model is reversed so that sources of referral are central and specialist units and general hospitals. These findings are in line with model, and Khanali's model in which patients are referred from specialist centers to the palliative care clinics (Mojen et al., 2017). It seems that the specialty-oriented culture dominant in the Iranian society and the patient's visiting specialists even for the most minor issues, the public and caregivers' insufficient knowledge and awareness, palliative service delivery's being a novel care approach, the shortage or lack of different settings which offer this type of service, easier access to specialized services and more facilities in the secondary and tertiary levels of the health system in Iran, as well as the above mentioned items are the main reasons for which the experts have allocated higher scores to the indicator *patients' referral by oncologists to receive hospice services*. Referral by an oncologist may be effective in providing the infrastructures for the establishment of a hospice care system and a starting point for the provision of this type of service at the community.

In the developed model, it was also determined that the hospice care team should at least include a general practitioner, a trained nurse, and a spiritual caregiver/cleric. Psychologists and social workers came next. These results are in line with WHO guidelines (World Health Organization, 2016). Furthermore, the spiritual caregiver is a key member of the hospice care team according to the cultural-religious background of the country.

TABLE 5 | The operational stages of establishing hospice care delivery system for adult patients with cancer.

At the level of the Ministry of Health and Medical Education

- Providing documents regarding the importance of establishing hospice care system and presenting it to the policymakers
- Forming a joint taskforce for hospice care policymaking in the Ministry of Health
- The development of national program and its general executive policies
- The development of care service packages in hospice centers
- The insurance coverage of hospice services, the necessary drugs and narcotics
- Developing and approving guidelines and protocols for ethical decision-making < resuscitation at hospice centers and pain and symptom management
- Defining the documents, the required specialized licenses and expected competencies for each discipline to provide services at hospice centers
- Developing and approving supervisory rules
- Communicating the national plan, executive policies, service packages, the formulated guidelines and protocols and the supervisory rules to medical universities and insurance organizations

At the level of medical universities

- Forming a joint taskforce for the establishment of hospice care system at the level of medical universities
- Demographic needs assessment at the provincial level
- Cost estimation and conduction of cost-effectiveness studies
- Involving private sector, charitable organizations and NGOs
- Formulation of hob descriptions and the jurisdiction of each sector
- Allocating physical (space, equipment and medicine), financial and human resources
- Forming hospice care delivery teams
- The development and implementation of short-term skill training programs
- Launching hospice centers (providing training and care delivery settings)
- Developing and implementing programs at the community level to raise public awareness
- Providing hospice centers and other service delivery settings with the access to SEPAS
- Developing and communicating executive instructions (the referrer, the type of referral, transfer and admission at each level) to comprehensive health centers, long-term care centers, hospitals and educational and treatment centers affiliated to the university
- Developing and communicating instructions on how to supply narcotics
- Strategies for strengthening the family physician system and the referral system
- Developing standard educational content and the integration of concepts into educational curriculums
- Monitoring care provision at hospice centers

At the level of comprehensive health centers, long-term care centers, hospitals and educational and medical centers

- Forming a hospice care committee
- Holding periodic and in-service courses of hospice care Offering 24/7 services
- Implementing programs to raise public awareness
- The implementation of communicated instructions regarding the establishment of hospice care delivery system
- Conducting research and assessing the effectiveness of the provided services Providing feedback

At the level of hospice centers

- Providing 24/7 services and support to patients and families
- Identifying the needs of the patient and family in all aspects through standard methods
- Pain control and managing physical, mental, social and spiritual needs of the patients
- Family management in mental, social and spiritual dimensions
- The training and empowerment of the family Providing bereavement services
- Formal communication with health service providers to identify patients in need of hospice care
- Training service providers at hospice centers and other health service delivery settings
- Public education at the community level Recording data, documentation and having access to SEPAS for sharing information
- Conducting research and evaluating the provided services
- Communication with policymaking taskforces at the university level
- Developing plans for service development

Contrary to the results of other studies in the field of palliative care in Iran, using volunteer forces for the provision of hospice services was not a priority. It should be noted that volunteers and charity donors are active members of the palliative care system in many countries around the world, and the provision of many services, especially hospice care, depends on their participation (Hunter and Orlovic, 2018). In Iran, most volunteers work in charities and in the field of community services, especially psychological and financial support (Ansari et al., 2018).

In regard with academic education and the expected skills and competencies, according to the experts, the development of the specialized discipline, the fellowship course, and master's degree program for hospice care are not mandatory. *Holding short-term hospice care skill training courses* to provide specialist manpower obtained higher feasibility, which is in line with the policies adopted in other countries around the world and even in other Middle Eastern countries (Barasteh et al., 2020).

On ***organizing the accountability system***, In the developed model, *the formation of a hospice care policymaking taskforce at the university level*, *the formation of a hospice care committee at the hospital level*, *the participation of all the sectors involved in hospice care delivery, including private and public sectors,* and *the formulation of job descriptions, roles, responsibilities and the jurisdiction of each department* were of great feasibility. In line with our model, Ansari's palliative care model in Iran also shows that the formation of executive committees at various levels of the

Ministry of Health, including the Deputy Minister of Education, universities, and hospitals will play an important role in the development of palliative care services (Ansari et al., 2019).

Laying the groundwork *in the health system* in our developed model is possible in the following ways. *Providing documents in regard with the importance of hospice care for health policymakers* and *developing a national hospice care plan,* which is regarded as the official starting point for the provision of these services in many communities (Zarea et al., 2020). Another important point in the designed model is providing financial prerequisites through various funding approaches including public funding, private funding, insurance coverage, development of service packages and setting service costs, as well as supporting charitable organizations. However, funding through the private sector and seeking support from donors seems to be more important. Privatization in the health system can lead to positive outcomes. More funding and investments in the private sector, flexibility, and the freedom of managers in utilizing and making better use of the available funds will enable them to perform more activities with more diversity to increase patients and families' satisfaction, as well as providing higher quality services (Eshaghian-dorcheh et al., 2020). Since governments alone cannot address the growing needs of cancer patients, charities and volunteers can be of great help to the government and patients at the community level, provided that all the activities are monitored and organized based on the set rules and regulations (Groeneveld et al., 2017).

In the present model, making plans to meet the structural prerequisites of care delivery in hospice centers, including physical space and supportive care equipment, is considered a priority and has high feasibility. This factor is one of the principles of designing every health care system, which is line with the results of other studies (Rassouli and Sajjadi, 2016; Barasteh et al., 2020).

Based on this model, the greatest need in regard with the legal and ethical requirements of care in hospice centers is the development of guidelines for administering narcotics and necessary medications and clarifying legal issues related to care, drug prescription, death, and resuscitation operations in these centers. Developed systems of palliative, hospice and end-of-life care around the world have clear rules regarding euthanasia, DNR, and decision making based on patients' interests (Economist Intelligence Unit, 2015).

In regard with research and public awareness, there is an evident need for providing cultural and social infrastructures to increase the acceptance of hospice care and conduct cost-effectiveness studies, which has the highest level of feasibility based on our model. Given the growing rate of research in the field of palliative care in Iran in recent years, and cancer-related research centers and journals' prioritizing the publication of articles in the field of palliative care, it is important to create proper infrastructures in the existing centers, determine research priorities in this area, and finance further research (Rassouli and Sajjadi, 2014).

Due to the limited number of palliative care experts in Iran, there were few knowledgeable individuals participating in the surveys. On the other hand, cultural, social, and religious factors are reported as underlying factors in the development of the care system. However, given that Iran has a wide cultural diversity, this issue may limit the generalization of the research findings. Therefore, its pilot implementation in different regions is recommended.

CONCLUSION

The aim of this study was to develop and validate a model for establishing a hospice care delivery system for the adult patients with cancer in Iran. Based on the developed model, it was discovered that the Iranian health system needs to launch hospice centers and provide comprehensive services to cancer patients and their families. It is also necessary to design a comprehensive care plan and train specialist manpower in order to address this need. In addition, the integration of these services into the health system and organizing the accountability system before the establishment of this type of services is an inevitable necessity. To this end, policy making through providing the infrastructure in the health system, and capacity-building at the community is also of great importance. Therefore, in order to implement the developed model in Iran, it is necessary to consider the following items: creating job titles in regard with these services especially for physicians and nurses, implementing the program with the presence of community health nurses and family physicians based on the referral process according to the similar and approved instructions, formulating clinical guidelines and the necessity of its implementation at all levels, encouraging the private sector and NGOs for investment, developing service packages for palliative hospice care and end-of-life care based on the insurance costs, and developing the indicators of quality care to audit in this field.

By proposing a model tailored to the structure of the Iranian health system, in addition to being applicable in policymaking, launching relevant services in the country, and developing palliative care network. It can be used in various educational, research and clinical fields including hospice care specialty courses, the design and implementation of pilot studies and the management of end-stage cancer patients and their families in different health system settings.

AUTHOR'S NOTE

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DATA AVAILABILITY STATEMENT

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

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Ahvaz Jundishapur University of Medical Sciences under the code of IR.AJUMS.REC.1397.306. Participants provided their written informed consent to participate in the study.

AUTHOR CONTRIBUTIONS

SB and KZ conceived the study and contributed to its design. SB did the quantitative and qualitative data collection, analysis, and interpretation and drafted the manuscript. MR, MH, and SM contributed to study design, draft preparation, and study coordination. KZ, MR, MH, and SM supervised and coordinated the study project. SH participated in study consultation and critical revision of the article. All authors read and approved the final manuscript.

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How Do We Talk With People Living With Dementia About Future Care: A Scoping Review

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A diagnosis of dementia often comes with difficulties in understanding a conversational context and expressing how one feels. So far, research on how to facilitate advance care planning (ACP) for people with dementia focused on defining relevant themes and topics for conversations, or on how to formalize decisions made by surrogate decision makers, e.g., family members. The aim of this review is to provide a better scope of the existing research on practical communication aspects related to dementia in ACP conversations. In November 2020, seven databases were searched to select papers for inclusion (Proquest, Medline, Embase, Scopus, Psycinfo, Amed, and Cinahl). This search was updated in December 2021. The search strategy consisted of three tiers (related terms to "dementia," "communication" and "ACP"), intersected by using the Boolean term "AND," and resulted in 787 studies. Two researchers followed explicit criteria for two sequential levels of screening, based on titles and abstracts and full papers. A total of 22 studies were included for data analysis. Seven topics (i.e., importance of having ACP conversations, knowledge gap, inclusion of people with dementia in ACP conversations, policy vs. practice, adapting to cognitive changes, adapting to psychosocial changes, and adapting to emotional changes) emerged clustered around two themes (i.e., communicating with people with dementia in ACP, and changes in communication due to dementia). This scoping review provides practical suggestions for healthcare professionals to improve ACP communication and uncovered gaps in research on communication aspects related to dementia in ACP conversations, such as non-verbal behavior, timing and implementation, and personal preferences.

Keywords: dementia, advance care planning, communication, scoping review, palliative aged care

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INTRODUCTION

Advance care planning (ACP) can be described as the process that supports adults at any age or stage of health in understanding and sharing their personal values, life goals and preferences regarding medical care (Sudore et al., 2017). Performing ACP in early stages of any disease potentially increases the likelihood that a patient's wishes will be incorporated into care decisions, and reduce unwanted hospitalization and intensive treatments at the end of life [Detering et al., 2010; Silveira et al., 2010; for a systematic review on effects of ACP on end-of-life care, see Brinkman-Stoppelenburg et al. (2014)]. ACP has been associated with a range of positive end-of-life outcomes. Although the evidence base is still limited, this could be especially relevant for people with dementia, as they are likely to experience a prolonged period of cognitive decline, starting in early stages of the disease trajectory [for a systematic review on effects of ACP in improving end-of-life outcomes for people with dementia, see Dixon et al. (2018)].

Changes in cognitive abilities caused by dementia may not only impact decision-making capacities, but are also likely to affect the ability to process information and to participate in conversations. Difficulties in understanding a conversational context and expressing feelings may lead to several communication challenges, depending on the dementia condition (Rousseaux et al., 2010; Visser et al., 2020). For people with dementia, a simple chat with family members can be difficult, let alone having conversations with healthcare professionals about ACP. As a result, it is not very common for healthcare professionals to initiate ACP conversations with people with dementia (Azizi et al., 2022), and if any important ACP topics need to be discussed, they gradually turn to a proxy decision maker, like a family caregiver. However, people with dementia should have an active role as long as possible as well, when planning their future care (Van den Block, 2019). Healthcare professionals may adapt their communication strategies in ACP conversations to the cognitive abilities of people with dementia.

So far, research on how to facilitate ACP for people with dementia specifically focuses on defining relevant themes and topics for conversations, or on how to formalize decisions made by surrogates (Ampe et al., 2016; Tilburgs et al., 2018). As Van den Block (2019) describes in her editorial on the current state of research on ACP for people with dementia, much of the existing literature emphasizes on answering questions concerning the "what" of ACP, rather than understanding the "how" of ACP communication. Questions such as "How do we overcome the barriers for ACP conversations with people with dementia?" and "How to tailor communication in order to facilitate ACP conversations with people with dementia?" are still to be answered. A better scope of the existing research on communication aspects related to dementia in ACP conversations is needed to uncover any gaps in research in order to formulate communication strategies for healthcare professionals to improve their ACP practices, impacting the lives of people with dementia and their family members.

The aim of this scoping review is to examine how communication difficulties related to dementia are addressed in the literature (following the explicit scoping review method by Arksey and O'Malley, 2005). This paper serves as a starting point for future research and clinical practice, highlighting the contribution of involving people with dementia in making decisions and plans around their care.

METHODS

Eligibility Criteria

The study involved original empirical research papers focusing on communication and conversations about ACP or future palliative care, excluding discussions of immediate care relief. Subjects in these studies were to be people with dementia, family caregivers and/or healthcare professionals caring for people with dementia. Following scoping methods, no papers were excluded based on quality assessments of design and analyses, and no meta-analyses were used to aggregate findings (Arksey and O'Malley, 2005).

Information Sources and Search

In November 2020, a total of seven databases were searched to select papers for inclusion (Proquest, Medline, Embase, Scopus, Psycinfo, Amed and Cinahl). This search was updated in December 2021. Three tiers of the search strategy (related terms to "dementia," "communication" and "ACP") were intersected by using the Boolean term "AND," as presented in **Table 1**.

Paper Screening, Extraction and Analysis

Two sequential levels of screening were undertaken independently by two researchers (MV and HS), based on: (1) titles and abstracts; and (2) full-text papers. Any disagreement was reconciled by consensus. Inclusion criteria were as follows: (1) papers must describe original empirical research; (2) involve human subjects that have been diagnosed with dementia or care for people diagnosed with dementia; (3) include communication aspects related to dementia in either variables or outcomes, and (4) study conversations around future palliative care or ACP. Papers were excluded (1) if published in a language other than English, (2) if the study focused on nursing homes residents or older people in general (with no results reported on an identifiable subgroup of people with dementia). and (3) focused on discussions of immediate care relief. An initial data extraction was conducted by MV, focusing on type of research, participants, setting, and aims of the papers. Following this, two researchers (MV and JS) independently performed an initial content analysis to map out potential topics related to the aim of this research. The research team (MV, HS, JS, and DP) engaged in a iterative

TABLE 1 | Overview search strategy.

Tier 1[1]	AND	Tier 2	AND	Tier 3
Dementia OR Alzheimer's disease OR Frontotemporal dementia OR Vascular dementia OR Lewy body disease OR Parkinson's disease OR Cognitive impair		Communicate OR Social OR Conversation		Advance care planning OR Palliative

[1] *Tier 1 is based on the most common forms of dementia according to Alzheimer's Association (https://www.alz.org/alzheimers-dementia/what-is-dementia/types-of-dementia): Alzheimer's disease, frontotemporal dementia, vascular dementia, Lewy body disease, Parkinson's disease.*

process of discussing findings related to the topics to enhance analytical rigor and achieve consensus on outcomes.

RESULTS

Search Results and Study Characteristics

As illustrated in **Figure 1**, our search strategy resulted in 1,728 references. After removing duplicates, 756 references were imported to Covidence systematic review software (Veritas Health Innovation, Australia) to manage abstract- and full paper screening. After the title and abstract screening, 735 of 787 papers were excluded as they did not met all inclusion criteria as described above; in the full-text screening, 30 out of 52 papers were excluded for several reasons [i.e., not being accessible (6), or not describing original empirical research (11), not reporting on communication aspects (6), or palliative care or ACP (2) and not reporting dementia specific outcomes (5), leaving 22 papers for analysis].

As listed in **Table 2**, of the 22 included papers, 13 were published since 2016 (Ampe et al., 2016, 2017; Morin et al., 2016; Sinclair et al., 2016; Aasmul et al., 2018; Givens et al., 2018; Hill et al., 2018; Tilburgs et al., 2018; de Vries and Drury-Ruddlesden, 2019; Goossens et al., 2020a,b; Sussman et al., 2020; Yeh et al., 2021). The majority of papers described qualitative studies (n = 14), using methods such as interviews, focus groups and ethnographic observations (Godwin and Waters, 2009; Johnson et al., 2009; Sims-Gould et al., 2010; Goodman et al., 2013; Poppe et al., 2013; Beernaert et al., 2014; Ampe et al., 2016; Givens et al., 2018; Hill et al., 2018; Tilburgs et al., 2018; de Vries and Drury-Ruddlesden, 2019; Goossens et al., 2020a; Sussman et al., 2020). Only three papers described quantitative methods; two cluster randomized controlled trials (Ampe et al., 2017; Goossens et al., 2020b) and one survey described a single survey (Morin et al., 2016). Two studies used a mixed methods approach (Karel et al., 2007; Yeh et al., 2021) and two Delphi studies were included (van der Steen et al., 2014; Sinclair et al., 2016; Sussman et al., 2020). Papers described studies that were conducted in ten different countries, with five studies conducted in Belgium (Beernaert et al., 2014; Ampe et al., 2016, 2017; Goossens et al., 2020a,b), four in the United Kingdom (Godwin and Waters, 2009; Goodman et al., 2013; Poppe et al., 2013; Sinclair et al., 2016) and four in the United States of America (Kayser-Jones, 2002; Karel et al., 2007; Givens et al., 2018; Yeh et al., 2021). Other studies were conducted in Canada (Sims-Gould et al., 2010; Hill et al., 2018; Sussman et al., 2020), Norway (Aasmul et al., 2018), France (Morin et al., 2016), Netherlands (Tilburgs et al., 2018), Australia (Johnson et al., 2009) and New Zealand (de Vries and Drury-Ruddlesden, 2019). One paper used Delphi study methodologies including different countries across the world (van der Steen et al., 2014).

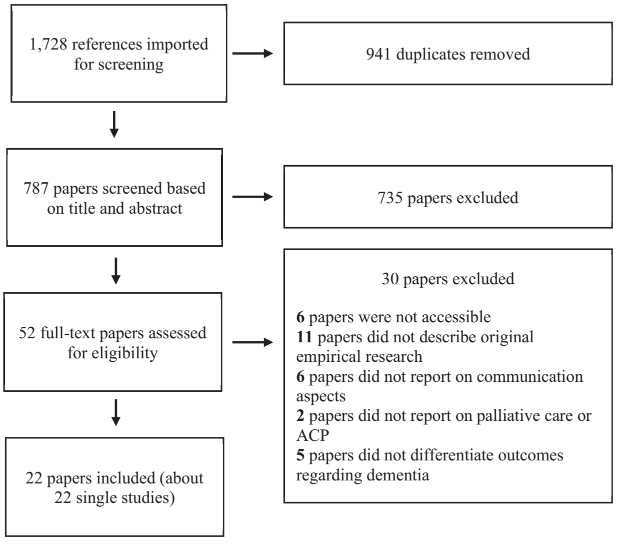

FIGURE 1 | PRISMA flowchart procedure paper screening.

Topics

A total of seven topics were identified clustered around two themes. The first four topics reflect on communicating ACP with people with dementia in general (i.e., covering the importance of ACP for people with dementia, the knowledge gap, the inclusion of people with dementia in ACP conversations, and how daily practice may differ from policy). Three more topics emerged around cognitive, psychosocial and emotional changes due to dementia, and how to adapt communication strategies to improve ACP conversations with people with dementia (see **Figure 2** for an overview).

Topic 1: Importance of Having Advance Care Planning Conversations

All papers underline that ACP is central to high-quality, holistic end-of-life care. Five papers in total specifically focused on the importance of having ACP conversations (Kayser-Jones, 2002; Sims-Gould et al., 2010; Goodman et al., 2013; van der Steen et al., 2014; Sinclair et al., 2016). It is likely to reduce unnecessary interventions, promotes comfort and increases clarity in important decision making (e.g., Goodman et al., 2013). An ethnographic study by Kayser-Jones (2002) showed that "*lack of attention to cultural needs, cognitive status, inadequate staffing, and inappropriate and inadequate communication between healthcare providers and nursing home residents and their families were the predominant factors that influenced the experience of dying.*" The two Delphi studies showed that proactive conversations around care and making decisions are essential for quality of care at the end of life (van der Steen et al., 2014; Sinclair et al., 2016). These conversations should be part of a continuous process of discussing values in life, rather than the single act of completing a form about care preferences (Goodman et al., 2013; Sinclair et al., 2016). To do so, ongoing communication amongst healthcare professionals, people with dementia and family caregivers appears to be essential (Sims-Gould et al., 2010).

TABLE 2 | Papers included.

Author (et al.)	Published	Title	Country	Type	Population, sample
Aasmul et al.	2018	Advance Care Planning in Nursing Homes—Improving the Communication Among Patient, Family, and Staff: Results From a Cluster Randomized Controlled Trial (COSMOS)	Norway	Quant*, CRCT*	545 residents, nursing home
		Aim: investigate effect of ACP on communication among NH staff, patient, and family and nursing staff distress			
Ampe et al.	2016	Advance care planning for nursing home residents with dementia: policy vs. practice	Belgium	Qual*, observations	20 nursing homes
		Aim: evaluate ACP policy in nursing homes and the involvement of residents and families, and how policy relates to practice			
Ampe et al.	2017	Advance care planning for nursing home residents with dementia: Influence of “we DECide” on policy and practice	Belgium	Quant*, Pre-test-post-test CRCT*	90 care workers, 18 dementia care units
		Aim: pilot ACP intervention in terms of influence on ACP policy and practice in nursing homes			
Beernaert et al.	2014	Early identification of palliative care needs by family physicians: A qualitative study of barriers and facilitators from the perspective of family physicians, community nurses, and patients	Belgium	Qual*, focus groups, interviews	20 GPs, 12 community nurses, 18 people with dementia
		Aim: explore barriers and facilitators of early identification by family physicians of the palliative care needs			
de Vries and Drury-Ruddlesden	2019	Advance care planning for people with dementia: Ordinary everyday conversations	New Zealand	Qual*, interviews	23 family caregivers
		Aim: explore experiences of preparedness and support for family members of people with dementia, before, during and following death.			
Givens et al.	2018	Advance care planning in community-dwelling patients with dementia	United States	Qual, observations	62 family caregivers
		Aim: examine aspects of ACP among patients with dementia and health care proxy acceptance of patients’ illness			
Godwin and Waters	2009	“In solitary confinement”: Planning end-of-life wellbeing with people with advanced dementia, their family and professional carers	United Kingdom	Qual*, observations, interviews	10 people with dementia and family caregivers, 4 wards in nursing home
		Aim: explore opinions of people with advanced dementia, family and professional carers on good terminal care and well-being			
Goodman et al.	2013	Preferences and priorities for ongoing and end-of-life care: A qualitative study of older people with dementia resident in care homes	United Kingdom	Qual*, interviews	18 people with dementia, nursing home
		Aim: explore how older people with dementia discuss priorities and preferences for end-of-life care			
Goossens et al.	2020a	Shared decision-making in advance care planning for persons with dementia in nursing homes: a cross-sectional study	Belgium	Qual*, Observations	65 wards in nursing home
		Aim: explore how health professionals and residents with dementia perceive the level of SDM during ACP conversations			
Goossens et al.	2020b	Improving shared decision-making in advance care planning: Implementation of a cluster randomized staff intervention in dementia care	Belgium	Quant*, Pre-test-post-test CRCT*	311 healthcare professionals, nursing home
		Aim: examine effects of ACP intervention on SDM for persons with dementia in nursing homes, perceived importance, competence and frequency of staff members concerning SDM and facilitating and hindering context elements for the sustainability of the training results			
Hill et al.	2018	Staff Perspectives of Barriers to Access and Delivery of Palliative Care for Persons With Dementia in Long-Term Care	Canada	Qual*, interviews	22 healthcare professionals, nursing home
		Aim: investigate experiences of staff delivering palliative care to individuals with dementia to determine how care was delivered, to learn which guidelines were used, and whether policies affected the delivery of palliative care			
Johnson et al.	2009	The communication challenges faced in adopting a palliative care approach in advanced dementia	Australia	Qual*, focus groups, interviews	34 dementia medical experts
		Aim: examine communication issues affecting implementation of palliative care for persons with dementia in residential aged care facility			
Karel et al.	2007	Three methods of assessing values for advance care planning: comparing persons with and without dementia	United States	Mixed*, interviews, surveys	176 older adults (of whom 88 people with dementia)
		Aim: examine the utility of health care values assessment tools for older adults with and without dementia			
Kayser-Jones	2002	The Experience of Dying: An Ethnographic Nursing Home Study	United States	Qual*, observations, interviews	35 residents, 52 family caregivers: 52, 102 healthcare professionals, nursing home
		Aim: investigate the process of providing end-of-life care to residents who were dying in nursing homes			
Morin et al.	2016	Discussing end-of-life issues in nursing homes: a nationwide study in France	France	Quant*, survey	674 family caregivers, nursing home

(Continued)

TABLE 2 | (Continued)

Author (et al.)	Published	Title	Country	Type	Population, sample
		Aim: investigate how discussing end-of-life issues and frequency of conversations associate with care outcomes			
Poppe et al.	2013	Qualitative Evaluation of Advanced Care Planning in Early Dementia (ACP-ED)	United Kingdom	Qual*, interviews	12 people with dementia, 8 family caregivers, 6 healthcare professionals
		Aim: explore the acceptability of discussing ACP with people with memory problems and mild dementia shortly after diagnosis			
Sims-Gould et al.	2010	Care Provider Perspectives on End-of-life Care in Long-Term-Care Homes: Implications for whole-person and palliative care	Canada	Qual*, ethnography	10 residents, nursing home
		Aim: explore experiences of dying and end-of-life care for persons with dementia in long-term care from the perspective of care providers			
Sinclair et al.	2016	Consensus views on advance care planning for dementia: a Delphi study	United Kingdom	Delphi study	
		Aim: investigate consensus views of how ACP should be explained and carried out with people with dementia			
Sussman et al.	2020	Engaging persons with dementia in advance care planning: Challenges and opportunities	Canada	Qual*, focus groups	10 people with dementia, 8 family caregivers
		Aim: explore experiences with ACP, concerns related to end-of-life care, and practices supporting positive engagement with ACP			
Tilburgs et al.	2018	The importance of trust-based relations and a holistic approach in advance care planning with people with dementia in primary care: a qualitative study	Netherlands	Qual*, interviews, focus groups	10 people with dementia, 10 family caregivers, 10 GP's, 1 focus group (nurses, case managers)
		Aim: explore barriers and facilitators for ACP with community-dwelling people with dementia			
van der Steen et al.	2014	White paper defining optimal palliative care in older people with dementia: A Delphi study and recommendations from the European Association for Palliative Care	Worldwide	Delphi study	
		Aim: define optimal palliative care in dementia			
Yeh et al.	2021	Improving end-of-life care for persons living with dementia: Bereaved caregivers' observations and recommendations	United States	Mixed*, surveys interviews	53 family caregivers
		Aim: elicit recommendations for improving end-of-life care experiences of people with dementia from the perspective of bereaved caregivers			

**Quant, quantitative research; Qual, qualitative research; Mixed, mixed methods (both qualitative and quantitative research); CRTC, cluster randomized controlled trial.*

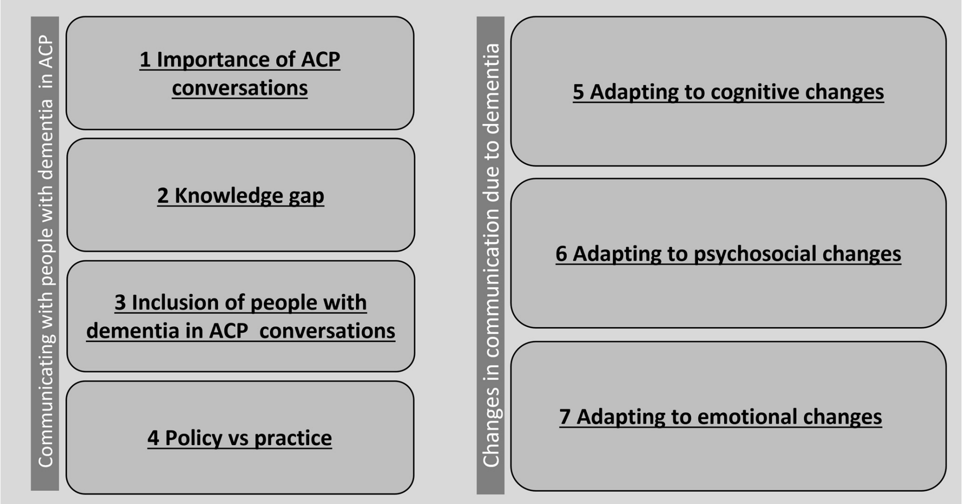

FIGURE 2 | Topics around communication aspects related to dementia and ACP found in the scoping review.

Topic 2: Knowledge Gap

Although it is clear that communication about care between healthcare professionals and patients is an important determinant of quality end-of-life care, so far, characteristics and determinants of good quality communication have been hard to identify. Two papers described the lack of knowledge on how to provide good quality communication (Goodman et al., 2013; Aasmul et al., 2018). For example, Goodman et al. (2013) interviewed 18 people with dementia about their ACP preferences, and they found it is unclear how people with dementia and their current experiences can inform planning and decision making around ACP. Although numbers of empirical studies on ACP with people with dementia are increasing, it seems that well-powered controlled trials investigating communication between people with dementia and healthcare professionals in long term care facilities (LTCF) are needed (Aasmul et al., 2018).

Topic 3: Inclusion of People With Dementia in Advance Care Planning Conversations

Five papers showed that people with dementia are rarely included when discussing ACP (Johnson et al., 2009; Goodman et al., 2013; Poppe et al., 2013; Ampe et al., 2016; Givens et al., 2018). Ampe et al. (2016) clarified that healthcare professionals (1) were more comfortable to talk with family caregivers, and (2) underestimated the capability of people with dementia themselves to talk about preferences related to ACP. However, a decision made by a family caregiver may be inconsistent with the person with dementia their wishes, and family caregivers state making decisions on behalf of someone else around their end-of-life care is difficult (Poppe et al., 2013). Therefore, there is a need to invest in including people with dementia discussing their preferences around ACP.

Topic 4: Policy Versus Practice

Only two studies included in this review (Ampe et al., 2016, 2017) show a lack of strategies for implementing ACP conversations in dementia care. Apparently, organizational healthcare policies concerning ACP are present and promising, although often not implemented in daily practice of healthcare professionals. For example, although the intervention "We DECide" by Ampe et al. (2017) positively influenced the intension of performing ACP in participating dementia care units, the actual practice did not improve. Healthcare professionals only managed to involve residents or their family caregivers on a baseline skill level. It is clearly not enough for healthcare professionals to understand and acknowledge evidence-based practices of ACP, and practically oriented training is required to change habits and behaviors. Ampe et al. (2017) suggest adding a strong focus on the organizational context of ACP training (e.g., by including the management of the team in discussions on the topic, or by organizing in service training for trainers).

Topic 5: Adapting to Cognitive Changes

A total of eleven papers describe how changes in cognitive abilities with people with dementia may affect the way they participate in ACP, or how to deal with conversation difficulties that arise because of cognitive changes (Karel et al., 2007; Godwin and Waters, 2009; Johnson et al., 2009; Goodman et al., 2013; Poppe et al., 2013; van der Steen et al., 2014; Ampe et al., 2016; Sinclair et al., 2016; Aasmul et al., 2018; Tilburgs et al., 2018; Goossens et al., 2020a). Older people living with dementia show loss of memory, communication, orientation, control, autonomy, independence, self-esteem and relationships (Goodman et al., 2013). Poppe et al. (2013) stated this may affect the way people with dementia make decisions and how they are informed. Johnson et al. (2009) showed that "*dementia has a trajectory that causes a diminishing ability for the person to express their care needs and treatment wishes*." In their study, Ampe et al. (2016) found that LCTF residents with dementia were no longer capable of discussing important decisions. Moreover, healthcare professionals appear to underestimate communication skills of people with dementia, and their ability to process abstract concepts (Godwin and Waters, 2009). According to van der Steen et al. (2014) communicating with patients and families of people with dementia requires special skills due to cognitive problems that come with dementia complicating decision making. We found several suggestions to overcome these communication problems related to cognitive changes:

Question Formulation

After interviewing general practitioners, Tilburgs et al. (2018) suggest ACP should be tailored to the cognitive level of the person with dementia by asking closed instead of open questions. Communication training may help healthcare professionals to remain aware of the way they formulate questions (Aasmul et al., 2018). Moreover, people with dementia should be offered the time to prepare themselves for certain questions and the ability to process them (Goossens et al., 2020a). Apparently, the way questions are asked in ACP conversations should be tailored according to dementia severity. However, interestingly, Karel et al. (2007) found no differences between participants with mild dementia and control participants (without dementia) with regards to question formulation in their survey research. People with mild dementia were able to answer open-ended questions about quality of life and responded to forced-choice questions regarding healthcare values.

Timely Initiation

According to Poppe et al. (2013), ACP is usually completed in the last 6 months of life, which may cause problems for people diagnosed with dementia: decision-making capacity and ability to communicate decrease as the disease progresses. Therefore, the initiation of ACP should happen as early in the disease trajectory as possible, preferably shortly after diagnosis, while people with dementia still have the capacity to make informed decisions (Aasmul et al., 2018; Tilburgs et al., 2018), but only when they are ready to do so (Sinclair et al., 2016). Ampe et al. (2016) found that rather than to wait for crisis situations, ACP should be discussed before. They mention admission in a LTCF to be a good moment to talk about ACP. Godwin and Waters (2009) underlined the importance to keep a conversation ongoing; we cannot presume views and conceptions around ACP remain unchanged.

Non-verbal Behavior

Several papers underline the importance of observing behavior of people with dementia while in conversation about ACP. Healthcare professionals should not only focus on well formulated (spoken) views (Ampe et al., 2016). If we neglect non-verbal behavior, we may miss important connotations (Godwin and Waters, 2009). Recognition of behaviors as potential signs of discomfort need to be observed and addressed (Johnson et al., 2009).

Topic 6: Adapting to Psychosocial Changes

A total of nine papers discussed how psychosocial changes due to dementia may affect ACP conversations or how to overcome difficulties that arise because of these changes (Kayser-Jones, 2002; Karel et al., 2007; Johnson et al., 2009; Goodman et al., 2013; Poppe et al., 2013; Ampe et al., 2016; Aasmul et al., 2018; Tilburgs et al., 2018; Goossens et al., 2020a). Awareness of dementia may affect one's self-esteem and confidence in a negative way (Goodman et al., 2013). Still, the opportunity to participate in ACP and contribute to choices around their care may help people with dementia feel more empowered (Goossens et al., 2020a). We found several suggestions to improve one's confidence in ACP conversations:

Familiar People

A well-established relationship with a healthcare professional may have a positive effect on the course of an ACP conversation (Ampe et al., 2016). Moreover, follow-up conversations may help to build relationships further, and are also necessary to maintain an ACP routine (Aasmul et al., 2018). However, several papers underline familiarity with the healthcare professional is not as important for successful ACP communication as well-trained and empathetic conversation partners (Karel et al., 2007; Poppe et al., 2013; Tilburgs et al., 2018). According to Karel et al. (2007), familiarity is not essential for good ACP practice. In fact, in their study, non-familiar people who behaved attentive and empathetic, were successful in eliciting care preferences with people with dementia. This suggests more research on the relationship between healthcare professionals and people with dementia is needed.

Training of Healthcare Professionals in Effective Communication Skills

A lack of communication skills can be a barrier for conducting successful ACP conversations, such as not listening, trivializing situations and being distant in conversations (Kayser-Jones, 2002; Johnson et al., 2009; Tilburgs et al., 2018; Goossens et al., 2020a). In addition to possessing knowledge about the purpose and goals of ACP, healthcare professionals need to possess effective communication skills in order to initiate and sustain dialog on the future care of people with dementia (Johnson et al., 2009; Poppe et al., 2013). In a study by Poppe et al. (2013), healthcare professionals identified knowledge about dementia, available resources and knowledge of one's own limitations as key skills and competencies for discussing ACP. Feeling confident when discussing ACP was mentioned to be important and healthcare professionals found having experience in dealing with difficult conversations increased their confidence.

Healthcare Professionals Empathy

Discussing their findings of their study on improving ACP communication between healthcare professionals and people with dementia, Aasmul et al. (2018) stated that a low level of empathy with staff may have affected ACP conversations in a negative way. This fits well with work of Tilburgs et al. (2018), who found non-empathic attitudes by general practitioners to be a barrier of ACP with people with dementia. It seems to be important for healthcare professionals to have an empathetic attitude while talking to people with dementia about ACP.

Topic 7: Adapting to Emotional Changes

Dementia causes substantial changes in one's life that may elicit different emotions and feelings compared to feelings people had before their diagnosis, or compared to those with other diseases. A total of ten papers reported on how emotional changes that come with dementia may possibly affect ACP conversations, or on how to overcome challenges due to these changes (Kayser-Jones, 2002; Godwin and Waters, 2009; Poppe et al., 2013; Beernaert et al., 2014; Ampe et al., 2016; Aasmul et al., 2018; Givens et al., 2018; Hill et al., 2018; de Vries and Drury-Ruddlesden, 2019; Yeh et al., 2021). People with dementia often experience agitation, fear, confusion, and pain. This may be difficult to cope with, for themselves or those around them (Hill et al., 2018). The thought of needing palliative care can be confronting, and therefore ACP conversations tend to be avoided by some (Beernaert et al., 2014). In general, ACP conversations are practically oriented, and not so much focusing on emotions. Although crisis situations often contain emotional moments, there may be little room for discussing emotions when making decisions, possibly related to time pressure. This could be a missed opportunity to understand which and how emotions influence certain decisions (Ampe et al., 2016). We found several suggestions to cope with emotions better in ACP conversations:

Not Avoiding the Topic

Often, in day-to-day conversations between people with dementia and healthcare professionals, safe topics and social chit-chat are most apparent (Kayser-Jones, 2002). Beernaert et al. (2014) found that neither people with dementia nor healthcare professionals initiate discussion of non-acute care needs in day-to-day conversations. According to Godwin and Waters (2009), the ability and especially the willingness of people with dementia to talk about abstract concepts such as death is underestimated with healthcare professionals. Yeh et al. (2021) underline that initiative for ACP conversations between people with dementia and healthcare professionals can also come from family caregivers.

Regular Meetings

It may be beneficial for healthcare professionals to initiate regular conversations with people with dementia to talk about ACP. Aasmul et al. (2018) suggested quarterly meetings with nursing home residents with dementia and monthly meetings with their family caregivers. According to a study in nursing homes by Ampe et al. (2016), people with dementia should be engaged actively in the ACP process, not only when they are admitted to a LTCF, "but throughout their stay." They state it is the responsibility of healthcare professionals to stimulate colleagues and other care workers to pick up on conversations and communicate relevant information about ACP.

Normalizing Advance Care Planning

Conversations around important topics such as end-of-life and care preferences may cause some anxiety in patients, but there is a need to normalize these kind of conversations. Poppe et al. (2013)

showed that ACP conversations gave people with dementia time to think about their future. Some were relieved and less worried after discussing their preferences, and they felt supported by their family and services. It was important for them that family caregivers and healthcare professionals were attentive to their preferences. Moreover, discussing ACP within families makes room for having those conversations in an informal, day-to-day setting (de Vries and Drury-Ruddlesden, 2019). According to Givens et al. (2018), ACP conversations in family settings do not happen enough but would benefit everyone involved.

DISCUSSION

With this scoping review, we mapped existing literature on communication aspects related to dementia in ACP conversations. Seven topics (importance of having ACP conversations, knowledge gap, inclusion of people with dementia in ACP conversations, policy vs. practice, adapting to cognitive changes, adapting to psychosocial changes, and adapting to emotional changes) emerged. Practical suggestions for healthcare professionals to improve ACP communication are provided and summarized in **Box 1**. This review underlines the scarcity of studies focusing on "how" to talk to a person with dementia about their ACP preferences; more research is needed on non-verbal communicative aspects of ACP conversations, timing and implementation of these conversations, and preferences of individuals with dementia related to these conversations, as results of studies so far have been inconsistent or have not given any or little attention to these topics.

One topic that was mentioned in the literature several times but seems to lack empirical evidence, is how expressive behavior related to dementia may affect the process of an ACP conversation between a patient and a healthcare professional. Research underlines the importance of reading non-verbal behavior for ACP conversation processes and outcomes (Godwin and Waters, 2009; Johnson et al., 2009; Ampe et al., 2016), but how to do so is still to be studied. Numerous studies show changes in expressive abilities of people with dementia [e.g., emotional expressions, review by Lee et al. (2019); expressions of apathy, Kumfor et al., 2018; and mutual eye gaze, Sturm et al., 2011], and may be extra prone to affect behavior in conversations around sensitive topics of ACP.

BOX 1 | Practical suggestions for healthcare professionals to improve ACP communication.

- Make continuous and active conversations part of the ACP practice, rather than the single act of completing a form about care preferences.
- Make an effort to include people with dementia themselves in ACP conversations.
- Tailor the questions asked in ACP conversations according to dementia severity and personal needs.
- Initiate ACP as early in the disease trajectory as possible, preferably shortly afterward the diagnosis.
- Focus not only on well formulated (spoken) views but also pay attention to non-verbal behavior of person with dementia.
- There needs to be empathy and attentive listening.
- Do not underestimate the ability and the willingness of people with dementia to talk about abstract concepts such as ACP.
- Initiate ACP conversations on a regular basis.
- Normalize talking about ACP by initiating ACP conversations in informal settings.

Another important topic that needs more research is timing and implementation of ACP conversations and practice. Research seems to underline the significance of "early" initiated conversations, and that initiation in care facilities may be too late to include people with dementia (Poppe et al., 2013; Ampe et al., 2016; Sinclair et al., 2016; Aasmul et al., 2018; Tilburgs et al., 2018). More research is needed on how to define the right moment. Several papers suggest healthcare professionals to "have a continuous conversation" with people with dementia about ACP, and to "talk about ACP in an informal way and often." However, it seems to be difficult to implement such suggestions in practice. Healthcare professionals often feel incapable of initiating conversations on sensitive topics like ACP and tend to avoid these (Beernaert et al., 2014). Future research should have a strong focus on how to implement ACP in practice, rather than only formulating ACP policies. Also, although our findings suggest that ACP conversations are needed on a regular base, and healthcare professionals investing time seems necessary, the ideal duration of an ACP conversation remains unclear (e.g., de Vries and Drury-Ruddlesden, 2019).

Further research is needed on the optimal timing and frequency of conversations, but also on models of care in which healthcare professionals who are best positioned to conduct and personalize ACP, are supported to do so. A review by Piers et al. (2018), describes evidence-based guidelines for healthcare professionals across settings in the practical application of ACP in dementia care. Amongst recommendations on topics such as documentation and end-of-life decisions, limited suggestions are made around communication strategies in ACP conversations that are in line with the recommendations as presented in **Box 1**(e.g., adjust one's communication style and content to their own level and rhythm). Still, papers included in this scoping review showed different findings with regard to the importance of familiar healthcare professionals leading ACP conversations, the effect of (lack of) empathy by the healthcare professional, and whether or when an approach should be more or less directive. Preferences for the way an ACP conversation is held may be personal, and can change over time, depending on many factors, such as cultural background or even mental conditions. For example, in some cultures, fostering autonomy of the person with dementia is important, while in other cultures, a paternalistic approach may be useful at times. As this is barely reflected in the studies included in this scoping review, future work needs to focus on personal preferences that demand a level of flexibility of healthcare professionals to adopt different approaches as needed.

This scoping review has several strengths and limitations. Our strength is that we focused specifically on communicative aspects of ACP conversations with people with dementia, uncovering significant gaps in literature and providing recommendations for research and practice. Unfortunately, most studies covered this topic within a broader scope, focusing on ACP practice related to dementia in general, leaving several important factors underexposed. No papers that were included in the scoping review focused on cultural differences as a determinant for how to communicate ACP with people with dementia, limiting the generalizability while also exposing a need for future studies on ACP to include non-Western populations. In fact, all studies included were conducted in Western high-income studies. A reason for this could be we only included papers written in English. Another complicating matter when interpreting results and recommendations of the papers included in the scoping review, was that papers referred to different ACP policies that were custom for specific countries. Legislation and guidelines at national and international level may determine how ACP is approached and is still being developed (Alzheimer Europe, 2020). Still, scoping the literature, we did find a number of practical implications for ACP practice in general, to be adapted by healthcare practitioners and family caregivers, in order to improve ACP practices. Conversations about ACP should be continuous practice, with an active role for people with dementia themselves. Timing, content and form have to be tailored to their cognitive, psychosocial and emotional needs. This study underlines the need for more thorough, empirical studies that look into how to talk to people with dementia about future care.

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

MV, HS, JS, and DP: conceptualization. MV and HS: data curation. MV and JS: formal analysis. MV: funding acquisition and writing—original draft. HS, JS, and DP: writing—review and editing. All authors approved the final version to be published and read and agreed to the published version of the manuscript.

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Psychometric Properties of the Persian Version of Palliative Care Outcome Scale (POS) in Adult Patients With Cancer

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Background: Measuring the outcomes of palliative care plays an important role to improve the quality, efficiency, and availability of these services in patients with cancer. Using valid, reliable, and culturally appropriate tools has a considerable role to measure these outcomes. This study aimed to assess the psychometric properties of the translated version of the Palliative care Outcome Scale (POS).

Methods: This methodological study was conducted in two outpatient clinics related to Shohada Tajrish and Baqiyatallah hospitals in Tehran in 2019–2020. The translation was done using the Forward-Backward approach after gaining permission from the developer. Face validity was tested with 10 patients with cancer through cognitive interviewing, as well as content validity with four experts. Construct validity was performed by ($n = 203$) exploratory factor analysis and confirmation ($N = 150$). To assess the reliability, internal consistency was assessed by using Cronbach's alpha coefficient, and relative stability was assessed using the interclass correlation coefficient (ICC). Furthermore, interpretability and ceiling and floor effects were assessed.

Results: A total of 353 patients with cancer under palliative care were included in the study. Then, three psychological (30%), physical (12.25%), and social factors (12.08%) with a cumulative variance of 54.34% were extracted in exploratory factor analysis. Confirmatory factor analysis showed that the model has a good fit of information. Cronbach's alpha coefficient for scale was 0.719. Furthermore, the ICC was 0.812. The scale was interpretable, and ceiling and floor effects were 0%.

Conclusion: Persian version of the POS was evaluated as a valid and reliable tool. Therefore, it can be used by the clinician to monitor the consequences of palliative care in Iranian cancer patients.

Keywords: palliative care, measurement, validity, reliability, psychometrics, scale, symptom, outcome and process assessment

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INTRODUCTION

There were an estimated 10 million deaths caused by cancer worldwide in 2020 (Ferlay et al., 2021). In the same year, 131,191 new cases of patients with cancer were diagnosed and 79,136 of them died in Iran (International Agency for Research on Cancer, 2020).

Patients with cancer and their families need physical care, as well as spiritual, psychological, and social support in Iran (Aghaei et al., 2021). Thus, palliative care is used as a response to these needs by improving the quality of life for patients and their families *via* the prevention and relief of pain (Barasteh et al., 2020), reducing the burden of disease, improving satisfaction, and increasing the quality of life of patients (Hugar et al., 2021).

Palliative care is the science and art of improving the quality of life for dealing with a chronic illness over many years (Bakitas et al., 2009). Providing care and supporting patients and their families through the various stages of life-threatening diseases such as cancer are among the goals of palliative care (Aghaei et al., 2021). Receiving palliative care improves the quality of life of patients and their families, but only ∼14% of 40 million people needing palliative care each year actually receive it according to WHO (2022).

The establishment and development of palliative care in Iran is one of the goals of the Iranian health system. The first palliative care services in Iran were provided in 2006 at the Comprehensive Cancer Center in Tehran (Rassouli and Sajjadi, 2016). In 2014, the Ministry of Health and Medical Education launched a palliative care working group using various sources and opinions from related fields (Barasteh et al., 2020). At present, palliative care is provided in the form of counseling and comprehensive care in 5–6 centers in Tehran, Isfahan, and Zanjan (Rassouli and Sajjadi, 2016).

Generally, palliative care services in Iran are mostly provided to patients with cancer, although in developed nations, this sort of care is provided to a broad spectrum of patients with terminal conditions, such as heart failure, COPD, and dementia. European Union emphasizes the provision of palliative care at the primary health care level for reasons such as increasing aging populations and the incidence of chronic and debilitating diseases (Heydari, 2018).

At first, providing palliative care faced many challenges in terms of the lack of clear definition, dimensions, and principles according to the cultural context of Iran. The Ministry of Health and Medical Education has helped develop palliative care by helping to form the Iranian Cancer Association and the presence of experts in various fields, as a result of designing and implementing related projects and studies and holding a congress (Barasteh et al., 2021). However, palliative care in Iran encounters a struggle due to weakness in policy-making, insufficient training, shortage of specialists, insurance system concerns, unclear roles and teamwork, medication access issues, and the need for civic support (Barasteh et al., 2020). Also, the development of clinical and research practices is ambiguous due to the lack of standard tools and measurement indicators.

Palliative care professionals must assess the efficacy of the care they offer as the demand for it grows, owing to the inclusion of a broad variety of chronic conditions in patients.

As a result, assessing palliative care outcomes is critical for improving the quality, efficiency, and accessibility of these treatments. Measuring outcomes as a principle in quality assurance and continuous quality improvement (Bausewein et al., 2011), is affected by changes in current and future health status of the patients (Donabedian, 1988; Porter et al., 2016).

In clinical settings, outcome measurement is being used to understand changes in patients' health status or quality of life, to facilitate communication with patients/families and health care team, and to help in clinical decision making and evaluation of the effectiveness of clinical interventions (Gruenewald et al., 2004; Pantaleon, 2019). Palliative outcome care is an essential tool to be used in clinics and research. It has been used for updating clinical practice, monitoring service interventions, and assessing and enhancing the quality of care (Kocatepe et al., 2020).

Patients are the primary source of information on changes in their health status and quality of life, hence the outcomes of health care are linked to their experiences. Examining the effects of palliative care may also aid in achieving the best potential outcomes. Therefore, it is important to use the appropriate tools to measure outcomes in palliative care (Porter et al., 2016).

Thus, various tools including the Memorial Symptom Assessment Scale (MSAS), Edmonton Symptom Assessment Scale (ESAS), and Palliative care Outcome Scale (POS) (Bruera et al., 1991; Aaronson et al., 1993; Roth et al., 1998; Groenvold et al., 2006) are designed to measure the outcomes of palliative care in various dimensions. The POS is a comprehensive tool developed by Hearn and Higginson as a multidimensional benchmark for people with advanced cancer. This scale includes physical and psychological symptoms, spiritual considerations, emotional concerns, and psychological and social needs of the patient and their family (Hearn and Higginson, 1999). One of the important strengths of this tool is its optimal validity and reliability, as well as its design based on a comprehensive care approach. The physical, psychological, social, and spiritual components of the patient's experience are all taken into account while creating this tool (Porter et al., 2016). The tool was translated into various languages, including Spanish (Eisenchlas et al., 2008), Turkish, German (Bausewein et al., 2005), and several other languages.

In Iran, the need for palliative care and its consequences was increasingly considered in recent years. Therefore, by considering the need to study the outcomes of palliative care in the centers which are providing these services in Iran, it is necessary to validate an international tool and provide sufficient information about its psychometric process. Therefore, this study was conducted to translate and assess the psychometric properties of the Persian version of the POS in adult cancer patients.

METHODS

Study Design

In this methodological study, the POS translated into Persian and assessed its psychometric properties in 2019–2020.

Study Population/Sampling

This study was performed on patients with cancer who were referred to outpatient clinics Shohada Tajrish and Baqiyatallah and who were in advanced stages of the disease. For this study, 353 patients with cancer were included by the convenience sampling method. Besides, 10 patients participated in the study on face validity. Furthermore, 4 experts were invited for qualitative content validity. Over 18 years old, cancer diagnosis based on physician and patient records, desire in participating in the research, capacity to speak vocally in Persian, and no cognitive or psychiatric issues were the inclusion criteria. Non-cooperation and refusal of the patient or caregiver during the study and incompletion of the scale were considered the exclusion criteria. After obtaining permission, the researcher went to the Baqiyatallah and Shohada Tajrish to start sampling. Statistical Package for the Social Sciences (SPSS) software, version 22.0 was used for data analysis. The maximum error of the first type was considered to be 5%.

Study Instruments

Demographic Information Questionnaire

Researcher-made questionnaire was used to collect the demographic information of the patients, such as age, sex, type of cancer, and painkillers usage history.

Palliative Care Outcome Scale (POS)

The POS was developed by Hearn et al. in 1999. It consists of 12 questions, and these questions are related to the patient's physical, mental, emotional, and social states. Except the last question, the answers to all questions are obtained using 0–4 Likert with 0–4 numerical labels. POS scores of each individual question from 1 to 10 can be described in a general score. The overall score is 0–40 so the highest score indicates the maximum disability. Questions 1–8 have 5 options that are rated from zero to four points. Question 9 has three options, and the scores of which are zero, two, and four, respectively. Question 10 has four options, and the points of which are zero, two, four, and zero, respectively. Question 11 completes question 10 and is an open answer. Question 12 of the questionnaire is also related to how to answer the questionnaire, which has 3 points. The measure demonstrated construct validity (Spearman rho = 0.43–0.8). Test/retest reliability was acceptable for seven items. Internal consistency was good (Cronbach's alpha = 0.65) (Hearn and Higginson, 1999).

Translation Procedure

The translation was done after receiving permission from the developer of the POS. First, the scale was translated from English (main language) to Persian (target language) by two translators (one health specialist and the other general translator). This version was translated back into English by two more translators after analyzing the translations and obtaining a final form from the original Persian version (third and fourth translators). Following the evaluation of the two translations, a copy was retrieved and given to the developer to check the quality of the translation using the International Quality of Life Assessment (IQOLA) translation process (Bullinger et al., 1998).

Face and Content Validity

Cognitive interviews were conducted to assess the qualitative face validity. In cognitive interviews, the source of error in the tools identified by focusing on the cognitive process of the respondents when filling out the questionnaire (Willis, 2004). Briefly, 10 patients with cancer were interviewed for face validity. Patients were interviewed face-to-face and changes were made to the Persian version of the POS. Furthermore, they were asked to score readability, clarity, item structure, ease of understanding, item complexity, and ambiguous terms, as well as question categorization, ease of replying, language forms, and wording. Furthermore, in order to match the translated version with the original English one and make sure the content can be correctly conveyed, we sent the questionnaire to four experts in Persian Literature to receive their feedback on language forms, diction, and the placement of the words and phrases (qualitative content validity assessment).

Construct Validity

Patients with cancer receiving palliative care were referred to Shohada Tajrish Center, Baqiyatallah Hospital. Data collection was performed from April 2019 to August 2020. The minimum sample size required for exploratory factor analysis (EFA) is 3–10 participants per item (Plichta, 1965). For EFA, 203 patients with cancer receiving palliative care services were included in the study by available sampling. The principal component analysis (PCA) method was used to extract the factors, and the Promax rotation was used to interpret the factors (Samitsch, 2014).

In order to conduct confirmatory factor analysis (CFA), Munro (2005) recommended 20–30 items per factor (Munro, 2005). Given that the original version of the scale considers 5 dimensions, 100–150 samples were required for CFA. Hence, 150 patients were included in the study for CFA. Indicators of model fit in CFA in three general categories are the following: (1) absolute fit: the root mean square error of approximation (RMSEA), standardized root means square residual (SRMR), Goodness-of-Fit index (GFI), and chi-square; (2) comparative fit: index (CFI), incremental fit index (IFI), relative fit index (RFI), normed fit index (NFI), and Tucker-Lewis index (TLI); (3) affordable fit: parsimony comparative fit index (PCFI), parsimony normal fit index (PNFI), adjusted goodness-of-fit index (AGFI), and Akaike's information criterion (AIC) (Plichta, 1965; Samitsch, 2014).

Reliability

The correlation among the items referred to internal consistency in a tool that was assessed by Cronbach's alpha coefficient. Furthermore, relative stability was assessed using the interclass correlation coefficient (ICC). The sample size of 28 patients and the interval between two measurements of 1 week were considered.

Interpretability

To assess the interpretability, the correlation of the total score of scale with gender, age using Pearson Correlation and *t*-test were assessed. Furthermore, the SEM and minimal detectable changes (MDC) were calculated. The following equation was used to calculate the standard error of measurement:

$$\text{SEM} = \ \text{SD}\sqrt{1} - -\text{ICC}$$

Where SD is the standard deviation of the sum values obtained in test and retest phases, while the ICC is the coefficient of repeatability. To calculate the MDC, we used the following equation:

$$\text{MDC} = \text{SEM}\times \text{z}\ \sqrt{2}$$

Furthermore, MDC can be calculated as a percentage of the MDC% to determine the actual relative changes after treatment or among repeated measurements over time to further show the relative value of the random error of measurement. To calculate this, the following equation was used:

$$\text{MDC\%} = (\text{MDC}\div\text{mean})\times\ 100$$

Where "MDC%" is acceptable if it is smaller than 30%, and the excellent MDC% value is assumed to be below 10% (Wu et al., 2011; Sajadi et al., 2020). Percentage of minimum and maximum scores (floor or ceiling effect was considered to be present if >15% of the subjects achieved the lowest or highest possible scores, respectively) (Terwee et al., 2007).

Data Analysis

We used SPSS software, version 22.0. for EFA and software Linear Structural Relationship (LISREL) version 8.8 for CFA. In all analyses, the significance level was considered $P < 0.05$.

TABLE 1 | Rating of exploratory factor analysis (EFA) and confirmatory factor analysis (CFA) based on demographic characteristics.

	Category	EFA *N* (%)	CFA *N* (%)
Gender	Male	95 (46.8)	70 (46.7)
	Female	108 (53.2)	80 (53.3)
Age-group (year)	0–30	36 (17.7)	33 (22.00)
	30–50	93 (45.8)	63 (42.00)
	50–70	66 (32.5)	47 (31.3)
	<70	8 (3.9)	7 (4.7)
Type of cancer	Gastrointestinal	76 (37.4)	63 (42.00)
	Blood	37 (18.2)	25 (16.7)
	Uterus and ovary	33 (16.3)	23 (15.3)
	LUNG	16 (7.9)	10 (6.7)
	Others	41 (20.2)	29 (19.3)
Type of pain relief	Non	14 (6.9)	7 (4.7)
	OPOIDS	96 (47.3)	75 (50.00)
	NSAIDS	93 (45.8)	68 (45.3)

Ethical Consideration

The present study was conducted in the ethics committee of Baqiyatallah University of Medical Sciences with the ethics code IR.BMSU.REC.1396.134. The researcher described the study approach to all participants and acquired their signed consent after getting the appropriate authorization and coordinating with the relevant authorities. Participants were also told that the study's data would be kept confidential and that they may exit at any moment.

RESULTS

Socio-Demographic and Clinical Status

Participants in EFA included 203 patients 95 (46.8%) were men and 108 (53.2%) were women with a mean age of 44.81 ± 14.37. In the CFA, another 150 patients had a mean age of 43.84 years, of which 70 (46.7%) were men and 80 (53.3%) were women. Other details are given in **Table 1**.

Face and Content Validity

Face validity was confirmed using 10 adult patients with cancer. Moreover, content validity was determined by 4 expert specialists. The items did not change in face and content validity in terms of their simplicity and clarity.

Construct Validity

Exploratory Factor Analysis

The Kaiser-Meyer-Olkin (KMO) test at 0.733 and Bartlett's test were significant ($P = 0$). According to the results, three factors of psychological factors (items 7, 8, 4, and 3), physical factors (items 1, 2, and 9), and social factors (items 6, 10, and 5) were extracted. These three factors accounted for 54.34% of the consequences of palliative care (**Table 2**).

Confirmatory Factor Analysis

Another sample consisting of 150 patients was selected for CFA. The results of the chi-squared test (x2 = 65.11 and $P = 0$) and other fit incises showed that the three-factor model extracted from EFA has a good fit of the data (RMSEA: 0.072; NFI: 0.88;

TABLE 2 | EFA of the Persian version of the Palliative care Outcome Scale (POS).

Factor	Item	Factor loading	variances	Eigenvalues
Factor 1 (emotional dimension)	Q7	0.842	30.007	3.001
	Q8	0.784		
	Q3	0.681		
	Q4	0.631		
Factor 2 (physical dimension)	Q1	0.866	12.256	1.226
	Q2	0.856		
	Q9	0.460		
Factor 3 (social dimension)	Q6	0.751	12.088	1.209
	Q10	0.717		
	Q5	0.441		
Cumulative %			54.351	

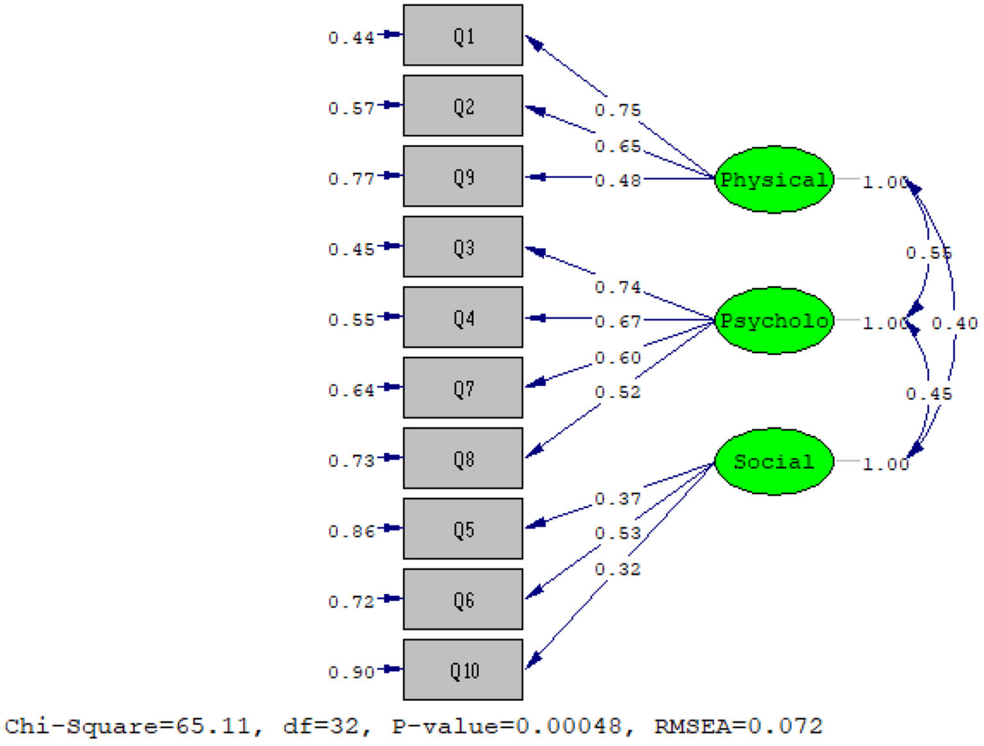

FIGURE 1 | The final structure of the model.

CFI: 0.93; IFI: 0.93; RFI: 0.82; AGFI: 0.9; PGFI: 0.55; RMR: 0.077; standardized RMR: 0.077).

Finally, the results showed that CFA based on the three-factor model extracted from EFA with the obtained data has a good fit (**Figure 1**).

Reliability

For reliability, internal consistency with a 95% CI was performed using Cronbach's alpha coefficient (0.719). The total ICC was 0.812. Furthermore, the ICC of the factor was 0.798, the psychological factor was 0.862, and the social factor was 0.77.

Responsiveness and Interpretability

The result of ANOVA showed that there are no significant differences between the overall POS score and the ages of participants ($P = 0.739$). Moreover, the result of the t-test showed that the overall POS score between men and women was not statistically significant ($P = 0.642$). Furthermore, the ceiling and floor effects for the scale were zero, but it was acceptable because it is below 15%. The MDC percentage was calculated at 8.11%. An MDC of <10% was considered excellent. Therefore, MDC was suitable for this study. The SEM was also calculated at 3.81.

DISCUSSION

The results of the current study deal with the psychometric properties of a 12-item Persian version of the POS in Iran.

Assessing the face and content validity of the scale using the opinions of 10 patients showed that the items are simple and clear. Reviewers judged the measure adequate for tracking palliative care outcomes and authorized coverage beyond the physical issues faced by patients with advanced illness after conducting content validation on the original version of the scale (Hearn and Higginson, 1999). Similar to the results of this study, content validity was confirmed in the Italian version by interviews with patients (Costantini et al., 2016), as well as in the Thai version with CVI report at 0.96 (Pukrittayakamee et al., 2018) and in the Turkish version with CVI report at 0.8–100 (Kocatepe et al., 2020). In addition, the content validity and agreement in the German version of the instrument were evaluated from the perspectives of professional staff and patients in terms of the relevance of the items, forgotten components, and reflection of the patients' true situation. From the perspective of most patients, the tool addressed their real problems, while one-third of them was unsure. Half of the staff were able to communicate with the tool, whereas half were hesitant that the tool could cover patients' real problems. However, the study of Bausewein et al. did not include construct validity due to research limitations (Bausewein et al., 2005).

Confirmatory and exploratory factor analyses of the POS was performed using a sample of 150 and 203 patients with cancer. The fit indices of the Persian version of the POS model based on the main model extracted three psychological, physical, and social factors with a cumulative variance of 54.34%. Goodness-fit indices were confirmed in all three factors. Acceptable values for the RMSEA index (goodness ratios of the mean squared error of the approximation), as well as a 90% CI, should be less than or equal to 0.08. In this study, RMSEA = 0.072 was obtained. The study of Siegert et al. (2010) in the psychometric properties of this scale showed that two factors of "psychological well-being" and "quality of care" were identified, and the three items act separately (Siegert et al., 2010). In the Turkish version of the POS, the compatibility values of the scale ($s = 69$) according to CFA were RMSEA = 0.059, 062, and 0.047 ($P < 0.05$) for the patients, caregivers, and staff, respectively (Kocatepe et al., 2020).

In the research of Harding et al. (2010), the cultural adaptation and psychometric assessment of the POS revealed the tool's excellent qualities, as well as its acceptability and good applicability in clinical settings in Africa. This scale is shorter than similar tools, such as the Missoula-VITAS Quality of Life Index (MVQOLI), and requires less time to complete, which is easy to use in routine clinical evaluations (Harding et al., 2010). The internal stability was acceptable (Cronbach's alpha = 0.719), which is consistent with the results of Eisenchlas et al. (2008).

The ICC of the physical factor was 0.798, the psychological factor was 0.862, and the social factor was 0.77. In the main version of the scale test-retest reliability was acceptable for seven items, and had shown Cronbach's alpha = 0.65 for patients and 0.7 for staff; although they had reported change over time but without statistical significance (Hearn and Higginson, 1999). Internal consistency varies in different studies. The internal consistency in the Thai version scale was 0.9 (Pukrittayakamee et al., 2018), while the Argentine version scale was acceptable at Cronbach's alpha = 0.68–0.69 and 0.66–0.73 for patient and staff ratings, respectively, and test-retest reliability showed very high agreement for every item (>0.8) (Eisenchlas et al., 2008). Although Cronbach's alpha of 0.6 in the African POS had shown moderate internal stability of the scale, test-retest has shown high intra-class correlation coefficients for all items (0.78–0.89) (Harding et al., 2010). Furthermore, Cronbach's alpha of 0.64 in the Turkish POS had shown moderate internal stability of the scale (Kocatepe et al., 2020).

According to the psychometric properties of the Persian version of the POS, the construct validity can be concluded that the mentioned tool in three factors, namely emotional, physical, and social, covers the outcomes of palliative care in patients with cancer. Based on the research of Kocatepe et al. (2020), the Turkish POS is also a valid and reliable tool that can be used with patients, caregivers, and staff members in three dimensions for the evaluation of physical and psychological symptoms, including spiritual, practical, and emotional concerns, as well as psychosocial needs (Kocatepe et al., 2020). Pukrittayakamee et al. (2018) showed that this tool is valid and reliable for use in primary research and clinical setting (Pukrittayakamee et al., 2018).

In this study, the MDC was calculated at 8.11%. A minimum percentage of detectable change of <10% was considered excellent. Therefore, MDC was suitable for this study. The SEM was also calculated at 3.81. These indicators suggested the desirable characteristics of the scale. However, these features have not been studied in similar studies.

Finally, it can be concluded that this tool is suitable for use in clinical settings to assess the symptoms and concerns of the patients and monitor changes in them over time. The tool is concise and takes <10 min to complete. This tool is widely evaluated and is available in various language versions. This tool can be used in a wide range of diseases and different clinical settings such as hospitals, nursing homes, and hospices (Van Vliet et al., 2015).

CONCLUSION

The results of this study showed that the POS tool has a favorable face, content, and structural validity (including EFA and CFA). The reliability of the tool was also reported as desirable. Therefore, due to the need of the Iranian community for palliative care, this tool is suggested to be widely used in clinical, educational, and research in Iran.

Limitation

One of our limitations was the absence of psychometric methods to assess the study's criterion validity. Iranian scholars may utilize these tools to investigate additional validation approaches such as criterion and concurrent validity after psychometric examination of numerous acceptable measures in the area of palliative care.

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

MS, SB, AE, and MR: conceptualization, methodology, draft preparation, and supervision. SM, MP, FH, HS, and AF: writing-reviewing and editing, data collection, and draft preparation. MN, MK, AE, and SB: visualization, investigation, software, data collection, and draft preparation. All authors contributed to the article and approved the submitted version.

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Psychometric Properties of the "Quality of Life in Life-Threatening Illness—Family Carer Version" (QOLLTI-F) in Persian-Speaking Carers of COVID-19 Patients

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Background: Measuring family caregivers' quality of life plays a significant role in improving the quality, efficiency, development, and provision of efficient services for patients with COVID-19. As a result, evaluating the quality of life requires the use of valid and reliable measures that are culturally appropriate. This study was conducted to determine the psychometric properties of the Persian version of the Quality of Life in Life-Threatening Illness–Family Carer Version (QOLLTI – F) in patients with COVID-19.

Methods: This methodological study was carried out in 2021 at Baqiyatallah Hospital in Tehran. After gaining approval from the tool creator, the translation was carried out utilizing the forward-backward approach. Cognitive interviews with 10 family caregivers of COVID-19 patients were used to demonstrate face validity. Moreover, construct validity was identified by performing exploratory factor analysis (EFA) (n = 251), confirmatory factor analysis (CFA) (n = 200), and convergent validation using Zarit Burden Interview (ZBI) questionnaire. For scale reliability, internal consistency and stability were performed using Cronbach's Alpha Coefficient and test-retest, respectively.

Results: 451 family caregivers of patients with COVID-19 were enrolled in this study. Three factors with a cumulative variance of 51.85% were extracted during EFA: (1) Caregiver's physical-emotional status, (2) Satisfaction with the situation, and (3) Caregiver's concerns. CFA showed that the model enjoyed a moderate to a good fit of information (RMSEA: 0.087; NFI: 0.98; CFI: 0.91; IFI: 0.91; GFI 0.89; standardized RMR: 0.070). A significant correlation was found between the Persian version of the ZBI and participants' total scores of QOLLTI – F v3 (r = –0.196, P = 0.000).

Cronbach's Alpha Coefficient = 0.719 and ICC stability reliability = 0.71 of the questionnaire were confirmed.

Conclusion: The Persian version of the QOLLTI – F v3 is a valid and reliable scale that can measure family caregivers' quality of life during a Life-Threatening illness in patients with COVID-19. This instrument may be utilized in clinical trials and research to enhance the quality of life for family carers in Iranian society.

Keywords: quality of life, family caregiver, measurement, validity, reliability, psychometric properties, scale

INTRODUCTION

In the last 2 years, the increase in COVID-19-related deaths has become the greatest threat to human health in the current century (Plohl and Musil, 2020); By November 26, 2021, 5,174,646 million people worldwide have lost their lives. By the widespread outbreak of the SARS CoV-2 virus, World Health Organization [WHO], 2020 has declared a public health emergency worldwide (Afrashteh et al., 2020).

The high prevalence of COVID-19 has negatively affected individuals and communities' physical, social, psychological, and mental functioning which has had significant economic, social, and health consequences (Algahtani et al., 2021). Besides, it has led to increased psychosocial and physical stress such as fear of infection and death, the spread of fake news and rumors, interference in activities of daily living, travel bans and restrictions, social isolation in terms of quarantine or other distancing measures, and occupational and financial problems (Alipour et al., 2020). In this respect, the findings of Wang et al. (2021) demonstrate that COVID-19 has moderate to severe psychological effects that might cause feelings of anxiety, depression, and fear in people. Godinic et al.'s research looks at the effects of the COVID-19-related economic crisis on people's mental and psychological health (Godinic et al., 2020; Ganesan et al., 2021). The unique nature of COVID-19, the need to maintain public safety, social distancing, limited educational and care resources, prolonged illness duration, inadequate knowledge about providing care and being an emerging disease, along with physical problems resulting from the disease might have led to increased isolation and solitude and intensified psychological stress in these patients (Holt-Lunstad et al., 2015). Moreover, compared to other patients, caring for these patients has posed additional challenges to family caregivers (Mirzaei et al., 2020). By managing care, creating proper communication, and educating patients, family caregivers assist minimize psychological and health stresses on the country's health system and empower people to cope with disease (Dixe et al., 2019). These caregivers are often affected by the biological and physiological damage caused by the patient care process, and they are likely to experience reduced social activities, lack of leisure time, and reduced relationships with family and friends (Magliano et al., 2005). In addition to the caring role, most family caregivers are confronted with other challenges such as job management, housekeeping, childcare, concerns about their children's education due to school closures, new economic pressures, high medical costs (Kent et al., 2020), and protecting self and family members against virus spread, all of which affect their quality of life (Dinç and Erdoğan, 2021).

Quality of life is an important measure of a society's health and well-being, and its assessment may lead to the identification of a broad variety of issues that influence people's everyday lives. World Health Organization defines the quality of life as an individual's mental perception of the impact of illness or medical condition on various domains of life, including physical, mental, social, and occupational functioning (Algahtani et al., 2021). COVID-19 pandemic has negatively affected the quality of life of all strata of society, especially family caregivers (Dinç and Erdoğan, 2021). During a pandemic, support systems for family caregivers are essential to enable them to efficiently cope with spiritual and physical problems (Dinç and Erdoğan, 2021).

Therefore, it is necessary to measure family caregivers' quality of life using reliable tools to develop and provide the most efficient services (Axelsson et al., 2020). Quality of Life in Life-Threatening Illness–Family Carer Version was designed by Cohen et al. (2006) in English and French. Its validity and reliability were confirmed. This scale measures caregivers' quality of life and their perception of the patient's life-threatening and challenging condition (Cohen et al., 2006). The instrument has been translated, and its psychometric properties were investigated in various cultures, including Australian (Bradford et al., 2012), Swedish (Axelsson et al., 2020), German (Schur et al., 2014), Spanish (Arias-Rojas et al., 2021), Malaysian (Alnjadat et al., 2014), Indian (Nayak et al., 2014), and Czech (Bužngová et al., 2015). However, a Persian tool to measure family caregivers' quality of life in Life-threatening illnesses has not been designed or psychometrically investigated. Therefore, this study aimed to translate and determine the psychometric properties of the Persian version of the Quality of Life in Life-Threatening Illness–Family Carer Version in the families of patients with COVID-19 (QOLLTI – F).

MATERIALS AND METHODS

Study Design

The present methodological study has investigated Persian version and the psychometric properties of QOLLTI – F v3 scale.

Study Population/Sampling

The target population was family caregivers of the patients with COVID-19 referred to Baqiyatallah Hospital. Availability sampling was used in this study, and sampling was performed in 2021 for a period of 1 month. Inclusion criteria included: at least 18 years of age, caring for a patient with confirmed COVID-19 according to a physician's diagnosis and laboratory tests, willingness to participate in the study by signing a written consent form, literacy of reading and writing in Persian, and no cognitive or mental disorders according to the individual's report. Family caregivers who were reluctant or unable to cooperate in completing the questionnaire or who delivered an incomplete questionnaire were excluded.

Study Instruments

Demographic Information Questionnaire

A researcher-made questionnaire was used to collect demographic information, including gender, age, marital status, education level, job status, and satisfaction with monthly income.

Quality of Life in Life-Threatening Illness–Family Caregiver Version 3

This scale was designed by Cohen et al. (2006), and its validity and reliability were confirmed. The present scale includes 16 items and seven subscales, including Carers Own State (5 items), Relationships (2 items), Carers Outlook (3 items), Quality of Care (2 items), Patient Condition (1 item), Finances (1 item), and Environment (2 items). Scale scores range from 0 to 10. The overall score of scale is calculated based on the average score of 7 subscales. The minimum and the maximum scores are 0 and 160, respectively. The scale's validity was confirmed *via* content and construct validity, and its reliability was obtained by calculating the internal consistency and Cronbach's alpha coefficient of 0.857. The test stability was reported to be 0.77 for the first and second tests and 0.80 for the second and third tests using the test-rest method. ICC for seven subscales was reported to be 0.50–0.79 (Cohen et al., 2006).

Zarit Burden Interview

Zarit Burden Interview (ZBI) is the most widely used tool to assess the care burden of family caregivers, designed by Zarit et al. (1986) (Bédard et al., 2001; Yap, 2010). This questionnaire consists of 22 items and assesses caregivers' individual, social, emotional, and financial dimensions. The items are rated on a 5-point Likert scale ranging from 0 = never, 1 = rarely, 2 = sometimes, 3 = often, 5 = always. Each individual's score is between zero and 88 (Zarit et al., 1986). A higher score indicates a greater psychological burden. Scores in the range of 61 to 88 indicate severe, 31–60 moderate, and less than 30 mild care burden. The validity and reliability of this questionnaire have been investigated by Navidian et al., according to Iranian culture. The Persian version of this tool has desirable psychometric indices. Besides, the test-retest reliability coefficient of the intra-cluster correlation coefficient was 0.94 at a time interval of 2 weeks (Navidian and Bahari, 2008).

Translation Procedure

The forward-backward translation method was used. After obtaining permission from the developer in the forward translation stage, the original English version of the QOLLTI-F was translated into Persian by two specialized English translators, according to the International Quality of Life Assessment (IQOLA) protocol (Bullinger et al., 1998). Afterward, two versions of the translated scale were reviewed in a meeting with researchers in the incorporation stage. Finally, an initial joint translation was obtained as the researchers reached a consensus. In the backward translation stage, the joint Persian translation prepared in the previous stage was translated into English by two native speakers fluent in Persian and English, and an English version was obtained. Two English versions prepared in the previous step were sent to the tool developer to be compared. The developer compared the provided questionnaire to the original version conceptually and approved it.

Face Validity

Cognitive interviews were used to assess the qualitative face validity following the translation process. A cognitive interview is conducted to identify the source of error in the scale by focusing on respondents' cognitive process when completing the scale (Willis, 2004). Furthermore, ten family caregivers with diverse economic, social, and education levels were interviewed. They were requested to rate the legibility, clarity, and structure of the items, ease of comprehension, item difficulty, confusing words, item classification, ease of responding, language forms, and wording. Subsequently, the modifications were applied in the Persian version of QOLLTI-F.

Construct Validity

Exploratory factor analysis (EFA), confirmatory factor analysis (CFA), and convergent validity methods were used to assess the construct validity of this scale.

Exploratory Factor Analysis

Exploratory factor analysis is used to discover the underlying structure of a relatively large set of variables. The minimum sample size required for EFA is 3–10 participants per item (Kellar and Kelvin, 2013). To assess EFA, 251 family caregivers of patients with COVID-19 were enrolled in the study using the convenience sampling method. Moreover, Keiser-Meyer-Olkin (KMO) and Bartlett's tests were performed to evaluate the sample adequacy and suitability. A value of KMO closer to one is more suitable for factor analysis; however, a score greater than 0.5 is generally acceptable, and a score greater than 0.7 is more appropriate (Ebadi et al., 2017). Bartlett's test with a significance level below 0.05 is acceptable (Kaiser, 1974; Hutcheson and Sofroniou, 1999). Favorable results of KMO and Bartlett's tests suggest a favorable correlation matrix for factor analysis (Mohammadbeigi et al., 2015). The association between each component and each questionnaire item is known as factor loading. The association must be suitable for a question to be retained. The minimum factor loading in this study was considered 0.3. In case that factor loading is less than 0.3, the correlation between the factor and the item is weak (Hair, 2009; Rashidi Fakari et al., 2020). For factor extraction and interpretation, the principal components analysis (PCA) and the PROMAX rotation methods were used, respectively (Samitsch, 2014).

Confirmatory Factor Analysis

The extracted factors were assessed using CFA following EFA (Waltz et al., 2010). Afterward, 200 family caregivers were included in the study to evaluate the CFA. CFA was performed using Partial Least Squares to assess the fit of the proposed model with the data. In general, in CFA, the correlation between latent factors and measurable variables is clear; this way, the significance, and intensity of the correlation are determined. Model fit indicators in CFA are classified into three general categories (1) Absolute fit: root mean square error of approximation (RMSEA), standardized root mean square residual (SRMR), goodness-of-fit index (GFI), and Chi-square, (2) Comparative fit: comparative fit index (CFI), incremental fit index (IFI), RFI, normal fit index (NFI), TLI, and (3) Parsimonious fit: PCFI, PNFI, AGFI, AIC.

Convergent Validation

The last method to assess the construct validity was Convergent validation. Respondents simultaneously completed the ZBI and the Persian version of the QOLLTI-F. To confirm this method, the correlation of QOLLTI-F with the ZBI was measured using a correlation coefficient (Krabbe, 2016).

Reliability

To assess the reliability of the scale, two methods of determining Internal Consistency and Stability were used. Cronbach's Alpha Coefficient was calculated to measure the internal consistency of the tool. Cronbach's alpha between 0.8–0.7 indicates acceptable and sufficient internal consistency (Rattray and Jones, 2007). To determine the tool stability, the test-retest method with a sample size of 30 was used. The retest was performed at a time interval of 14 days, and the scores obtained in these two stages were compared using the Intra-cluster correlation index (ICC). ICC index greater than 0.80 is assumed as the desired stability (De Boer et al., 2004). The total item correlation was also examined. We assessed the association between each item and the scale's overall score before deciding whether or not to keep the questions. Questions having a correlation of less than 0.3 were eliminated from the analysis (Stevens, 2012).

Ethical Consideration

The permission to conduct the study was obtained from the ethics committee of Baqiyatallah University of Medical Sciences (ethics code: IR.BMSU.BAQ.REC.1400.48). After obtaining permission from the tool developer *via* email, the translation was performed. Before initiating the research, study participants were informed of the study objectives, and they were recruited after signing written informed consent. The participants were ensured of data confidentiality and the right to withdraw from the study at any stage.

Data Analysis

SPSS software version 22 and LISREL version 8.8 were used for data analysis. In all analyses, the significance level was considered $P < 0.05$.

RESULTS

Socio-Demographic

Among 451 participants, 274 (60.8%) were male, and 177 (39.2%) were female, their mean age was 41.38 ± 13.62. **Table 1** shows the mean score of quality of life, based on demographic characteristics. There was a statistically significant relationship between the patient's marital status ($P = 0.01$) and the income with his/her quality of life ($P = 0.04$). However, no statistically significant relationship was found between gender ($P = 0.01$), marital status ($P = 0.89$), level of education ($P = 0.65$), and job status ($P = 0.83$).

Translation Procedure

After the translation and verification process by Robin Cohen, a Persian questionnaire with one phrase about family caregivers' overall quality of life and 16 items under seven subscales were obtained.

Face Validity

Face validity was verified using ten family caregivers. The items did not change, while assessing face validity due to their simplicity and clarity.

TABLE 1 | Mean score of quality of life, based on demographic characteristics.

Variable	*n*	%	Mean (SD)	Statistical test	Results
Gender					
Male	274	60.8	96.95(23.12)	Independent *T*-test	$P = 0.897$
Female	177	39.2	95.42(2270)		$T = 0.691$
Age					
0–20	11	2.4	88.09(21.51)	One way ANOVA	$P = 0.2$
21–40	240	53.2	95.25(23.50)		$F = 1.59$
<40	200	44.3	98.13(22.26)		
Marital status					
Single	122	27.1	93.97(26.41)	One way ANOVA*	$P = 0.01$
Married	315	69.8	96.56(21.34)		$F = 4.16$
Divorced/widowed	14	3.1	112.42(19.76)		
Education					
Elementary	31	6.9	99.67(24.20)	One way ANOVA	$P = 0.65$
Diploma	148	32.8	94.70(22.69)		$F = 0.54$
Bachelor's	187	41.5	97.16(24.16)		
Master's/Ph.D.	85	18.8	96.24(20.17)		
Income					
High	48	10.6	102.89(24.28)	One way ANOVA*	$P = 0.04$
Average	268	59.4	96.66(21.78)		$F = 3.09$
Dissatisfied	135	29.9	93.43(24.32)		
Job status					
Employed	243	53.9	96.92(24.15)	One way ANOVA	$P = 0.833$
Unemployed	124	27.5	95.97(22.69)		$F = 0.183$
Retired	84	18.6	95.28(19.68)		

**Post hoc analysis show that there is statically significance between single and divorce (p = 0.012) and high and dissatisfied income (p = 0.037).*

TABLE 2 | Exploratory Factor Analysis of the Persian Version of the QOLLTI – F v3.

Factor	Items	Factor loading%	Variance
Factor 1	Q6	0.869	29.58
	Q7	0.816	
	Q5	0.795	
	Q8	0.707	
	Q11	0.527	
Factor 2	Q13	0.839	13.05
	Q12	0.824	
	Q1	0.571	
	Q9	0.532	
	Q10	0.474	
	Q2	0.473	
Factor 3	Q14	0.725	9.21
	Q15	0.713	
	Q16	0.645	
	Q4	0.567	
	Q3	0.539	
Cumulative%			51.85

Construct Validity

The three methods of EFA, CFA, and convergent validity were employed to assess the construct validity of scale.

Exploratory Factor Analysis

A KMO value was found to be 0.832, and Bartlett's test of sphericity was significant (X^2 = 2222.856, df = 120, *p* = 0.000). Three factors were extracted and labeled since they described 51.85% of the total variance of family caregivers' quality of life (**Table 2**).

Confirmatory Factor Analysis

In the CFA, the model showed a good fit. The indices examined for goodness fit included: NFI = 0.98, RMSEA = 0.087, GFI = 0.89, standardized root mean SRMR = 0.070, CFI = 0.91, and IFI = 0.91. The results of the CFA are provided in **Figure 1**

Convergent Validity

For the convergent validity, a significant correlation was found among participants' total scores of QOLLTI – F v3 and Persian version of ZBI (*r* = –0.196, *P* = 0.000). In other words, the higher the burden of care, the lower the quality of life of the family caregiver will be.

Reliability

For scale reliability, internal consistency was obtained to be 0.719 using Cronbach's Alpha Coefficient. Besides, stability was confirmed using the test-retest method. Pearson correlation coefficient was reported to be 0.71.

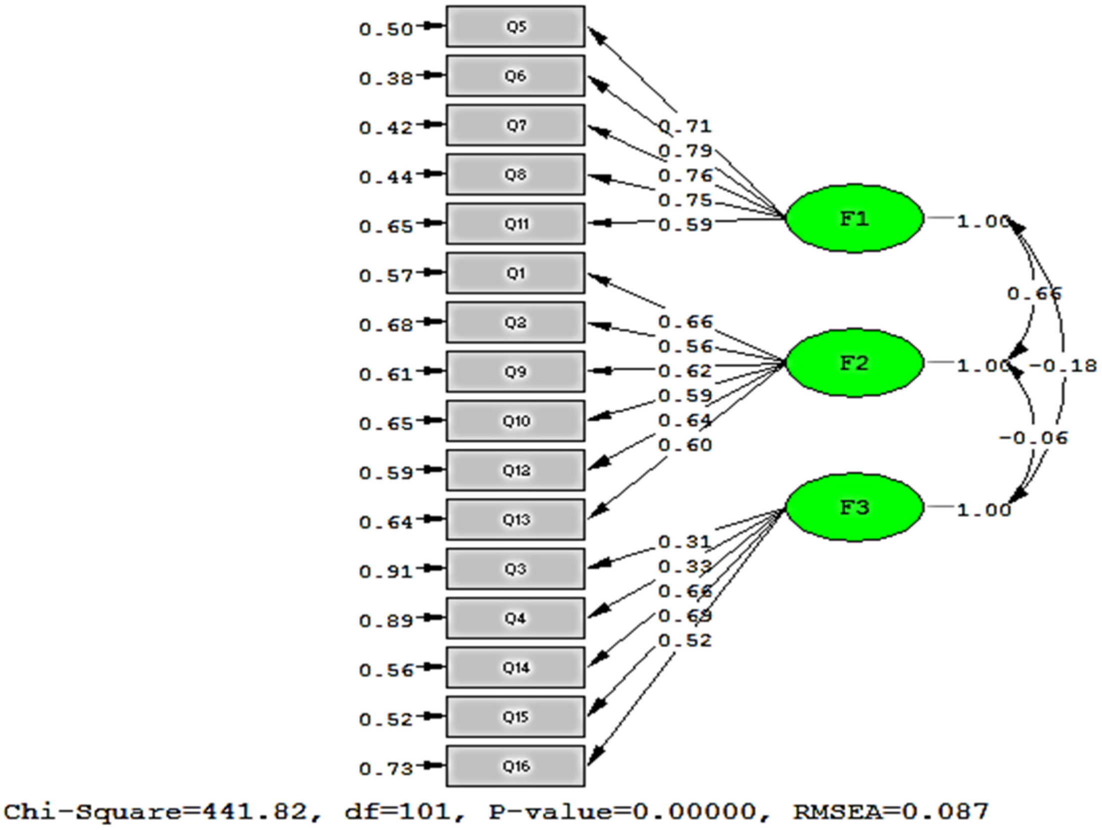

FIGURE 1 | The final structure of the model.

DISCUSSION

During COVID-19 pandemic, family caregivers' quality of life was severely affected. Family caregivers provide measures associated with the symptom management, treatment, and the patient's physical, psychological, and social care (Arias-Rojas et al., 2021). Therefore, addressing caregivers' problems is essential to provide appropriate treatment and social support to patients and ensure the continuity of care (Karimirad et al., 2018). Consequently, QOLLTI-F is a valid tool to assess family caregivers' quality of life. The present study was conducted to investigate the psychometric properties of the Persian version of the QOLLTI – F.

This study discreetly performed the translation process until a final Iranian version was obtained. Similar to the results of this study, the face validity of the original English version of scale was assessed by Cohen et al. with a sample size equal to 30 caregivers (Cohen et al., 2006). In addition, face validity was assessed in the German version (Schur et al., 2014) by 30 caregivers, the Latin American–Spanish version (Arias-Rojas et al., 2021) by 21 caregivers, the Swedish version (Axelsson et al., 2020) by 15 patients and nine caregivers, and in the Malaysian version (Alnjadat et al., 2014) by 10 caregivers, which indicated that the items were straightforward and clear after being translated.

The present study used EFA, CFA, and convergent validation to confirm the construct validity using the ZBI. Regarding the number of extracted factors (3 factors) following EFA, the present study is consistent with Arias-Rojas et al.'s study (Arias-Rojas et al., 2021), conducted to prepare the American–Spanish version. However, the result of EFA in this study was inconsistent with the study by Schur et al. (2014) to develop the German version (4 factors extracted), the study by Alnjadat et al. (2014) in preparing the Malaysian version (7 factors extracted), and the study by Cohen et al. (2006) to prepare the original version of the scale (7 factors extracted). This difference is probably due to the disease nature in the studied populations and sample size. In contrast to the present study, in the studies by Alnjadat et al. (2014) and Schur et al. (2014), no CFA was performed; however, the researchers in this field recommend performing this stage of construct validity in future studies.

Pearson correlation between two scales (QOLLTI-F-V3 scale and the ZBI) indicated an appropriate correlation and confirmed the convergence validity of this scale. In other words, the higher the burden of care, the lower family caregivers' quality of life will be.

Similar to the present study, in the study by Schur et al. (2014) the Integrative Hope Scale (IHS) was used to assess concurrent validity. Besides, the Hospital Anxiety and Depression Scale (HADS) and the Subjective Carers Burden questionnaire were used to assess discriminant validity, revealing a significant relationship between the two scales.

The present study indicated the appropriate reliability (Cronbach's alpha and Test-retest) of the scale. This result was in line with the results of the original version of the QOLLTI-F scale with reliability (Cronbach's alpha = 0.86) and stability (Test-retest = 50–0.79) (Cohen et al., 2006). Furthermore, the results of the present study regarding reliability were in agreement with the results of the study conducted by Schur et al. (2014) to localize the German version (Cronbach's alpha = 0.83) and stability (Test-retest = 0.92) (Schur et al., 2014), the study by Alnjadat et al. (2014) to localize the Malaysian version (Cronbach's alpha ≥ 0.74), and the study by Arias-Rojas et al. (2021) to localize the Latin American–Spanish version of QOLLTI-F scale (Cronbach's alpha = 0.83) and stability (Test-retest = 0.87).

To conclude, according to the psychometric properties of tool in the Persian version, it can be stated that it is an excellent scale to be used to measure family caregivers' quality of life. Another advantage of the tool is its conciseness, which requires ten minutes to complete. This tool was widely validated, and its different versions are available in various languages.

Limitations

So far, an adequate number of tools in the field of palliative care in Iran have not been made or psychometric. As a result, one of our constraints was the absence of psychometric methods to assess the study's criterion validity. Another disadvantage was that this research only included family caregivers of COVID-19 patients. There are other types of life-threatening illnesses, such as cancer, advanced heart failure, chronic obstructive pulmonary disease, in which the quality of life may be affected dissimilarly. The researchers recommend that this study be performed on other patients who use palliative care services. In addition, future studies are needed to investigate this new structure using other tests to evaluate concurrent and discriminant validity and fully validate the QOLLTI-F in the Persian language.

CONCLUSION

QOLLTI – F is a valid and reliable questionnaire to measure the family caregiver's quality of life during a life-threatening illness. Therefore, it can be used in clinical evaluation and research to improve family caregivers' quality of life in Iranian society. This tool can be used in various diseases and medical centers, including hospitals, nursing homes, and hospice centers.

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

AF: conceptualization, methodology, draft preparation, and data collection. AE and MR: methodology and draft preparation.

SH: methodology and draft preparation. MM: supervision, data collection and draft preparation. AK, MJ, BA, and MSR: data collection and draft preparation. SB: supervision, conceptualization, methodology, draft preparation and data collection. All authors contributed to the article and approved the submitted version.

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Screening for Distress in Oncological Patients: The Revised Version of the Psychological Distress Inventory (PDI-R)

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Background: Psychological research in oncological settings is steadily increasing and the construct of psychological distress has rapidly gained popularity—leading to the development of questionnaires aimed at its measurement. The Psychological Distress Inventory (PDI) is one of the most used instruments, but its psychometric properties were not yet deeply evaluated. The present studies aimed at investigating the psychometric properties of the PDI (Study 1) and providing a revised version of the tool (Study 2).

Methods: Oncological outpatients were enrolled at the Department of Medical Oncology of the Presidio Ospedaliero of Saronno, ASST Valle Olona, Italy. For the first study ($N = 251$), an Exploratory Graph Analysis was used to explore the item structure of the PDI. In the second study ($N = 902$), the psychometric properties of the revised PDI (PDI-R) were deeply assessed.

Results: Study 1 showed that the PDI has a not clear structure and it should be reconsidered. On the opposite, Study 2 showed that the revised version (PDI-R) has a solid factorial structure, it is invariant across gender and age, and it has good psychometric properties.

Conclusion: Results suggest that the PDI-R is a reliable measure of psychological distress in different samples of oncological patients, with stronger psychometric properties than the original version. Its use in the clinical and research field is therefore recommended to improve the quality of both assessment and treatment of psychological distress in patients with oncological problems.

Keywords: distress, palliative care, distress thermometer, psychological distress, oncology, psycho-onchology, psycho-oncological distress, psycho-oncological care

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INTRODUCTION

Cancer disease is one of the most common chronic diseases (Crocetti et al., 2006; Jemal et al., 2006) and, with about 9.6 million deaths per year (Ferlay et al., 2019; Teo et al., 2019), represents one of the major causes of death worldwide (WHO, 2014). In Italy, in 2019 cancers alone accounted for 29% of deaths, the most frequent types being breast (53,500), colorectal (49,000), lung (42,500), prostate (37,000), and bladder (29,700) (AIRTUM Working Group, 2021).

Despite the advances in the medical field, the diagnosis of cancer brings with it physical changes associated with the disease itself or to treatment side effects and certainly represents a stressful life event that may induce psychological distress in both patients and their loved ones (Norton et al., 2004; Sellick and Edwardson, 2007; Rossi et al., 2021)—also significantly altering the quality of their relationship (Mannarini and Boffo, 2014; Mannarini et al., 2017; Saritas et al., 2017; Catania et al., 2019; Granek et al., 2019; Rakici and Karaman, 2019).

Negative emotions might relate to the threat to life, and the uncertainty for both the treatment outcomes and the entailed suffering (Peck, 1972; Maguire, 1985; Holland, 1992; Amadori et al., 2002; Grassi and Riba, 2012; Wise et al., 2013; Clerici and Veneroni, 2014)—and studies reveal that at least one-third of the individuals diagnosed with cancer report high levels of psychological distress; which increase as the disease worsens (Carlson and Bultz, 2003; Sellick and Edwardson, 2007).

Psychological distress in the oncological setting corresponds to the cancer-related totality of emotions and feelings experienced by patients that may affect their ability to cope with cancer itself (Holland and Bultz, 2007; Compen et al., 2018). Indeed, evidence exists for the association between high levels of distress and decreased quality of life (Derogatis et al., 1983; Maguire, 1985; Fallowfield, 1988; Fallowfield et al., 2001; Riba et al., 2019), adherence to treatment, self-management (Newell et al., 1998; Partridge et al., 2002; Spiegel and Giese-Davis, 2003; Moore, 2010; Granek et al., 2019; Oliveri et al., 2019; Panzeri et al., 2021a), and survival in patients with oncological problems (Trask et al., 2002; Kwak et al., 2013; Ng et al., 2017). Further, Bultz and Carlson (2006) considered psychological distress a fundamental marker of wellbeing and—due to its reciprocal influence with the physical status of the sufferers (Zabora et al., 1997, 2001; Carlson et al., 2004; Louison et al., 2019; Panzeri et al., 2021b; Rossi Ferrario et al., 2021)—a key indicator of the patient's global health (Bultz and Carlson, 2006) to the point of being listed as the *sixth vital sign* to be investigated in medicine (Grassi and Riba, 2012; Wise et al., 2013; Riba et al., 2019) along with blood pressure, temperature, heart rate, respiratory rate, and pain (Rose and Clarke, 2010; Cutillo et al., 2017).

However, this construct is still is often misinterpreted in literature and confused with other terms, such as «*symptom distress*» (Ridner, 2004)—which corresponds to patients' discomfort related to the perceptions of their symptoms (McCorkle and Young, 1978). Moreover, it is also seriously underreported in the oncological field (Brain et al., 2006).

Physicians, oncologists, and psycho-oncologists should, therefore, properly screen psychological distress levels in patients with cancer at various (treatment) stages (Carlson and Bultz, 2003; Jacobsen, 2007). To facilitate its measurement, the National Comprehensive Cancer Network® (NCCN®) guidelines have indicated that psychological distress may be thought of in terms of (a conjunction of) anxiety and depression (Trask et al., 2002). Massé (2000) has attempted to identify the components of psychological distress and his results show that it consists of a change from a baseline emotional state to the experience of anxiety, depression, aggressiveness, self-depreciation, and demotivation (Massé, 2000). Indeed, when a clinician diagnoses cancer it is generally not possible to make an accurate prognosis; the patient has no guarantee that treatment will restore health. This situation increases the fear of worsening and death. The patient feels hopeless and unable to adopt behaviors useful for the improvement of health. These feelings of total helplessness contribute to the development of depressive symptoms (Rosselli et al., 2015). Furthermore, Pandey et al. (2007) pointed out that anxiety and depression are highly associated with distress in cancer patients to the point of supporting the idea of an overlap between these constructs (Pandey et al., 2007).

Accordingly, studies show that 23.4% of cancer patients report anxiety symptoms which worsen when the cancer symptoms appear, during examinations, and at diagnosis (Stark and House, 2000; Ng et al., 2017; Naser et al., 2021). Moreover, the presence in oncological patients of depressive symptoms ranges from 8 to 24% in non-palliative-care treatment during or after intervention (Krebber et al., 2014). Still, the presence of depressive symptoms might differ according to the cancer type, how these symptoms are evaluated, and the intervention phase—with a higher prevalence in more severe patients (Pirl, 2004; Krebber et al., 2014; Naser et al., 2021). Starting from this background, numerous researches have aimed to create and validate psychological tools for the assessment and measurement of distress in patients with organic pathologies (Carlson and Bultz, 2003; Herschbach et al., 2004; Mitchell, 2010).

However, the Psychological Distress Inventory (PDI, originally developed and validated in Italian context) (Morasso et al., 1996), one of the most worldwide used instruments for the screening and assessment of psychological distress in oncology (Vodermaier et al., 2009; Muzzatti and Annunziata, 2012), was validated without an in-depth evaluation of its psychometric properties, (e.g., factorial structure, measurement invariance, etc.). Indeed, it provides encouraging, albeit incomplete, information on reliability and criterion, concurrent, and discriminant validity but no data on construct validity (Muzzatti and Annunziata, 2012).

Notably, a screening tool that does not have strong and established psychometric properties may lead to misleading results. However, an even more serious problem might be represented by the measurement biases, which may, in turn, lead to underestimation of the measured problem or misdiagnosis—and therefore increase the patient's suffering (Chad-Friedman et al., 2017). Given the strong impact of distress on health as well as on (also) medical care of cancer patients (DiMatteo et al., 2000), it is therefore essential to have well-established, well-structured, reliable, and that can be used in samples with different characteristics.

Consequently, the first goal (Study 1) of this study is to explore the psychometric properties (i.e., factor dimensionality) of the Psychological Distress Inventory (PDI) (Morasso et al., 1996). On the basis of results of the Study 1, the second aim (Study 2) is to develop and extensively assess the psychometric properties (i.e., structural validity, measurement invariance, screening ability, etc.) of a shortened version of the PDI: the Psychological Distress Inventory—Revised (PDI-R).

STUDY 1. EXPLORING THE DIMENSIONAL STRUCTURE OF THE PSYCHOLOGICAL DISTRESS INVENTORY

Materials and Methods

Procedure

An observational research design was used to investigate the psychological distress experienced by oncological outpatients during the first few weeks between the diagnosis of cancer and the first psychological clinical session—in line with the procedure provided by the HuCARE study protocol (Passalacqua et al., 2016; Marconi et al., 2020; Caminiti et al., 2021; Rossi et al., 2021).

This study was approved by the Ethics Committee of the Ospedale di Saronno (protocol n° 23247). All procedures were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Participants

Oncological outpatients were consecutively recruited from the Department of Medical Oncology, Presidio Ospedaliero di Saronno, ASST Valle Olona, in Saronno (Italy).

Inclusion criteria for participating in the study were: (A) having received a diagnosis of cancer within the last 6 months; (B) being an oncological outpatient (C) not being hospitalized for cancer-related problems within the last year, (D) following intravenous therapy for cancer; (E) being 18 years or older; (F) providing informed consent to participate in the study; and (G) being a native Italian speaker. Exclusion criteria were: (A) inability to understand the items of the questionnaire; (B) impossibility to be assessed due to speaking impairments and/or upcoming medical commitments.

A sample of 270 participants was initially assessed. However, 19 subjects were excluded due to missing data/answers. The final sample comprised, therefore, 251 participants: 120 males (47.80%) and 131 females (52.20%), aged from 20 to 86 years (mean = 63.24, *SD* = 12.56). Considering the type of cancer, 30.7% patients had breast cancer, 27.1% patients had lung cancer, 19.9% patients had gastrointestinal cancer, 10.6% patients had urogenital cancer, 4.2% patients had oncohematological cancer, and 7.5% patients had other type of cancer (e.g., head-and-neck cancer or skin cancer). Considering education level, 22.9% patients had an elementary school diploma, 35.4% patients had a middle school diploma, 34.3% patients had a high school diploma, and 7.4% patients had a bachelor/master's degree. Considering civil status, 77.2% patients were either in a relationship or married, 11.4% patients were either separated or divorced, 8.3% patients were widowed, and 3.1% patients were single. Considering working status, 50.1% patients were retired, 27.6% patients were dependent workers, 12.4 patients were entrepreneurs, 3.7% patients were housewifes, 4.2 patients were unemployed, and 2% patients declared "other."

Sample Size Determination

The sample size was calculated *a priori* considering the statistical analysis of this study. However, to date, within the framework of Exploratory Graph Analysis (EGA), no "gold standard" rule for determining the minimum sample size required to correctly estimate model parameters seems to exist (Golino and Epskamp, 2017). Therefore, a minimum sample size of 200 individuals was considered adequate.

Measures

Psychological Distress Inventory

The Psychological Distress Inventory (PDI) was used to measure the degree of psychophysical distress experienced by the sample (Morasso et al., 1996). It represents a self-report questionnaire aimed at detecting distress in individuals suffering from oncological problems by investigating the experience of discomfort related to both emotional and physical domains. The PDI comprises a total of 13 items—with good reliability properties (Morasso et al., 1996)—on a 5-point Likert-type response scale ranging from 1 (*not at all*) to 5 (*very much*). High scores correspond to a high degree of distress perceived by the subject. In this study, Cronbach's alpha was equal to 0.804 and McDonald's omega was equal to 0.846.

Statistical Analyses

The R software (R Core Team, 2017) was used with the following packages: bootnet (Epskamp et al., 2018); corrplot (Wei and Simko, 2017); EGAnet (Golino and Christensen, 2020), igraph (Csardi and Nepusz, 2006), networkTools (Jones, 2020), qgraph (Epskamp et al., 2012) psych (Revelle, 2018), and psychTools (Revelle, 2020).

Preliminary analyses were performed before carrying out the EGA (Christensen et al., 2020b). First, the normality of items, and the presence of excessive correlations ($r > 0.70$) between items, were inspected (Howell, 2013; Tabachnick and Fidell, 2014). Second, for each item, the level of informativeness was evaluated (Mullarkey et al., 2019; Bottesi et al., 2020). An item should be considered as badly informative as its SD is 2.5 SDs below the average of all the items (Mullarkey et al., 2018, 2019; Marchetti, 2019). Third, item redundancy was checked (Christensen et al., 2020a) by using a Unique Variable Analysis (UVA) approach with weighted topological overlap (wTO) method and adaptive alpha. However, as suggested by existing guidelines, possible item redundancies should be carefully evaluated—for example, considering the aims of the study and/or by inspecting the semantic content of the items (Christensen et al., 2020b).

Consequently, an EGA (Golino and Epskamp, 2017; Christensen et al., 2020b) was performed to assess the item clustering (i.e., dimensionality) of the PDI—given its several advantages over traditional exploratory factor-analytic techniques; it provides greater accuracy in identifying the correct number of factors/dimensions (Golino and Demetriou, 2017; Golino and Epskamp, 2017; Christensen and Golino, 2020; Golino et al., 2020a,b). The EGA produces a plot that might be considered as a "visual guide" (Golino and Epskamp, 2017; Golino et al., 2020a; Panzeri et al., 2021c). It displays the correct number of dimensions—by highlighting which items cluster together and their level of association: the thicker is an edge, the strongest is the relationship between item of a specific cluster (dimension/factor) (Mair, 2018; Christensen and Golino, 2020). The Exploratory Graph Analysis (EGA) was carried out by using a 5,000 parametric bootstrap procedure. Moreover, the GLASSO method with polychoric correlations was used to estimate model parameters (Costantini et al., 2015; Epskamp, 2017; Golino and Epskamp, 2017). In addition, the correct number of dimensions were detected by using the *"Louvain community detection algorithm"* (Blondel et al., 2008). It has demonstrated better performances than the Walktrap algorithm (Pons and Latapy, 2006) in recognizing clusters of items/dimensions (Christensen et al., 2020b).

Once the EGA revealed the number of dimensions composing the PDI, the questionnaire and item statistics were explored. First, item stability (IS) was computed to evaluate the proportion of times the original dimension is exactly replicated across bootstrap resamples—thus, assessesing the occurrence of each item within a certain specific dimension (Christensen et al., 2020b). IS ranging from 0 (" = *perfect instability*") to 1 (" = *perfect stability*") and values higher of 0.80 ($IS \geq 0.80$) suggest that the item can be considered "stable" and consistently identified in the dimension (Christensen and Golino, 2019). The contribution of each node to the coherence of the dimensions was then assessed using the standardized node strength—namely, network loadings. It is important to note that they represent partial correlation loadings, and the magnitude of these loadings should therefore be interpreted according to the following benchmarks (Christensen and Golino, 2020; Golino et al., 2020c): small: $\lambda_{EGA} \geq 0.15$; moderate: $\lambda_{EGA} \geq 0.25$; large: $\lambda_{EGA} \geq 0.35$.

The internal consistency of each factor was evaluated with Cronbach's alpha and McDonald's omega (McDonald, 1999; Howell, 2013).

Lastly, correlations between items were assessed using Pearson's correlation coefficient and interpreted using Cohen's (Cohen, 1988) classical benchmarks: $r < 0.10$, trivial; r from 0.10 to 0.30, small; r from 0.30 to 0.50, moderate; $r > 0.50$, large.

Results

Preliminary Analyses

First, as reported in **Table 1**, univariate normality was observed for the large majority of the PDI items. Considering non-normal distributed items, only three of them showed small-to-moderate deviations from normality (item#5, item#9, and item#13). Moreover, none of the bivariate correlations exceeded a critical level ($r \geq 0.70$).

Second, the level of informativeness of each item was tested. Results showed that none of the 13 items of the PDI was badly informative (i.e., $SD_{item} < 2.5$ SD below the mean level of informativeness, $M_{SD} = 1.16 \pm 0.18$)—suggesting that each item of the PDI provides adequate variability across the sample as well as a good level of informativeness.

Third, item redundancy was inspected. The UVA showed possible redundancy between some items of the PDI. Considering the aim of the study—exploring the structure of the PDI and redefining its psychometric properties—no item was removed. However, item redundancies were carefully considered and deeply studied before setting up Study 2.

Exploratory Graph Analysis

As reported in **Figure 1** and **Table 1**, the EGA (5000 bootstrap) clearly identified three dimension/factor solution: median = 3; SE = 0.489; 95%CI [2.040, 3.960]. Moreover, the bootstrapped EGA showed that the probability of a three dimension/factor solution was = 0.682 (68.2%) and the probability of a four-dimension/factor solution was = 0.309 (30.9%).

Item Statistics

As reported in **Table 1** and **Figure 2**, IS revealed that the PDI items were—on average—stable within and between their designated dimension/factor: $Dim\#1_{IS\_replication\_mean} = 0.936$; $Dim\#2_{IS\_replication\_mean} = 0.708$; $Dim\#3_{IS\_replication\_mean} = 0.813$. More in detail, the IS analysis (**Table 1**) showed that most items had a good replication index. Indeed, the items in the first dimension (item#3, item#7, item#9, and item#10) displayed a replication index higher than 0.88; also the items in the second dimension (item#1, item#5, item#11, and item#13) displayed a replication index higher than 0.88. Lastly, the items in the third dimension (item#2, item#6) displayed a replication index higher than 0.99. It should be noted that item#4 (feel tired), item#8 (body image), and item#12 (sexual difficulties) did not achieve the recommended threshold of 0.80 in none of the three aforementioned dimensions.

Then, EGA-based network loadings (λ_{EGA}) were computed to assess the contribution of each node to the coherence of the dimensions. As reported in **Table 1**, each item showed a high association with its more stable dimension/factor. Considering the first dimension/factor, λ_{EGA} ranged from 0.257 (large) to 0.426 (large). For the second dimension/factor, λ_{EGA} ranged from 0.231 (large) to 0.380 (large). For the third dimension/factor, λ_{EGA} ranged from 0.302 (large) to 0.334 (large).

Then, an in-depth examination of the semantic content of the items—grouped according to EGA—led to the labeling of the three dimensions. The first dimension measures internal states of psychological distress: anxiety (item#3), depression (item#7), lack of self-esteem (item#9), and lack of motivation (item#10). The second dimension assesses distress related to relationships and the external world: lack of desire to talk to others (item#1), loneliness (item#5), lack of external interests (item#11), worsening of relationships (item#13). Lastly, the third dimension relates exclusively to reverse items and thus a situation of wellbeing/tranquility (item#2 and Item#6).

STUDY 2—VALIDATION AND AN IN-DEPTH ANALYSIS OF THE REVISED VERSION OF THE PDI (PDI-R)

Materials and Methods

Procedure

According to the HuCARE project protocol (Passalacqua et al., 2016; Marconi et al., 2020; Caminiti et al., 2021; Rossi et al., 2021), the same procedure and inclusion/exclusion criteria of

TABLE 1 | Study 1. Descriptive statistics of items and Exploratory Graph Analysis (EGA) results.

	Descriptive statistics				Item stability (IS)			EGA loadings		
	Mean	***SD***	**Skwn.**	**K**	**Stab#1**	**Stab#2**	**Stab#3**	**Dim#1 $\|\lambda\|_{EGA}$**	**Dim#2 $\|\lambda\|_{EGA}$**	**Dim#3 $\|\lambda\|_{EGA}$**
Item#1	1.47	0.776	1.654	2.318	0.001	0.995	0.000	0.082	0.359	0.087
Item#2*	2.97	0.802	−0.137	0.670	0.002	0.000	0.998	−0.049	0.000	0.334
Item#3	2.22	0.976	0.622	−0.010	0.913	0.006	0.001	0.257	0.014	0.000
Item#4	2.49	1.129	0.455	−0.514	0.784	0.063	0.035	0.139	0.053	0.062
Item#5	1.36	0.774	2.413	5.468	0.079	0.888	0.001	0.163	0.231	0.000
Item#6*	2.73	1.030	0.045	−0.146	0.002	0.000	0.998	0.000	0.016	0.302
Item#7	1.86	0.917	0.971	0.617	0.884	0.021	0.001	0.295	0.203	−0.072
Item#8	1.80	1.061	1.349	1.226	0.281	0.140	0.315	0.100	0.019	−0.181
Item#9	1.33	0.667	2.368	6.244	0.954	0.003	0.002	0.273	0.187	−0.069
Item#10	1.53	0.826	1.579	1.987	0.957	0.001	0.001	0.426	0.032	0.096
Item#11	1.46	0.786	1.686	2.018	0.001	0.996	0.000	0.063	0.380	0.089
Item#12	1.97	1.261	1.001	−0.273	0.062	0.635	0.092	0.064	0.167	0.097
Item#13	1.37	0.760	2.344	5.647	0.005	0.986	0.000	0.151	0.272	0.089

**Reverse score item (not reversed); Skwn, Skewness; K, kurtosis; Stability#(. . .), stability of the item (5,000 replication) on the EGA-based dimension; Dim#(. . .), EGA-based dimension;* $|\lambda|_{EGA}$*, absolute value of the network loading.*

Study 1 were applied. This study was approved by the Ethics Committee of the Ospedale di Saronno (protocol n° 23247). All procedures were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Also in this study, oncological outpatients of the study were still consecutively recruited at the Department of Medical Oncology, Presidio Ospedaliero di Saronno, ASST Valle Olona, in Saronno (Italy).

Participants

An initial sample of 936 oncological outpatients was assessed. However, 34 subjects were excluded from the sample due to missing data/answers. Therefore, the final sample comprised 902 oncological outpatients: 487 males (54%) and 415 females (46%), aged from 31 to 89 years (*mean* = 68.39, *SD* = 9.073). Considering the type of cancer, 34.8% patients had breast cancer, 24.9% patients had lung cancer, 18.8% patients had gastrointestinal cancer, 7.9% patients had urogenital cancer, 3.5% patients had pancreatic cancer, 2.7% patients had oncohematological cancer, and 7.4% patients had other type of cancer (e.g., head-and-neck cancer or skin cancer). Considering education level, 26.9% patients had an elementary school diploma, 32.8% patients had a middle school diploma, 30.8% patients had a high school diploma, and 9.5% patients had a bachelor/master's degree. Considering civil status, 73% patients were either in a relationship or married, 6.8% patients were either separated or divorced, 10.3% patients were widowed, and 9.9% patients were single. Considering working status, 58.8% patients were retired, 20.7% patients were dependent workers, 9.6% patients were entrepreneurs, 5.6% patients were housewifes, 3.8 patients were unemployed, and 1.5% patients declared "other."

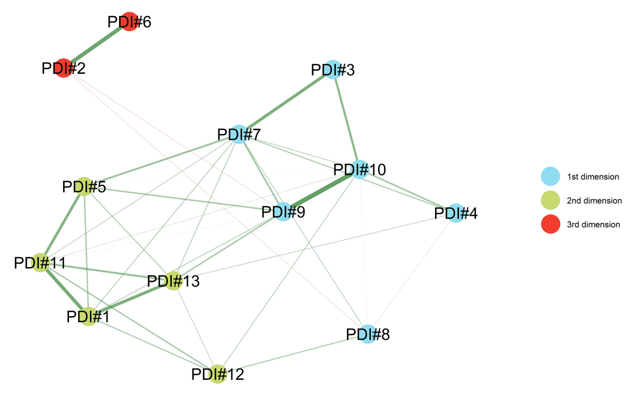

FIGURE 1 | Study 1. Exploratory graph analysis (EGA) of the Psychological Distress Inventory.

Sample Size Calculation

Since the present study aimed to assess psychometric properties of a ("new") questionnaire, the "*n:q* criterion" was used to determine the minimum sample size. In this formula, n is the number of subjects and q is the number of (free) model parameters to be estimated (Hu and Bentler, 1999; Muthén and Asparouhov, 2002; Yu, 2002). A ratio of five subjects per parameter (10:1; $n_{\text{minimum}} = 480$) was guaranteed (Bentler and Chou, 1987; Marsh et al., 1988; Hu and Bentler, 1999; Boomsma and Hoogland, 2001; Muthén and Asparouhov, 2002; Yu, 2002; Flora and Curran, 2004; Tomarken and Waller, 2005).

Measures

Psychological Distress Inventory—Revised

On the basis of the results of Study 1 and following consolidated methodological procedures (Pietrabissa et al., 2020a,b; Rossi et al., 2021), an in-depth analysis of items was performed. This procedure led to the removal of both the two reversed scored items of the original PDI (item#2 and item#6), and the three items that did not achieve IS threshold of 0.80 (item#4,

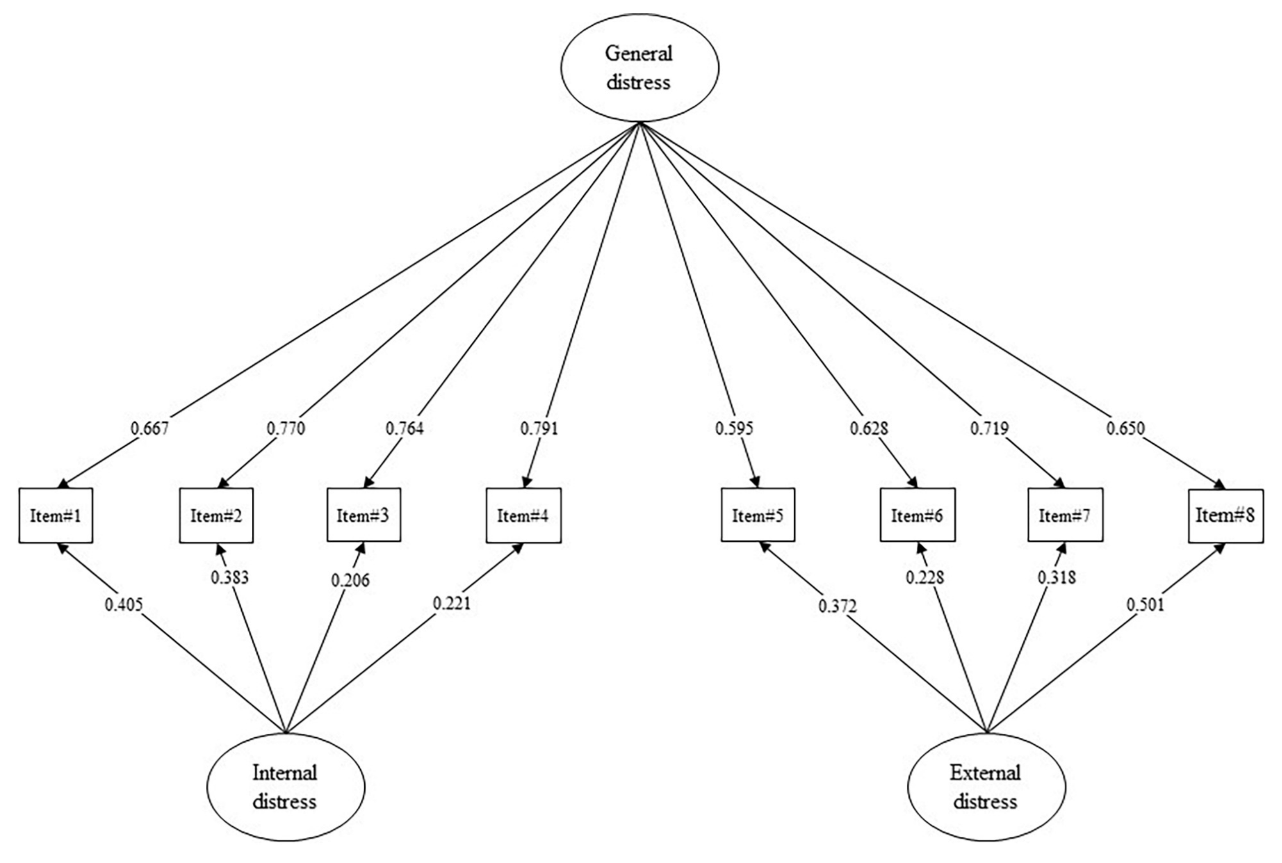

FIGURE 2 | Study 2. Confirmatory factor analysis (CFA) and factor loadings. All *p*-values are less than 0.001. Absolute values of standardized factor loading (|λ|) are reported.

item#8, and item#12). Consequently, on the basis of the results of Study 1, only two dimensions were retained: (A) *"internal"* and (B) *"external"* distress. The first dimension assessed internal states of psychological distress such as anxiety, depression, and lack of self-esteem. The second dimension assesses distress related to relationships and the external world: lack of desire to talk to others, loneliness, and worsening of relationships. Furthermore, considering the semantic content of the items, a hierarchical *"general distress"* dimension was also hypothesized. It is important to underline that the semantic content of the items has not been changed compared to the original version of the PDI provided by Morasso and colleagues (Morasso et al., 1996).

As for the original version of the PDI, the PDI-R relies on a 5-point Likert-type response scale (from 1 = "*not at all*" to 5 = "*very much*") see **Supplementary Appendix A** (English version) and **Supplementary Appendix B** (Italian version). High scores correspond to a high degree of distress perceived by the subject.

The Hospitalized Anxiety and Depression Scale

The Hospitalized Anxiety and Depression Scale (HADS) was used to measure the degree of psychological suffering in oncological patients (Zigmond and Snaith, 1983; Annunziata et al., 2011; Iani et al., 2014). It is a self-report questionnaire—with a bi-factorial structure—aimed at detecting anxiety and depression in hospitalized individuals (as well as outpatients) affected by organic pathology, as well as their perceived distress as the sum of the scores obtained on the anxiety and depression dimensions (Iani et al., 2014). The HADS comprises a total of 14 items—with good reliability properties—scored on a 4-point Likert-type response scale. High scores correspond to a high degree of anxiety, depression, and distress perceived by the subject. Two cut-offs threshold for the HADS total score (HADS-T; $\geq$ 16 = moderate distress; $\geq$ 22 = severe distress) were deemed appropriate in general clinical health settings to distinguish between individuals with no-psychological suffering and those with psychological suffering (Olssøn et al., 2005; Singer et al., 2009; Vodermaier et al., 2009; Stern, 2014). In this study, Cronbach's alpha was equal to: 0.836 for the anxiety subscale (HADS-A); 0.806 for the depression subscale (HADS-D); and 0.887 for the scale's total score (HADS-T).

The Distress Thermometer

The Distress Thermometer (DT) (Bulli et al., 2008; O'Donnell, 2013; Cutillo et al., 2017) is one of the most worldwide used instruments for the measurement of emotional distress in different contexts including oncological settings. The DT is a self-report single-item measure scored on a 10-point Likert scale (from 0 = "*no distress*" to 10 = "*extreme distress*") aimed at detecting psychological and emotive suffering. High scores correspond to a high degree of emotional distress perceived by the subject. Moreover, a cut-off threshold of ≥ 5 (Gil et al., 2005; Grassi et al., 2009; Vodermaier et al., 2009; Donovan et al., 2014) for the DT was deemed appropriate in general clinical health settings to distinguish between individuals with no-psychological distress and those who suffering from psychological distress (Grassi et al., 2009).

Statistical Analysis

R software (R Core Team, 2014, 2017) was used to perform statistical analyses with the following packages: cNORM (Lenhard W. et al., 2018); corrplot (Wei and Simko, 2017); irr (Gamer et al., 2019); lavaan (Rosseel, 2012; Rosseel et al., 2015), plotROC (Sachs, 2017); pROC (Robin et al., 2011), psych (Revelle, 2018), psychTools (Revelle, 2020), and semTools Contributors (2016).

A bi-factor model was specified: 4 items loaded onto the "*internal distress*" latent factor, while 4 items loaded onto the "*external distress*" latent factor, and each item also loaded onto a hierarchical "*general distress*" dimension.

The diagonal weighted least square (DWLS) estimator was used to assess the factorial structure of the PDI-R (Brown, 2015; Kline, 2016; Lionetti et al., 2016). Model fit was assessed by means of the Satorra-Bentler Chi-square statistics (S-Bχ^2), the Root-Mean Square Error of Approximation (RMSEA), the Comparative Fit Index (CFI), and the Standardized Root Mean Residual (SRMR) (Muthén and Muthén, 1998–2017; Hoyle, 2012; van de Schoot et al., 2012; Brown, 2015; Kline, 2016). Moreover, the following cut-off criteria were chosen to evaluate the goodness of fit: (A) statistically non-significance of the χ^2, (B) an RMSEA lower than 0.08, (C) a CFI higher than 0.95, and (D) an SRMR lower than 0.08 (Muthén and Muthén, 1998–2017; Hoyle, 2012; van de Schoot et al., 2012; Brown, 2015; Kline, 2016).

In addition, for a comprehensive evaluation of the factorial structure of the PDI-R, two alternative models were further specified and compared. First, a single factor model was specified: all of the 8 items loaded onto a single distress latent factor. Second, a first-order two-factor model was specified: 4 items loaded onto the "*internal distress*" latent factor, while 4 items loaded onto the "*external distress*" latent factor; thus, the general distress dimension was not specified.

Model evaluations were performed by using the test differences in three fit indices, with the following criteria as cutoffs for model equality: DIFFTEST (equal to $\Delta\chi^2$; p-value > 0.050) and ΔCFI (<0.010) (Cheung and Rensvold, 2002; Millsap and Yun-Tein, 2004; Millsap, 2012). The crossing of the cutoff of two out of three of these indices is evidence of model inadequateness.

Moreover, since the PDI-R is a new questionnaire, items' ability to discriminate subjects with low or high *internal, external*, and *general distress* was tested (Ebel, 1965; Chiorri, 2011). Item discriminant power (IDP) was computed. More in detail, the maximum total score and quartile rank for each subject were calculated. Subsequently, a series of independent sample t-tests—and their effect size (Cohen's d) (Cohen, 1988)—were calculated to assess item discriminating power by using the total score of the scale as a dependent variable and its lowest and highest quartile as grouping variable (Ebel, 1965; Chiorri, 2011). Moreover, item-total correlation (adjusted; i.e., $r_{\text{it}-\text{tot}}$) was also computed (Howell, 2013; Pallant, 2013; Tabachnick and Fidell, 2014).

The internal consistency of each factor was evaluated with Cronbach's alpha and McDonald's omega (McDonald, 1999; Howell, 2013). Convergent validity was assessed with the Pearson correlation coefficient (Tabachnick and Fidell, 2014) and interpreted using the aforementioned Cohen's benchmarks (Cohen, 1988).

Measurement invariance (MI) analyses were also performed to evaluate whether the factorial structure of the PDI-R was invariant between gender (male vs. female) and age (≤ 64 vs. ≥ 65) (Vandenberg and Lance, 2000). According to Meredith (1993) and Millsap (2012), the model structure was tested on each sample independently (Meredith, 1993). If the model fit was adequate in each sample, four nested models were sequentially specified and constrained to equality: the factorial structure (Model 1: Configural Invariance); the factorial structure and item factor loadings (Model 2: Metric Invariance); the factorial structure, item factor loadings, and item thresholds (Model 3: Scalar Invariance); the factorial structure, item factor loadings, item thresholds, and latent means (Model 4: Scalar Invariance); (Meredith, 1993; Vandenberg and Lance, 2000; Millsap, 2012). MI was assessed by using the above-mentioned test differences for model comparisons (Cheung and Rensvold, 2002; Millsap, 2012; van de Schoot et al., 2012).

Test-retest reliability of each scale was estimated on a subsample of 40 oncological patients by using the two-way mixed intraclass correlation coefficient (ICC$_{\text{consistency}}$) (de Vet et al., 2006; Berchtold, 2016; Koo and Li, 2016).

Considering that the PDI-R (as well as the PDI) was conceptualized as a screening tool, a Receiver Operating Characteristics (ROC) curve methodology was used to assess the PDI-R accuracy to discriminate between *non*-distressed and distressed patients (Zhou et al., 2002; Pepe, 2003; Ising et al., 2012; Savill et al., 2018). More in detail, the HADS-T score cut-offs (HADS-T ≤ 15 vs. HADS-T ≥ 16 and HADS-T ≤ 21 vs. HADS-T ≥ 22) and the DT cut-off (DT ≤ 4 vs. DT ≥ 5) were used as external criterion variable and the PDI-R total score was used as the dependent variable. The global accuracy-validity of the PDI-R was estimated with the area under the ROC curve (AUC; 5000 stratified bootstrap resamples)—interpreted using the Swets' benchmarks: AUC = 0.50, null; AUC from 0.51 to 0.70, small; AUC from 0.71 to 0.90, moderate; AUC from 0.91 to 0.99, high; and AUC = 1.00, perfect accuracy (Zweig and Campbell, 1993; Swets, 1998). Moreover, sensibility (Se) and specificity (Sp) were computed (Zhou et al., 2002; Pepe, 2003).

Lastly, as a supplemental analysis, according to previous studies procedures (Dakanalis et al., 2013), a "general sample" of patients with cancer was created by merging the sample of Study 1 and the sample of Study 2 ($N_{\text{total}} = 1153$). Thus, according to the procedure described by Gary et al. (2021), normative scores (T-scores) of the PDI-R were computed as well as the distribution percentiles of its total score (Lenhard A. et al., 2018; Gary et al., 2021).

Results

Structural Validity

The PDI-R showed an excellent fit to the data. The Chi-square statistic resulted to be not statistically significant: S-Bχ^2 (12) = 17.913; $p = 0.118$. Moreover, all the other fit indices revealed a good fit to the data: the RMSEA = 0.023; 90%CI [0.000, 0.044]; p(RMSEA < 0.05) = 0.984, the CFI = 0.999, the SRMR = 0.022. As reported in **Table 2** and **Figure 2**, all the items' loadings were statistically significant and ranged from | 0.206| (item#3, internal distress) to | 0.791| (item#4, general distress).

Moreover, this factor solution was compared with different competing models (Muthén and Muthén, 1998–2017; Brown, 2015). As reported in **Table 3**, model comparisons revealed the superiority of the proposed solution. Consequently, the bi-factor model solution was maintained to perform the following analysis.

Psychometrics Properties

The IDP analysis showed that 8 items of the PDI-R discriminated well between subjects with low or high internal and external distress (**Table 2**). The discrimination parameter t_i ranged from |20.34| (item#6—*external distress*) to |42.33| (item#2—*internal distress*), with an associated effect size (Cohen's d) ranging from 1.89 to 4.22, respectively. Also, the item-total correlation (adjusted) revealed a moderate-to-strong association between each item and the PDI-R scores.

Reliability analysis revealed satisfying results. Indeed, for the *internal distress* subscale, the Cronbach's alpha was equal to 0.776 and the McDonald's omega was equal to 0.842. The *external distress* subscale showed a Cronbach's alpha equal to 0.754 and a McDonald's omega was equal to 0.800. The *general distress* scale showed a Cronbach's alpha was equal to 0.853 and a McDonald's omega was equal to 0882.

Large correlations were found between the *internal distress* scale and the HADS-A scale ($r = 0.724$, $p < 0.001$); the HADS-D scale ($r = 0.630$; $p < 0.001$), the HADS-T ($r = 0.741$; $p < 0.001$), and the DT ($r = 0.595$, $p < 0.001$). Also, moderate-to-large correlations were found between the *external distress* scale and the HADS-A scale ($r = 0.583$, $p < 0.001$); the HADS-D scale ($r = 0.665$; $p < 0.001$), the HADS-T ($r = 0.681$; $p < 0.001$), and the DT ($r = 0.490$, $p < 0.001$). Lastly, large correlations were found between the *general distress* scale and the HADS-A scale ($r = 0.713$, $p < 0.001$); the HADS-D scale ($r = 0.705$; $p < 0.001$), the HADS-T ($r = 0.775$; $p < 0.001$), and the DT ($r = 0.576$, $p < 0.001$). In addition, the internal and external distress subscales revealed a large correlation: $r = 0.685$, $p < 0.001$.

Test-retest reliability showed satisfying results: the two-way mixed ICC was equal to 0.647, 95%CI [0.385, 0.797], for the internal distress scale, to 0.699, 95%CI [0.476, 0.827], for the external distress scale, and 0.685, 95%CI [0.452, 0.819], for the general distress scale.

Measurement Invariance

Gender (Male vs. Female)

Model Male. The Chi-square statistic resulted to be not statistically significant: S-Bχ^2 (12) = 13.490; $p = 0.334$. Moreover, all the other fit indices revealed a good fit to the data: the RMSEA = 0.016; 90%CI [0.000, 0.050]; p(RMSEA < 0.05) = 0.948, the CFI = 1.000, the SRMR = 0.025.

Model Female. The Chi-square statistic resulted to be not statistically significant: S-Bχ^2 (12) = 10.860; $p = 0.541$. Moreover, all the other fit indices revealed a good fit to the data: the RMSEA = 0.000; 90%CI [0.000, 0.046]; p(RMSEA < 0.05) = 0.968, the CFI = 1.000, the SRMR = 0.026.

TABLE 2 | Study 2. Item descriptive statistics, item psychometric properties, and factor loadings (λ) of the confirmatory factor analysis (CFA).

	Descriptive statistics				**IDP**		***Adj*** **$r_{(it-tot)}$**			**CFA**		
	Mean	***SD***	**SK**	**K**	***t***	***d***	**Internal**	**External**	**General**	**Internal**	**External**	**General**
Item#1	2.53	1.099	0.420	−0.441	−32.29	3.24	0.558		0.544	0.405		0.667
Item#2	2.29	1.079	0.565	−0.343	−42.33	4.22	0.638		0.560	0.383		0.770
Item#3	1.74	1.065	1.321	0.825	−23.69	2.34	0.547		0.530	0.206		0.764
Item#4	1.77	0.969	1.146	0.674	−25.93	2.57	0.575		0.651	0.221		0.791
Item#5	1.72	0.992	1.280	0.766	−24.78	2.31		0.532	0.589		0.372	0.595
Item#6	1.66	1.011	1.500	1.460	−20.34	1.89		0.475	0.618		0.228	0.628
Item#7	1.73	1.067	1.385	0.944	−26.37	2.45		0.591	0.649		0.318	0.719
Item#8	1.76	1.051	1.250	0.628	−30.97	2.88		0.609	0.608		0.501	0.650

All test are statistically significant with $p < 0.001$. Skwn, Skewness; K, kurtosis; IDP, item discriminant power; t, t-test; d, Cohen's d; Adj $r_{(it-tot)}$, item-total correlation (adjusted).

TABLE 3 | Study 2. Model comparison.

	S-Bχ^2 (*df*)	**RMSEA**	**CFI**	**Comparison**	**DIFF-TEST**	**|ΔCFI|**
Model 1: bi-factor model	17.913 (12)	0.023	0.999			
Model 2: single factor model	175.593 (20)	0.082	0.987	2 vs. 1	157.68***	0.013
Model 3: two factors model	112.556 (19)	0.074	0.990	3 vs. 1	94.64***	0.010

****p < 0.001. S-Bχ^2, Satorra-Bentler scaled chi-square test; df, degrees of freedoms; |Δ(. . .)|, absolute value of the differences between indices; RMSEA, root mean square error of approximation; CFI, comparative fit index.*

Configural Invariance. The configural invariance model showed good model fit indices: S-Bχ^2 (24) = 24.350, p = 0.442; the RMSEA = 0.006; the CFI = 1.000; and the SRMR = 0.025; suggesting that the factor structure was similar between males and females.

Metric Invariance. The metric invariance model well-fitted the data: S-Bχ^2 (40) = 77.762, $p < 0.001$; the RMSEA = 0.046, the CFI = 0.996, and the SRMR = 0.043. A statistically significant decrease in chi-square was found: DIFTEST (16) = 53.412; $p < 0.001$. However, a non-statistically significant decreases in CFI was found: $|\Delta \text{CFI}|$ = 0.004, indicating that items were equivalently related to the latent factor independently from gender.

Scalar Invariance. The scalar invariance model showed good model fit indices: S-Bχ^2 (61) = 78.150, $p < 0.001$; the RMSEA = 0.025, the CFI = 0.998; and the SRMR = 0.030. A non-statistically significant decrease in chi-square was found: DIFTEST (21) = 0.387; p = 1. Moreover, a non-statistically significant decreases in CFI was found: $|\Delta \text{CFI}|$ = 0.002, suggesting that males and females had the same expected item response at the same absolute level of the trait.

Latent Means Invariance. The latent mean invariance model well-fitted the data: S-Bχ^2 (64) = 103.994; the RMSEA = 0.037, the CFI = 0.996; and the SRMR = 0.033. A statistically significant decrease in chi-square was found: DIFTEST (3) = 25.844; $p < 0.001$. Moreover, a non-statistically significant decreases in CFI was found: $|\Delta \text{CFI}|$ = 0.002, suggesting that males and females had the same expected latent mean of the traits.

Age ($\leq$ 64 y.o. vs. $\geq$ 65 y.o.)

Model $\leq$ 64 y.o. The Chi-square statistic resulted to be not statistically significant: S-Bχ^2 (12) = 10.496; p = 0.573. Moreover, all the other fit indices revealed a good fit to the data: the RMSEA = 0.000; 90%CI [0.000, 0.051]; p(RMSEA < 0.05) = 0.947, the CFI = 1.000, the SRMR = 0.029.

Model $\geq$ 65 y.o. The Chi-square statistic resulted to be not statistically significant: S-Bχ^2 (12) = 11.080; p = 0.552. Moreover, all the other fit indices revealed a good fit to the data: the RMSEA = 0.000; 90%CI [0.000, 0.040]; p(RMSEA < 0.05) = 0.990, the CFI = 1.000, the SRMR = 0.021.

Configural Invariance. The configural invariance model showed good model fit indices: S-Bχ^2(24) = 21.577; the RMSEA = 0.000; the CFI = 1; and the SRMR = 0.023; suggesting that the factor structure was similar across age.

Metric Invariance. The metric invariance model well-fitted the data: S-Bχ^2 (40) = 67.042; the RMSEA = 0.039, the CFI = 0.997, and the SRMR = 0.040. A statistically significant decrease in chi-square was found: DIFTEST (16) = 45.465; $p < 0.001$. However, a non-statistically significant decreases in CFI was found: $|\Delta \text{CFI}|$ = 0.003, indicating that items were equivalently related to the latent factor independently from age.

Scalar Invariance. The scalar invariance model showed good model fit indices: S-Bχ^2 (61) = 68.991; the RMSEA = 0.017, the CFI = 0.999; and the SRMR = 0.032. A non-statistically significant decrease in chi-square was found: DIFTEST (21) = 1.949; p = 1. Moreover, a non-statistically significant decreases in CFI was found: $|\Delta \text{CFI}|$ = 0.002, suggesting that younger ($\leq$ 64 y.o.) and older ($\geq$ 65 y.o.) patients had the same expected item response at the same absolute level of the trait.

Latent Means Invariance. The latent mean invariance model well-fitted the data: S-Bχ^2 (64) = 73.815; the RMSEA = 0.018, the CFI = 0.999; and the SRMR = 0.033. A non-statistically significant decrease in chi-square was found: DIFTEST (3) = 4.825; p = 0.185. Moreover, a non-statistically significant decreases in CFI was found: $|\Delta \text{CFI}|$ = 0.000, suggesting younger ($\leq$64 y.o.) and older ($\geq$65 y.o.) patients had the same expected latent mean of the traits.

Accuracy of the Psychological Distress Inventory—Revised as a Screening/Diagnostic Tool

Based on the HADS-T scale cut-off for moderate distress (HADS-T $\leq$ 15 vs. HADS-T $\geq$ 16), the *"general distress"* scale of the PDI-R obtained excellent accuracy in discriminating between patients without distress and patients with distress: AUC = 0.908, 95%CI [0.889, 0.928] (**Table 4** and **Figure 3A**). Considering a cut-off point of 15 (i.e., PDI-R $\geq$ 15: risk of moderate distress), ROC curve revealed a SE equal to 0.881, 95%CI [0.884, 0.918], a SP equal to 0.786, 95%CI [0.753–0.818], and an ACC equal to 0.817, 95%CI [0.817, 0.817]. On the basis of the gold standard test (HADS-T; moderate distress), 67.3% of patients were classified as non-distressed and 32.4% of patients were classified as distressed. Thus, using the reported cut-off for the PDI-R, the ROC curve showed that 52.88% of individuals were correctly classified as "true negative" and 28.82% as "true positive" with a percentage of correct classification equal to 81.7%. On the contrary, 3.88% resulted to be "false negative" and 14.41% resulted to be "false positive" (18.9% of misclassification).

Based on the HADS-T scale cut-off for severe distress (HADS-T $\leq$ 21 vs. HADS-T $\geq$ 22), the *"general distress"* scale of the PDI-R obtained excellent accuracy in discriminating between patients without distress and patients with distress: AUC = 0.918, 95%CI [0.896, 0.939] (**Table 4** and **Figure 3B**). Considering a cut-off point of 18 (i.e., PDI-R $\geq$ 18: risk of severe distress), ROC curve revealed a SE equal to 0.893, 95%CI [0.837, 0.948], a SP equal to 0.790, 95%CI [0.761, 0.819], and an ACC equal to 0.804, 95%CI [0.803, 0.804]. On the basis of the gold standard test (HADS-T; severe distress), 86.6% of patients were classified as non-distressed and 13.4% of patients were classified as distressed. Thus, using the reported cut-off for the PDI-R, the ROC curve showed that 68.4% of individuals were correctly classified as "true negative" and 11.97% as "true positive" with a percentage of correct classification equal to 80.37%. On the contrary, 1.44% resulted to be "false negative" and 18.18% resulted to be "false positive" (19.2% of misclassification).

Based on the DT scale cut-off for distress (DT $\leq$ 4 vs. DT $\geq$ 5), the *"general distress"* scale of the PDI-R obtained good accuracy in discriminating between patients without distress and patients with distress: AUC = 0.857, 95%CI [0.817, 0.897] (**Table 4** and **Figure 3C**). Considering a cut-off point of 13 (i.e., PDI-R *geq* 13: risk of distress), ROC curve revealed a SE equal to 0.847, 95%CI [0.783, 0.912], a SP equal to 0.739, 95%CI [0.686, 0.792], and

TABLE 4 | Study 2. Results of the ROC analysis.

	HADS—moderate distress				HADS—severe distress				DT—general distress			
	Thr.	**Sens.**	**Spec.**	**Acc**	**Thr.**	**Sens.**	**Spec.**	**Acc**	**Thr.**	**Sens.**	**Spec.**	**Acc**
2	9	0.997	0.107	0.398	9	1.000	0.084	0.207	9	0.991	0.163	0.419
3	10	0.993	0.222	0.474	10	1.000	0.175	0.286	10	0.957	0.405	0.576
4	11	0.986	0.320	0.538	11	1.000	0.253	0.354	11	0.932	0.530	0.654
5	12	0.983	0.458	0.630	12	1.000	0.362	0.448	12	0.898	0.625	0.709
6	13	0.959	0.606	0.722	13	0.992	0.485	0.553	**13***	**0.847**	**0.739**	**0.772**
7	14	0.939	0.723	0.794	14	0.983	0.583	0.636	14	0.737	0.822	0.796
8	**15***	**0.881**	**0.786**	**0.817**	15	0.967	0.650	0.693	15	0.686	0.871	0.814
9	16	0.820	0.832	0.828	16	0.959	0.708	0.742	16	0.602	0.894	0.804
10	17	0.742	0.865	0.825	17	0.917	0.757	0.778	17	0.568	0.905	0.801
11	18	0.698	0.891	0.828	**18***	**0.893**	**0.790**	**0.804**	18	0.483	0.920	0.785
12	19	0.661	0.921	0.836	19	0.859	0.822	0.827	19	0.424	0.931	0.775
13	20	0.593	0.947	0.831	20	0.794	0.858	0.849	20	0.356	0.943	0.762
14	21	0.508	0.965	0.816	21	0.727	0.894	0.871	21	0.280	0.966	0.754
15	22	0.417	0.975	0.793	22	0.653	0.924	0.888	22	0.263	0.977	0.756
16	23	0.366	0.985	0.783	23	0.587	0.941	0.894	23	0.237	0.989	0.756
17	24	0.302	0.987	0.763	24	0.504	0.954	0.893	24	0.203	0.992	0.749
18	25	0.254	0.990	0.749	25	0.488	0.972	0.907	25	0.186	0.992	0.743
19	26	0.210	0.993	0.737	26	0.388	0.976	0.897	26	0.152	0.992	0.733
20	27	0.169	0.995	0.725	27	0.306	0.979	0.889	27	0.136	0.992	0.728
21	28	0.132	0.997	0.714	28	0.248	0.986	0.887	28	0.110	0.992	0.720
22	29	0.098	0.998	0.704	29	0.198	0.992	0.886	29	0.110	0.996	0.722
23	30	0.051	0.998	0.688	30	0.099	0.995	0.875	30	0.076	1.000	0.715
24	31	0.024	1.000	0.681	31	0.050	0.999	0.871	31	0.059	1.000	0.709
25	33	0.020	1.000	0.680	33	0.050	1.000	0.872	33	0.051	1.000	0.707
26	35	0.014	1.000	0.677	35	0.033	1.000	0.870	35	0.034	1.000	0.701
27	36	0.010	1.000	0.676	36	0.025	1.000	0.869	36	0.025	1.000	0.699
28	39	0.007	1.000	0.675	39	0.016	1.000	0.868	39	0.017	1.000	0.696

**Highest average of sensitivity and specificity. Thr, Threshold; Sesn, Sensitivity; Spec, Specificity; Acc, Accuracy. HADS cut-offs (moderate distress and severe distress) and the DT cut-off. In bold are reported the highest average of sensitivity and specificity.*

an ACC equal to 0.772, 95%CI [0.771, 0.773]. On the basis of the gold standard test (DT), 69.3% of patients were classified as non-distressed and 30.7% of patients were classified as distressed. Thus, using the reported cut-off for the PDI-R, the ROC curve showed that 51.05% of individuals were correctly classified as "true negative" and 26.18% as "true positive" with a percentage of correct classification equal to 77.23%. On the contrary, 4.71% resulted to be "false negative" and 18.06% resulted to be "false positive" (22.77% of misclassification).

Normative Scores

On the total sample of 1,153 patients with cancer (sample of Study 1 *plus* sample of Study 2), both the normative scores (T-score: *mean* = 50, *SD* = 10) and the percentile distribution were calculated. Results are reported in **Tables 5**, **6**.

GENERAL DISCUSSION

In the last decades, psychological research in medical and oncological settings constantly increased focusing on observed outcomes of psychological care (Mannarini et al., 2013; Caminiti et al., 2017, 2021; Rossi et al., 2021). In this context, the construct of psychological distress has rapidly gained popularity (Mitchell, 2010; Wise et al., 2013; Riba et al., 2019; Marconi et al., 2020). Indeed, due to its severe negative impacts on medical outcomes (DiMatteo et al., 2000), distress represents one of the most important indexes of suffering in oncological patients (Mitchell, 2010; Cutillo et al., 2017). This interest led to the development of several questionnaires aimed at measure this construct (Carlson and Bultz, 2003; Herschbach et al., 2004; Mitchell, 2010; Wise et al., 2013; Cutillo et al., 2017; Riba et al., 2019).

In particular, the PDI (Morasso et al., 1996) is one of the most used instruments worldwide (Wise et al., 2013; Riba et al., 2019). The PDI was originally developed in the Italian context: it is a thirteen-item self-report instrument (Morasso et al., 1996) and it is among the most recommended tools to screen for distress in oncology (Vodermaier et al., 2009; Muzzatti and Annunziata, 2012)—however, the original validation study showed that it lacks of a rigorous assessment of its (basic) psychometric properties such as its factorial structure. Indeed, The only identified validation study of PDI concerned Italian cancer patients (Morasso et al., 1996). In this study, criterion, concurrent and discriminant—but not construct—validity were reported, together with good internal consistency (Muzzatti and Annunziata, 2012). Despite

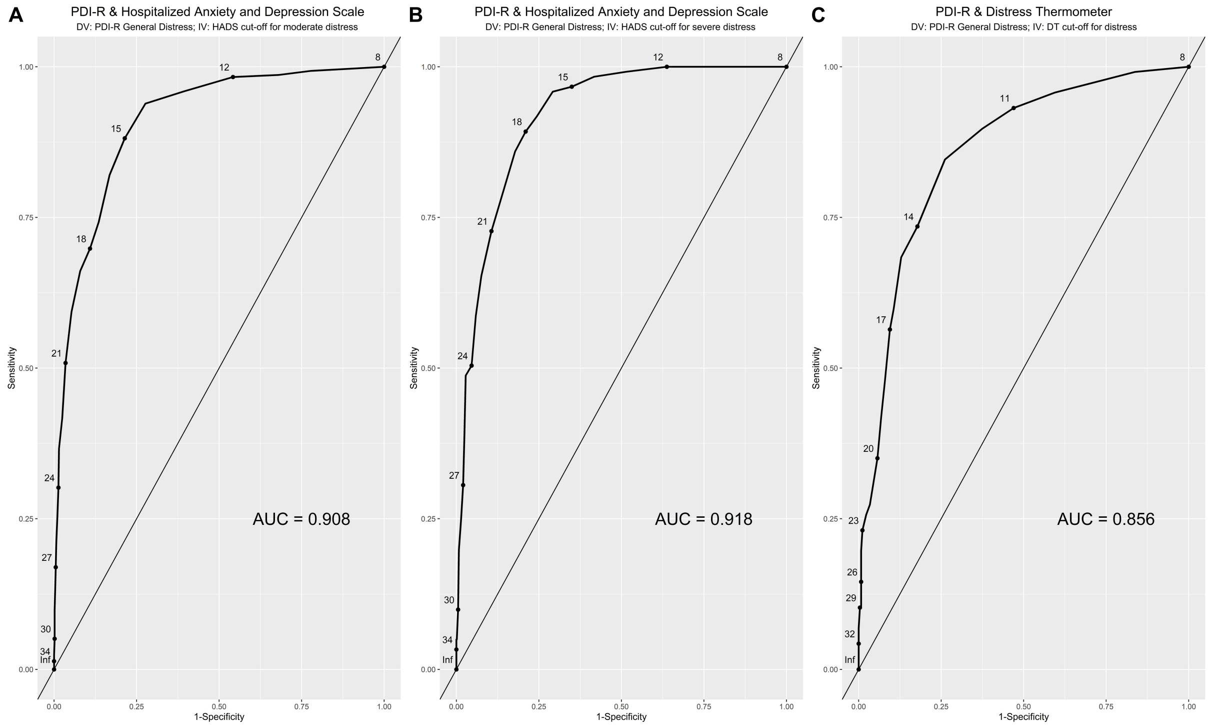

FIGURE 3 | Study 2. ROC curves analysis. HADS cut-offs (**A:** moderate distress; **B:** severe distress) and the DT cut-off **(C)**. x-axis: 1-specificity; y-axis: sensitivity; AUC = area under the (ROC) curve.

it provides encouraging, albeit incomplete, information on reliability and validity, data on construct validity are strongly requited for a screening tool (Muzzatti and Annunziata, 2012).

Starting from this background, in line with previous researches (Panzeri et al., 2021c; Parola et al., 2022; Rossi et al., 2022), the aim of the first study here reported was to extensively examine the factorial structure of the PDI. To achieve this goal, EGA was used: an innovative technique, particularly precise and sensitive in identifying the correct number of factors/dimensions (Golino and Epskamp, 2017). Moreover, on the basis of the results of Study 1, the second study (Study 2) aimed at developing a (shortened) revised version of the PDI (PDI-R) and to provide an in-depth analysis of its psychometric properties including its factorial structure and measurement invariance, thus filling the gap in the literature. To achieve this, gold-standard statistical techniques were used to analyze the psychometric properties of the questionnaires (e.g., CFA).

The first study revealed that the (original) supposed single-factor structure of the PDI (Morasso et al., 1996)—one latent dimension/factor accounting for all of the 13 items—was not adequate. Indeed, the EGA revealed that the original pool of items grouped onto three different dimensions/factors—instead of a single one (**Figure 1**). These dimensions refer to: (1) internal states of psychological distress such as anxiety and depression; (2) relationships and the external world: lack of desire to talk to others and loneliness; and (3) wellbeing/tranquility. It should be noted that this result might be attributed to the original structuring procedure of the PDI (Morasso et al., 1996) and the statistical analysis performed. Indeed, Morasso et al. (1996) did not test the factorial structure of the PDI but only evaluated its internal consistency—and, since Cronbach's alpha resulted good, a single-factor structure was inferred. In addition, despite the original validation article contained three studies attempting to validate the PDI, each of them was conducted relatively small sample sizes and thus, results may be biased.

In addition, scientific literature showed that modern techniques of statistical analysis—such as EGA (Golino and Epskamp, 2017)—are more strongly adequate and precise to extract the right number of factors of a questionnaire than classical exploratory techniques (e.g., exploratory factor analysis, EFA) (Golino and Demetriou, 2017; Christensen and Golino, 2019, 2020; Christensen, 2020; Christensen et al., 2020a; Golino et al., 2020b).

Results from Study 1 shed new light on the dimensional/factor structure of the PDI—inspiring the development of its revised (shortened) version: the PDI-R (Study 2).

For the validation and study of the psychometric properties of the PDI-R redundant items were removed: in particular, the two reversed scored items (PDI: item#2 and item#6). Moreover, also the three items that did not achieve the Item Stability threshold (PDI: item#4, item#8, and item#12) were removed. Consequently, only two first-order latent dimensions were retained: internal and external distress; and a second-order general distress dimension was hypothesized. Thus the factorial structure of a new, brief but solid questionnaire was tested on a large sample of oncological outpatients.

TABLE 5 | Study 1 *plus* Study 2. Normative scores (T-scores) and percentile distribution.

Overall			**Gender**				**Age**			
***N* = 1153**			**Male**		**Female**		**≤64 y.o.**		**≥ 65 y.o.**	
Raw	**T**	**$P_{.le}$**	**T**	**$P_{.le}$**	**T**	**$P_{.le}$**	**T**	**$P_{.le}$**	**T**	**$P_{.le}$**
8	31	3	32	3	32	4	32	3	33	4
9	39	14	40	15	38	11	36	8	39	14
10	43	24	43	25	42	22	40	17	42	22
11	45	33	46	33	45	32	44	27	45	30
12	48	41	48	41	48	41	46	36	47	37
13	50	48	49	48	50	49	48	44	48	44
14	51	55	51	54	52	56	50	51	50	51
15	53	61	53	60	53	63	52	58	52	57
16	54	67	54	66	55	68	54	64	53	62
17	56	71	55	70	56	73	55	69	54	67
18	57	76	57	75	57	77	56	73	56	72
19	58	80	58	78	59	81	58	77	57	76
20	59	83	59	82	60	84	59	81	58	80
21	61	86	60	85	61	87	60	84	60	83
22	62	88	61	87	62	89	61	86	61	86
23	63	90	62	89	63	91	62	89	62	88
24	64	92	63	91	65	93	63	91	63	91
25	65	94	65	93	66	95	64	92	64	92
26	66	95	66	94	67	96	65	94	65	94
27	67	96	67	95	68	97	66	95	67	95
28	69	97	68	96	70	98	69	96	68	96
29	70	98	69	97	71	98	69	97	69	97
30	71	98	70	98	73	99	70	98	70	98
31	72	99	71	98	76	99	72	98	71	98
32	73	99	72	99	78	99	73	99	72	99
33	75	99	73	99			75	99	74	99
34	76	99	74	99			79	99	75	99
35	78	99	76	99			79	99	76	99
36	80	99	77	99			79	99	77	99
37	80	99	78	99			79	99	79	99
38	80	99	80	99					80	99
39	80	99	80	99					80	99
40	80	99	80	99					80	99

Raw, General distress (PDI-R) raw score; T, T-score; $P_{.le}$, percentile. Overall sample, N = 1153; Male, n = 487; Female, n = 415; =64 y.o.: n = 327; =65 y.o.: n = 63.7.

A bi-factor structure was specified, and the CFA successfully confirmed that all the 8 items of the PDI-R loaded onto the supposed first-order latent factor (internal and external distress) and all the items loaded on the general dimension of distress (**Figure 2**). CFA also revealed that the PDI-R had good structural validity with good fit indices (Muthén and Muthén, 1998–2017; Hoyle, 2012; van de Schoot et al., 2012; Brown, 2015).

Moreover, considering that the PDI-R is a new scale, the proposed factorial structure was compared with possible competing models: a single-factor model and a two-first order model. Results of model comparison demonstrated that the proposed bi-factor structure is the best factorial solution for the PDI-R.

Item discrimination power was also tested. Results showed that each of the first four items composing the PDI-R well discriminated between subjects with low or high internal distress. At the same time, for the external subscale, the item discrimination power indicated that each of the second four items comprising the PDI-R well discriminated between subjects with low or high external distress. These results suggest the goodness of the items to discriminate between different types of distress in the individuals, and the ability of each item to represent its latent construct.

Reliability analyses were also performed, providing satisfying results for the internal, external, and general distress scale. In addition, the 1-month test-retest reliability provided good results – as shown by the two-way mixed ICCs (Pursey et al., 2016).

Convergent validity analyses were also performed. In line with the scientific literature, significant correlations were found

TABLE 6 | Study 1 *plus* Study 2. Normative scores (T-scores) and percentile distribution for gender and age.

	Male				**Female**			
	≤ 64 y.o.		**≥ 65 y.o.**		**≤ 64 y.o.**		**≥ 65 y.o.**	
Raw	**T**	**$P_{.le}$**	**T**	**$P_{.le}$**	**T**	**$P_{.le}$**	**T**	**$P_{.le}$**
8	33	3	34	5	32	4	32	3
9	36	8	40	16	36	9	37	9
10	40	16	43	24	41	18	41	18
11	43	24	45	32	45	30	44	26
12	45	32	47	39	48	41	46	35
13	47	39	49	46	50	50	48	42
14	49	46	51	52	52	58	50	49
15	51	52	52	58	54	64	52	56
16	52	58	53	63	55	70	53	62
17	53	63	55	68	57	75	54	67
18	55	68	56	73	58	79	56	72
19	56	72	57	76	59	82	57	76
20	57	76	58	80	60	85	58	80
21	58	80	60	83	62	88	60	83
22	59	83	61	86	63	90	61	86
23	61	85	62	88	64	92	62	88
24	62	88	63	90	65	93	63	91
25	63	90	64	92	66	95	64	93
26	64	92	65	93	68	96	66	94
27	65	93	66	95	69	97	67	96
28	66	95	67	96	70	98	68	97
29	67	96	68	96	72	99	70	98
30	69	97	69	97			72	99
31	70	98	70	98			74	99
32	71	98	71	98			77	99
33	73	99	72	99				
34	76	99	73	99				
35	78	99	74	99				
36	78	99	75	99				
37	78	99	76	99				
38			77	99				
39			78	99				
40			79	99				

Raw, General distress (PDI-R) raw score; T, T-score; $P_{.le}$, percentile. Male = 64 y.o.: n = 170; Male = 65 y.o.: n = 317; Female = 64 y.o.: n = 157; Female = 65 y.o.: n = 258.

between the PDI-R total score and other well-consolidated measures of psychological suffering such as the HADS and the DT (Wise et al., 2013; Riba et al., 2019). Strong correlations were found between the PDI-R general distress scale, the HADS total score (r = 0.775), its subscales (HADS anxiety, r = 0.713; HADS depression, r = 0.705), and the DT (r = 0.576). These correlations suggest a strong association of distress (measured by the PDI-R) with psychological and emotive difficulties in oncological patients—due to the possible presence of people with severe diagnoses and related preoccupations and fears in the sample (Rossi et al., 2021).

Moreover, MI was tested to explore at which level (structural vs. loadings vs. intercepts/thresholds, means) there were differences across gender (males vs. females) and age (≤ 64 vs. ≥ 65). MI analysis showed that latent means invariance was achieved. This suggests that the eight items were equivalently related to the latent distress factors across each sample, and indicates that samples had the same expected item response at the same absolute level of the trait. Thus, males and females as well as patients with different age interpreted the PDI-R items in the same way (the factorial structure was equal across samples),with the same strength (items were related to the latent construct equally between samples), with the same "starting point" (item thresholds were similar among samples) and had the same latent mean of the construct (latent means were similar across samples). Consequently, experienced psychological distress measured by the PDI-R can be evaluated and compared between males and females as well as between patients with different age (≤ 64 vs. ≥ 65).

In addition, results from the ROC analyses showed that the PDI-R is an excellent screening/diagnostic tool for the detection of psychological distress. Indeed, the PDI-R presented an excellent accuracy in discriminating between distressed and non-distressed oncological outpatients. More in detail, considering the principal aim of a screening tool—i.e., to capture the majority of "positives" patients (Zou et al., 2012)—the PDI-R showed excellent sensitivity properties. At the same time, it is important to underline that—in contrast to many screening tools—specificity is not compromised and it showed very good values. These results suggest that—in spite of the small number of items (n = 8)—the PDI-R is an excellent instrument for the screening of distress in samples of cancer patients. In this regard, it should be emphasized that the proposed cut-offs are those with the best average ratio between sensitivity and specificity on a sample of 902 cancer patients and should therefore be considered as guidelines for patient classification—which, however, cannot replace the clinical interview. However, these cut-offs should not be used in an absolutely inflexible way: they should be evaluated according to the situation and what is to be obtained from the screening assessment (Cohen, 1988).

Consequently, in order to guide the interpretation of the score obtained on the "general distress scale" of the PDI-R, normative scores (T-scores) and the percentiles distribution were calculated—on a large sample of 1,153 patients with cancer. Using the cut-offs that emerged from Study 2 (i.e., PDI-R ≥ 15: moderate distress; PDI-R ≥ 18: severe distress) it is possible to observe that the thresholds for moderate and severe distress are, respectively, located in the presence of the 60th and 75th percentiles—underlining the adequacy of the previously proposed guidelines.

Despite these interesting findings, several limitations have to be highlighted. First, despite the sample size being adequate to perform a CFA, the provided bi-factor model might be expensive in terms of sample-to-parameter ratio. Consequently, cross-cultural adaptation and validation studies aimed at computing the factorial structure of the PDI-R should consider enrolling an appropriate number of outpatients. Moreover, the number of participants (n = 40) to which the PDI-R was re-administered was enough for the assessment of the test-retest reliability but far from being adequate for a longitudinal MI analysis. In addition, the sample of the present research was only composed by oncological outpatients: future studies should investigate the factorial structure of this inventory in other categories of (hospitalized) patients.

Despite these limitations, it is also important to emphasize some strengths of the present research work. First of all, this work is based on a two-step methodology that has already been consolidated in previous research. Second, both questionnaires (i.e., PDI and PDI-R) were extensively analyzed using—for example—innovative statistics (e.g., EGA). Third, both studies are based on large sample sizes, which makes it possible to use robust and reliable statistics. Fourth, the study of factorial invariance showed that the PDI-R is an instrument that is widely applicable to heterogeneous contexts with patients of both sexes or different age groups.

Still, this contribution shows that the PDI-R might be a good instrument for the assessment of psychological distress in oncological settings and that it might also be used for research purposes.

Last, the PDI-R might also represent a starting point for the assessment of psychological distress and the planning of (psycho-)oncological treatments aimed to reduce of the individuals' psychological suffering and the general health status of patients in oncological settings.

DATA AVAILABILITY STATEMENT

The datasets presented in this article are not readily available due to privacy restrictions, data were available from the corresponding author on a reasonable request. Requests to access the datasets should be directed to corresponding author.

ETHICS STATEMENT

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

AUTHOR CONTRIBUTIONS

AAR conceived the study, wrote the first draft, performed statistical analyses, and displayed tables and figures. MM collected the data. FT wrote part of the first draft. CV and SM supervised the work and revised the final version of the manuscript. All authors read, approved the work, and contributed to the article and approved the submitted version.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2022.859478/full#supplementary-material

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Spirituality During the COVID-19 Pandemic: An Online Creative Arts Intervention With Photocollages for Older Adults in Italy and Israel

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The present study aimed to examine how expressions of spirituality were stimulated and reflected in an online creative arts intervention for older adults during COVID-19 lockdowns. The online process focused on the creation of digital photocollages together with narrative elements of dignity therapy. Twenty-four Israeli and Italian community-dwelling older adults aged 78–92 participated in a three-session online intervention involving the production of three photocollages. The visual and verbal data (participants' chosen photos and photocollages, and transcripts of the sessions) were qualitatively analyzed within an abductive framework. Four themes were generated, representing the four domains of spirituality that were stimulated by and expressed in the process: (1) Connectedness with the self, (2) connectedness with others, (3) connectedness with the environment, and (4) connectedness with the transcendent. The findings show how photographs can serve as projective visual stimuli which elicit personal content through spontaneous thinking, and they reveal the multifaceted nature of spirituality, wherein each domain nourishes the others. Overall, the findings illustrate how creative arts intervention guided by the tenets of dignity therapy can contribute to the spiritual care of older adults during periods of social isolation, or to the spiritual support provided in palliative care.

Keywords: spirituality, older adults, creative arts therapies, online interventions, COVID-19 lockdowns

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INTRODUCTION

Social restrictions introduced in response to the COVID-19 pandemic have caused many people psychological distress (Zhu et al., 2021; Keisari et al., 2022a). Social distancing mandates particularly affected older adults who live alone or rely on psycho-social support outside the home (Morrow-Howell et al., 2020; Steinman et al., 2020). The pandemic also led to a parallel outbreak of ageism, with an increasing portrayal of older adults as helpless, frail, and unable to contribute to society (Cesari and Proietti, 2020; Ayalon et al., 2021). Exposure to ageism is associated with a decline in self-esteem and dignity (Lev et al., 2018) and with poorer mental health (Lyons et al., 2018). The combination of isolation and ageism thus created a substantive need to support older adults and enhance their coping resources in this time of crisis (Wand et al., 2020; Wu, 2020). Studies point to the value of spirituality in promoting wellbeing and as a coping resource (Reutter and Bigatti, 2014; Lifshitz et al., 2019). Recent studies on mental health, spirituality and COVID-19 show that spirituality contributes to wellbeing and resilience in times of adversity (Prazeres et al., 2020; Del Castillo, 2021; Zhang et al., 2021).

This study reports an online creative arts intervention for older adults implemented during COVID-19 lockdowns in two countries, Italy and Israel, both of which instituted strict social distancing measures and curfews to mitigate the spread of the virus. The life expectancy in both countries is similar (in 2020 the life expectancy in Italy was 84.8 years for women and 79.7 for men and in Israel 84.8 years for women and 80.7 for men). However, the older population in Italy (age > 65) makes up a larger percentage (about 23.5% of the whole population) than in Israel (roughly 11.95%) (Central Bureau of Statistics, 2021; ISTAT, 2022). Another major difference between the two populations has to do with religiosity. While in Italy the majority of the population describe themselves as Christian (for example 79.2% of the population are Catholic; European Union, 2021), the population in Israel is mostly Jewish (for example 74% are Jewish and 18% are Moslem; Central Bureau of Statistics, 2021).

Creative arts therapies (CAT) is an umbrella term for healthcare professions employing the creative and expressive process of art-making to foster psycho-social wellbeing (Shafir et al., 2020). A growing body of research points to the value of creative art interventions for older adults, and their ability to improve various facets of mental health such as meaning in life, self-acceptance, wellbeing, positive relationships with others, and the reduction of symptoms of depression (Beard, 2012; Ciasca et al., 2018; Dunphy et al., 2019; Keisari et al., 2020). However, there is as yet scant literature on the ability of creative arts therapies to provide spiritual care for older adults. The current study aimed to help bridge this gap.

Our intervention combined a short-term individual creative arts therapy project, in which participants created their own photocollages, with narrative elements of dignity therapy, a psychotherapeutic mode aimed at helping individuals approaching the end of life maintain their dignity and sense of purpose (Chochinov et al., 2004, 2005). In this research, we sought to understand how engagement in the creative process, guided by the tenets of dignity therapy, helped participants connect to their spirituality as a coping resource. In so doing, we drew on theories that explore the dimensional structure of spirituality (Fisher, 2010; de Jager Meezenbroek et al., 2012) and its relationship with wellbeing in later life (Thauvoye et al., 2018; Lifshitz et al., 2019). This study examined how the spiritual care provided was manifested in four domains or orientations: connectedness with the self, connectedness with others, connectedness with the environment or nature, and connectedness with the transcendent.

Spirituality in Late Life

Before we proceed, let us clarify what we mean by spirituality. Social scientists draw a distinction between spirituality and religion, where the latter tends to refer to the external, institutionalized, formal, and doctrinal ways by which people engage with some concept of the sacred, transcendent, or divine (Dalby, 2007). Spirituality, by contrast, refers to personal, subjective, existential experiences (Gijsberts et al., 2011; de Jager Meezenbroek et al., 2012).

Theories of spirituality emphasize the human need to search for meaning and purpose in life (Muldoon and King, 1995; Daaleman et al., 2004; Emmons, 2006), and the universal desire to feel "at peace" (Steinhauser et al., 2006). These theories highlight how spirituality, especially in secularized Western societies, allows people to feel the divine within themselves, rather than as an external higher entity (Heelas, 2000), or link spirituality to a sense of connection with the essence of life (de Jager Meezenbroek et al., 2012).

There is a general consensus that spirituality is multidimensional in nature (Puchalski, 2012; Steinhauser et al., 2017). de Jager Meezenbroek et al. (2012), for example, described four dimensions of spirituality: Connectedness with transcendence as a higher being, connectedness with the self, connectedness with others, and connectedness with nature. Similarly, Fisher's (2010) model of spiritual health and wellbeing recognizes four domains: (a) The personal domain, which deals with self-awareness as a source of meaning, purpose and values in life; (b) the communal domain, which concerns the quality and depth of interpersonal relationships; (c) the environmental domain, which relates to the notion of unity with the environment; and (d) the transcendental domain, which relates to one's relationship with a higher entity. Those models both center around different orientations of spirituality. By contrast, Gijsberts et al. (2011), following a systematic review of instruments measuring spirituality in end-of-life populations, distinguish between three measurable dimensions of spirituality: spiritual wellbeing (e.g., peace); the spiritual cognitive behavioral context (e.g., spiritual beliefs, spiritual activities, and spiritual relationships); and spiritual coping.

Spirituality tends to develop during later adulthood, even in secular modern societies (Wink and Dillon, 2002, 2008; Moberg, 2008). Specifically, people tend toward more spiritual perspective from mid-life onward, where spirituality takes on an increasingly central role as people cope with aging and the approaching end of life (Gijsberts et al., 2011). Several explanations have been put forward for the development of spirituality in later adulthood, including awareness of one's mortality that comes at midlife, and changes of priorities in life, since before midlife, external constraints associated with launching a career and establishing a family tend to be dominant. Other studies suggest that the adversities related to old age, such as the loss of meaningful others, exposure to ageism, and restricted mobility can direct many older adults toward spirituality as a resource (Wink and Dillon, 2002). Tornstam's theory of gerotranscendence holds that as people age, they undergo a shift in perspective, from a materialistic and rational view of the world to one that is cosmic and transcendental (Tornstam, 1997). Gerotranscendence typically includes a redefinition of the self and relationships to others and a new understanding of fundamental existential questions. It is characterized by an increased feeling of affinity with past generations and a decreased interest in superfluous social interactions. Individuals become less self-concerned and interested in material things, and more selective in their choices of social and other activities (Hyse and Tornstam, 2009).

Studies have found associations between spirituality and various aspects of mental health and wellbeing in old age (McCauley et al., 2008; Reutter and Bigatti, 2014; Malone and Dadswell, 2018). Spirituality contributes for example to promoting and maintaining resilience in later life (Manning, 2013). It is associated with increased life satisfaction over time, and can influence older adults' interpretations of events, making events seem generally meaningful, and determining whether events are seen as opportunities, rather than threats and demands (Cowlishaw et al., 2013). Spirituality can also act as a buffer against death anxiety (Budhiraja and Midha, 2017). Spirituality is also associated with lower COVID-19-related anxiety (Prazeres et al., 2020). In addition, spiritual fortitude, which refers to the ability to draw on spiritual resources in the face of stressors, is related to various facets of mental health, such as meaning in life, spiritual wellbeing, and perceived posttraumatic growth (Zhang et al., 2021).

Some studies investigating how spirituality may improve mental health in older age (e.g., reducing depressive symptoms) have focused on specific domains or dimensions of spirituality (Rykkje et al., 2013; Han and Richardson, 2015; Thauvoye et al., 2018; Lifshitz et al., 2019). The present study adds to this literature by exploring how different domains of spirituality were stimulated by the current intervention, following the orientational models of Fisher (2010) and de Jager Meezenbroek et al. (2012). This intervention is focused on the creation of digital photocollages together with narrative elements of dignity therapy.

Dignity Therapy

Dignity therapy (Chochinov, 2002, 2012; Chochinov et al., 2005) is a brief individualized form of narrative psychotherapy, targeted at individuals of any age approaching the end of life (e.g., the terminally ill), as well as the older population (Fitchett et al., 2015). Dignity therapy involves the preparation of a physical *generative document*, which is based on the client's personal narratives. The process of creating the generative document is structured around eight themes: generativity, continuity of self, role preservation, maintenance of pride, hopefulness, aftermath concerns, and care tenor (Chochinov et al., 2005). The full process is intended to increase people's sense of purpose and preserve their dignity by giving them the opportunity both to reflect upon their lives, and to turn their life narratives into a resource for others (Chochinov et al., 2004). In the current intervention we related to these themes through three topics, one for each session: (1) Turning points in the personal narrative; (2) personal legacy; and (3) future perspectives. We describe our protocol in detail in the **Supplementary Material**.

Integrating the use of photographs within dignity therapy has been found to facilitate recollection and to help clients structure their narratives. Photographs serve as a projective stimulus, helping elicit past experiences, themes, and visual expressions that support the creation of the narrative (Testoni et al., 2019b, 2020, 2021). More precisely, visual images in photographs can reveal themes and topics which cannot be expressed by words alone, such as emotions, embodied expressions, and muted or sensitive aspects of personal experience (Weiser, 2018; Testoni et al., 2020, 2021). The photographs used in the therapy may be the personal snapshots or family photos belonging to the individual, or may come from some other source (e.g., stock images or art photos).

Crafting the Life Stories of Older Adults Through Photocollages

Collage is a popular practice in which participants select various materials and images, compose them, and attach them to a surface (Raffaelli and Hartzell, 2016; Russo-Zimet, 2016; Kaimal et al., 2020). Collage-making is an effective way of helping older adults process their memories and life experiences (Stallings, 2010; Keisari et al., 2022b). The creative process stimulates personal content and memories, while still allowing for a reflective distance that promotes verbalization and communication (Raffaelli and Hartzell, 2016). Collage also enables people to engage in art-making with less perceived threat, because it does not demand high technical skills (Stallings, 2010).

This study focused on the creation of digital photocollages, in which participants selected and composed photographs using software tools. Our previous findings show that the projective stimuli of digital photographs supported older adults' narratives and engaged them in a more embodied emotional experience. Creation of a digital photocollage can thus serve as a visual generative document for participants' life-stories (Keisari et al., 2022b). This study examined how expressions of spirituality were stimulated and reflected in this online creative arts intervention for older adults. Specifically, we explored which domains of spirituality were stimulated by the creative process, and expressed in the final created products (the photocollages).

MATERIALS AND METHODS

Sample

Twenty-four community-dwelling older adults took part in the study, including 12 in Italy and 12 in Israel. Participants were recruited through community coordinators, social workers at day centers, and family members. Inclusion criteria for the study were: (1) An age of 78 years or over (i.e., late aging); (2) a Mini-Mental State Examination above 24 (reported by social workers and family members), indicating normal cognitive performance (Woodford and George, 2007); (3) absence of any mental disorder or major depression; (4) vision and hearing sufficient to engage in online conversation; and (5) access to a computer screen large enough to engage in the creative process (smartphone screens were not appropriate).

After receiving initial consent to take part in the study, the researchers conducted an introductory phone call with all participants to explain the study aims and content. Participants provided formal verbal recorded informed consent at the start of the first online session (see below).

The study was approved by the Ethics Committee for Experimentation, University of Padua (confirmation number 0581B1B9C39761AE3C03AD3D93EFDEE9) and by the Ethics Committee of the Faculty of Social Welfare and Health Sciences at the University of Haifa, Israel (confirmation number 366/21). To preserve confidentiality, pseudonyms are used for the participants.

Crafting Life Stories Through Photocollages: Description of the Intervention

The short intervention involved a series of individual online sessions, in which participants made three photocollages. The sessions were structured around themes drawn from dignity therapy. Each session was focused on the exploration of one theme: (1) ***Turning points in the personal narrative***—significant life events and related themes, roles, and coping resources (McAdams, 2001; Chochinov et al., 2002; Keisari and Palgi, 2017); (2) ***personal legacy***—how the participant wished to be remembered, and what values and lessons they would like to pass on to their loved ones (Chochinov, 2002; Testoni et al., 2021); and (3) ***future perspectives***—desires, thoughts and concerns regarding the future and the end of life (Lang and Carstensen, 2002; Testoni et al., 2015, 2017). The full protocol (Keisari et al., 2022b) and TIDieR checklist (Hoffmann et al., 2014) can be found in **Supplementary Material**.

The sessions were conducted *via* Zoom, with the "share screen" function operational. At the start of each session, the themes and related questions were introduced by the therapist using as prompts a set of artistic photos, selected in advance for their ability to serve as visual metaphors for that session's theme (Weiser, 2004). The full pool comprised about 80 photographs, either original images by Israeli photographers Michal Fattal and Yehudit Liberman, or licensed photos obtained from the iStock website.[1] Participants were asked to view the presented photographs, choose those that most reflected their own personal experiences in relation to the themes raised by the therapist, and arrange them together within a blank space on the screen. This was done using PowerPoint software, which is available on most computers and enables flexibility in the creative process of selecting, cropping, positioning, and titling digital photographs. Participants were also permitted to select and incorporate personal photographs, or other photographs taken from the Internet (all such photos were replaced by licensed photographs from the iStock website for this publication). At the end of the process, the digital photocollages were printed and sent to the participants.

The sessions were conducted by the first (SK), second (SP), fourth (TE), and fifth (GM) authors, with each participant working with one therapist. The first author is a supervisor and drama therapist specializing in clinical gerontology. The second and fourth authors were doing internships in clinical psychology and drama therapy, and the fifth author was a master's degree student in clinical psychology. Two of the therapists (native Italian speakers) conducted the sessions in Italy, and two (native Hebrew speakers) in Israel. The sessions were conducted as a therapeutic process, so that the relationship with the researchers were therapeutic in nature, as this provided a safe, positive and corrective experience for the participants to promote self-development.

To ensure adherence to the protocol, the first (SK) and sixth (IT) authors conducted five training sessions with the team (2.5 h each) prior to the intervention in order to gain familiarity and practice how to deliver the intervention. In addition, 90-min online supervision sessions were held twice a week during the course of the study by the first author. During these sessions, the student therapists were able to discuss their work and receive professional support. Overall, 22 supervision sessions were conducted.

The Setting

The original protocol called for three online sessions for each participant, each lasting approximately 90 min. Some meetings were split into two sessions to accommodate the participants. Overall, 78 sessions were conducted between December 2020 and March 2021, ranging in length from 26 to 120 min, with 37 sessions held in Italy and 42 in Israel. Sessions were held about 2–7 days apart. In addition, participants would occasionally contact the therapist outside the formal sessions, *via* WhatsApp or email, to share thoughts, personal photographs, texts, and ideas they wanted to incorporate into their collage. Some participants were supported by family members or day center staff in their use of technology such as setting up the computer and the Zoom meetings.

Data Collection and Analysis

All sessions were video-recorded and later transcribed for analysis. The qualitative data collected for the study thus included both a verbal component (the transcribed sessions) and a visual component (each participant's chosen photographs and photocollages).

The analysis employed abductive reasoning, a pragmatist approach to analyzing qualitative data (Tavory and Timmermans, 2014). Following this approach, we began by drawing observations from the data (both verbal and visual), and then sought to make sense of those observations by examining them against theories and conceptualizations found in the literature (Timmermans and Tavory, 2012; Brinkmann, 2014; Earl Rinehart, 2020). This process led us to narrow our focus to the dimensions of spirituality revealed by the data. Specifically, we asked the following question: which dimensions or domains of spirituality were stimulated by the creative process, and expressed in the final created products (the photocollages)?

After selecting our research question, we conducted a polytextual thematic analysis (Gleeson, 2020) of both the verbal and visual data. The dataset was analyzed by the four researchers who had conducted the therapy sessions, using the Atlas.ti 9 cloud software, which allows for a team of researchers to work and analyze the same data together using a shared code set. The visual data were analyzed in relation to the participants' perspectives. Accordingly, the analysis

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[1] https://www.istockphoto.com/

focused on the participants' reflections on the visual images, their description (e.g., in terms of colors, shapes, movements, and texture), and the subjective experiences prompted by the photographs and the final photocollage. In addition, the final art products of photocollage were also analyzed by the research team, who took ongoing notes on the themes during the observation.

In the first step, the four researchers reviewed the data of the verbatim transcriptions along with the visual photograph and photocollage data. The verbal data were produced in three languages: Italian, Hebrew, and English (as three of the Israeli participants were native English speakers, their sessions were conducted in English).

In the second step, the researchers conducted an initial coding of the data. The initial coding was conducted in the original language. The two Italian researchers (SP and GM) coded the Italian data, and the two Israeli researchers (SK and TE) coded the Hebrew and English data.

Initial coding by two researchers served to maintain reflexivity since the researcher who conducted the session as a therapist, had an insider's perspective on the data, whereas the other had an outsider's perspective. Both perspectives shed light on the effects of one's position vis-à-vis the phenomenon (Berger, 2015).

To allow the Italian and Israeli researchers to work together, the codes were labeled and defined in English. During this stage, the four researchers met twice a week to review and discuss sample quotations from the transcripts, to ensure that the code definitions were consistent and appropriately applied to the responses. In cases of disagreement, the researchers discussed the coding process until an agreement was reached. These team consultations during the analysis process served to identify possible projections and content missed by the researchers (Berger, 2015).

In the third step, following the abductive reasoning (Tavory and Timmermans, 2014), after the initial set of codes was created, the researchers examined the codes against the theories and conceptualizations in the literature to identify significant broader patterns of meaning, and the codes were grouped into themes and subthemes. For example, the researcher identified the codes of "sense of contributing," "couplehood as coping resource," "generativity," "friendship as coping resource," "forgiveness," "universalism," "care in the family," and grouped them into three themes: "Connectedness with significant others," "a sense of contributing to others," and "caring for humanity." In the next step, the researchers created links between these three themes to form one core theme labeled "connectedness with others." The coding and analysis processes ended when theoretical saturation occurred and new data could not contribute to the theme (Fusch and Ness, 2015).

In the fourth step, the candidate themes were checked against the dataset and refined if needed. At this stage, selected quotations were translated to English to enable the third (HO) and sixth (IT) authors to review the thematic map. Note that the translation was verified by the two native speakers (Italian and Israeli) to ensure that no meaning was lost during the translation process. Finally, in the fifth step, the themes and subthemes were organized to create a model.

FINDINGS

Participants had a mean age of 83.96 (range: 78–92), with a mean of 84 (range: 78–88) for the Italians and 83.92 (range: 80–92) for the Israelis. Of the whole sample, 58.33% were women, 25% were currently married, and all the participants had children. Also, 58.33% of the sample considered themselves religious. Full demographic details of the sample can be found in **Table 1**.

As described above, our abductive analysis aimed to identify the domains of spirituality that were expressed through the intervention. Four main themes were generated, each representing a spiritual domain or orientation: (1) connectedness with the self; (2) connectedness with others; (3) connectedness with the environment; and (4) connectedness with the transcendent. The first two themes were further divided into several subthemes. The full model of themes and subthemes appears in **Figure 1**.

In what follows, we introduce and describe each of the main themes and subthemes. Where relevant, we also show how different themes linked together, such that participants' feelings and perceptions often glided seamlessly from one theme to another.

TABLE 1 | Participants' demographics.

Variables	Israeli participants	Italian participants	Total
Mean age (range)	83.92 (80–92)	84 (78–88)	83.96 (78–92)
Gender	6 Females	8 Females	58.33% Female
Place of birth	3 in Israel; 4 in America; 4 in Europe; 1 in Asia	All were born in Italy	37.5% Had immigrated in their lifetime (only Israeli participants)
Marital status	1 Married; 2 divorced; 9 widowed	5 Married; 7 widowed	25% Currently married
Education	4 Had a high school education; 8 had a college education	6 Had a primary school education; 6 had a high school education	25% Had a primary school education; 41.66% had a high school education; 33.33% had a college education
Religiosity	8 Defined themselves as secular, 4 as religious	2 Defined themselves as secular, 10 as religious	58.33% Defined themselves as religious
Religion	11 Were Jewish; 1 identified as an atheist	All were Catholic	45.83% Were Jewish, 50% Catholic, and 4.16% atheist

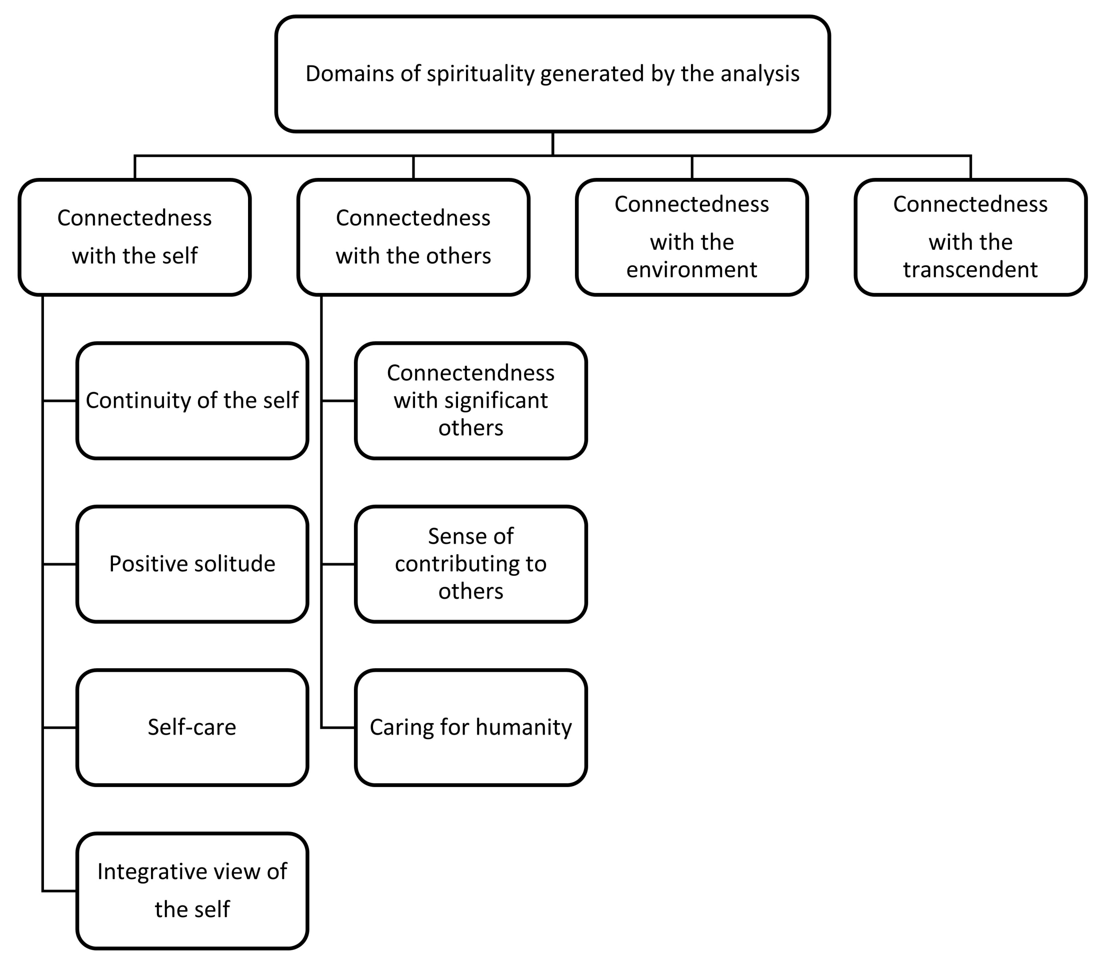

FIGURE 1 | The thematic map.

Connectedness With the Self

The first theme represents expressions of spirituality that involve connectedness to the self. This theme manifests as an interest in and pursuit of inner knowledge of the self, despite the changes that accompany older age. The theme has four subthemes: continuity of the self, positive solitude, self-care, and an integrative view of the self.

Continuity of the Self

The first subtheme refers to participants' perceived sense of self-continuity in older age, which was stimulated during the creative process. This subtheme was widespread in both the Israeli and Italian samples.

Elena, an 86-year-old Italian woman, chose the image of a candle (**Figure 2**) to represent her personal legacy. In describing her choice, she discussed how a sense of self-continuity helps her cope with facing the end of life:

@www.istockphoto.com

FIGURE 2 | Elena's chosen photograph and title: "The beauty of old age."

> That light over there, it looks a bit like my life. It's a candle [. . .] that has almost burned down. But it still makes light. And it leaves a trail [. . .] This ray of light, I like it [. . .] What I left behind, the things I've done and that I left as something I've built. That therefore remains, [it] doesn't go away [. . .] Good always remains, [the good things you have done] are not lost [. . .] The beauty of life, the beauty of old age. This is the last part of the candle, there

> is not a candle [anymore]. And the light is vitality. And those glowing red shadows that are behind, I see them as the love I left along the way, and love is the most important thing.

Miriam, an 82-year-old Israeli woman, selected a photograph showing a sheet of music to represent her personal legacy. She explained that the photograph reflects her appreciation of music, a legacy passed down by her parents which remains a key part of her identity. In her photocollage, Miriam positioned this photo next to another one representing a joyful family, and explained: "It looks like the family is dancing."

> Ah, music. plays an important part in my life. Mm. As long as I can remember, I took music lessons. I sang in the choir. I took violin lessons and, you know, and right now, first thing in the morning, I'm going to turn on some jazz, and listen to it peacefully [. . .] I love it, it played a major role in shaping our whole family life. My mother [a Jewish American] always sang in [a local] choir. My father sang in the choir. [Laughing.] My sister played for the choir. We all took piano lessons [. . .] Music is the central, has a central part in my life.

Positive Solitude

The second subtheme, positive solitude, refers to participants' ability to enjoy engaging in activities on their own. Interestingly, the positive solitude theme emerged among the Israeli participants, but none of the Italians. We will return to this difference in the discussion.

Rivka, an 80-year-old Israeli woman, included photos which represented her love of art, books, and the chess her father used to teach her in her photocollage (**Figure 3**). While reflecting upon her creation, she discussed how these photos also captured how her positive approach to solitude helped her cope with the COVID-19 lockdowns:

> Coping, undoubtedly, there are a few [photos] here. There is "toil." And "studying" and "chess," [these] are three of them [pointing to three photos in her photocollage]. [. . .] This means that even in such situations we need to find a way to do things by ourselves. [. . .] I really did not suffer during the whole Corona period [. . .] even though our social life was very very active [before the pandemic] [. . .] I found myself doing things alone and did not get bored for a second this year [. . .] Even though [. . .] I had a foot problem and I had to stay indoors for three months until this problem resolved [. . .] Even in this restricted situation I think I did not cope badly.

Self-Care

The third subtheme represents the ability to care for oneself—i.e., to understand what is best for the self and to make life choices that provide one with autonomy. In this study, self-care and the desire for autonomy were identified only among the Israeli participants. Itzhak, an 85-year-old Israeli man, chose the image of a telescope to represent his determination to take care of himself:

> I am optimistic. This keeps me going [. . .] Like in this picture with the telescope [. . .] I don't give up. I try to cope on my own and not be dependent on others. I do laser treatment every day to cope with the pain in my back and shoulder. Every day I do the treatment. I cope. Before I could not lift my arm, now see [shows how he lifts his arm]. Maybe it is the treatment and maybe it is my desire to succeed and to get better results. I am optimistic and I believe that within a month or two I will get to even better results than today.

Integrative View of the Self

The fourth and final subtheme under connectedness with the self-concerns an ability to integrate and accept different parts of the self, both positive and negative.

In his first collage, Aron, an 80-year-old Israeli man, composed a row of three photos, which he said symbolized his relationship with his father. One photo represented the disappointment he felt his father perceived in their relationship, while the other two represented love and pride. Aron explained:

> One of the things that most disappointed my dad, [. . .] he was also a chess player. He tried to teach me chess. And I was not attracted to it, as he had hoped for, and I know it terribly disappointed him [...] so this picture symbolizes disappointment... and here you add the picture with the books, he really liked books and my whole world was books. I read a lot. And he was always proud that I read like this. [...] And here is this picture of the porcelain [representing the factory where he worked]... and my father was very proud of his work, and I visited the factory, I was very proud of him and their achievements... So here we have in one line three pictures that actually relate to memories from my dad.

In integrating these contradictory aspects of his relationship with his father—disappointment, love, and pride—in his collage, Aron was able to accept both the positive and the negative as building blocks of his own identity.

Indeed, the use of collage is particularly suited to formulating and expressing an integrative view of the self, since collage itself is a medium in which distinct elements are crafted into a new whole. During the creative process, participants were invited to think about how different visual images might represent different feelings and life experiences. The images participants incorporated into their photocollages often captured contrasts and contradictions—sweet childhood memories along with representations of traumatic events, portraits of newborns next to photos of older adults, images of antiques and old-fashioned handicrafts juxtaposed with images representing modern high technology.

Caterina, an 87-year-old Italian woman, began to reflect on a photo that she saw as representing friendship. This prompted her to speak about her self-acceptance:

> I have a lot of friends, people who love me, old friends of mine. They really give me joy and remind me of the fact that, sure, I could have been better in my life, behaved in a better way, but you can see that a lot of things [I did] made other people happy. Then sure, I've made a lot of mistakes. And I made mistakes on a lot of things, I can see that, but I've always tried to fix them and to feel at peace with myself. [. . .] It's true, when you get to a certain age, you think about the life you've lived. And you say I did this and that wrong. But I did a lot of other things right. I justify it to myself, you know, if I did something wrong.

We can also see here how the spontaneous, creative nature of the collage-making process, and the tangible nature of the visual images that we presented to participants, allowed different domains of spirituality to link up and overlap with each other. In

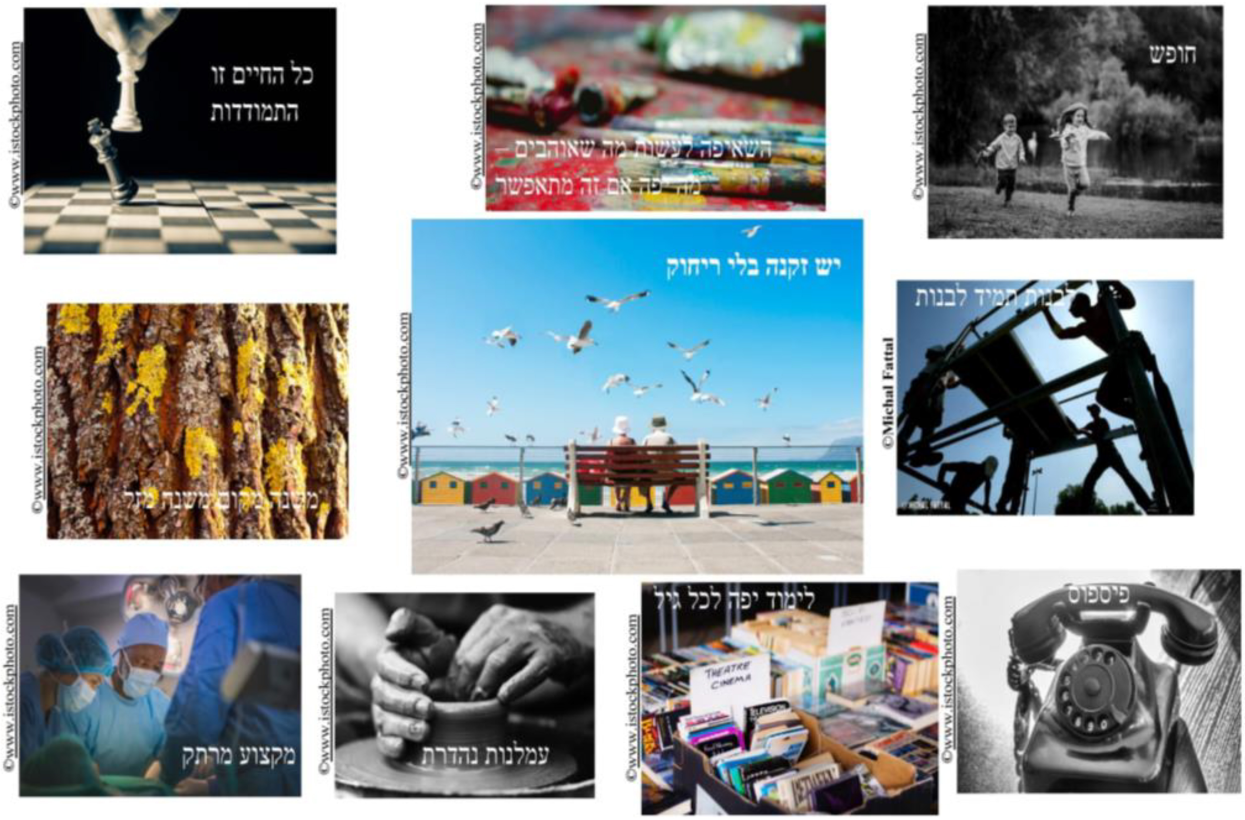

FIGURE 3 | Rivka's photocollage.

Caterina's reflections, an image which represented connectedness with others stimulated a more integrative view of herself, in which she was able to accept different pieces of her life.

Connectedness With Others

The second theme, already mentioned, represents expressions of spirituality that involve connectedness with other people. Three subthemes were identified within this theme: connectedness with significant others, a sense of contributing to others, and caring for humanity.

Connectedness With Significant Others

A major subtheme concerned participants' experiences of love and caring within their most proximal relationships. The most important of these relationships was often with a spouse. Many of our participants reported that couplehood played a major role throughout their lives. Asked to describe how she coped with aging and during the pandemic, Margherita, an 83-year-old Italian woman, chose a photo of two elderly people walking side by side (**Figure 4**):

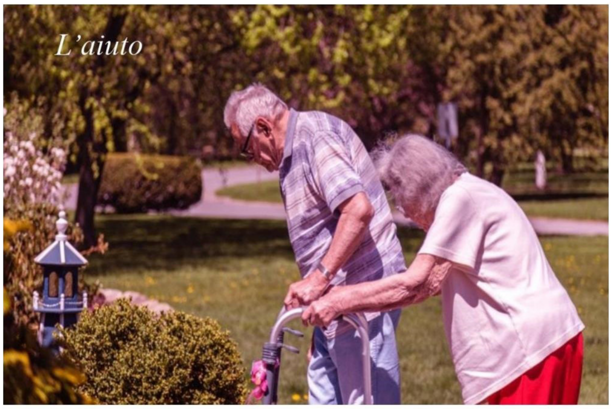

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FIGURE 4 | Margherita's chosen photograph and title: "The support."

> A love for life. I've been with my husband for my whole life. You know, at the age of seventeen I was already engaged. I got married at 24. It's been 59 years that we've spent together. We are married. Isn't it beautiful? You know why it's beautiful? Because there is love. [. . .] Yes, I see a value in this photograph because there is a person who is alive, and that's one thing, and second thing, you are hugging someone who's alive too. Then, you walk badly, you have a cane but you have a smile that keeps you going [. . .] being with my husband. Being together, the two of us. [. . .] In this difficult time [the pandemic], being together, husband and wife, pretty old [. . .] It helps me get through this period. [. . .] When you are two, it's easier to solve things.

Another important source of connectedness with others was friendship. Lea, an 83-year-old Israeli woman, described how friendship supported her in her childhood as a Holocaust survivor, and how friendship was important during COVID-19. From a photo of a snowy landscape that recalled her childhood memories as a Holocaust survivor in a boarding school, she shifted to a photo of migrating birds, which she saw as representing a sense of togetherness in life (**Figure 5**):

> I will take this picture, of the snow [. . .] It was in the Italian Alps. I was in a boarding school for Holocaust children like me [. . .] and I remember it snowed [. . .] And we played in the snow, and we were really happy. Snow symbolizes purity, something clean and pure and beautiful. We really had good time in this boarding school [. . .] So, at first I did not open up, I was very closed. But I met a friend there, a little older than me, and we were very good friends. And I really loved her, the way I love myself. What's happening now [the pandemic], all these fears, the anxieties, about Corona. So, a lot of people close themselves off, I closed myself off a little too, a little too much. [Short laugh.] And I remembered, in life I had many difficulties, during the Holocaust. And it helped me [being with others]. And it's the same [. . .] [points to the picture of the birds]. And here they [the birds] migrate, migratory birds. Together. And I guess if one is sick, they might help it.

Here again we can also see how the spontaneous flexible nature of the creative process allowed participants to glide smoothly from one domain of spirituality to another, revealing as well how the domains are inextricably connected. The photograph with which Lea began—the snowy landscape—reminded her of her connection to nature, her awe at the beauty of the snow. This intuitively led her to the memory of a good supportive friendship, which she represented by the image of the flock of birds.

Participants' musings and reflections often suggested the importance of their role as grandparents and their connectedness with their grandchildren. Caterina, an 87-year-old Italian woman, related to two different photographs to describe her relationship with her grandchildren, who infused her with hope for the future (**Figure 6**). The first depicted a grandmother laughing with her grandchild, and the second a young girl whose gaze reminded Caterina of her grandchildren:

> This grandmother [in the photograph] seems almost complicit with this child, she's got a sweet gaze, and these joys, you have to cultivate them. It's the wish to have children and them also to have grandchildren to love. Because I notice that when I look at my grandchildren, I don't care if they are good, I look them in the eyes and I see everything positive. That is, they help me think of a better future, I mean, a future for them [. . .]. This child's eyes [in the photograph] give me joy, hope for a future for everyone. For everyone, for the world, for us. They give me hope, I don't know. It's a feeling I have.

This subtheme also captures participants' experience of accepting other people, and their ability to forgive others despite previous difficulties in close relationships. Hana, an 87-year-old Israeli woman, talked about her experience of forgiving her mother. When describing this experience, she related to a personal photograph of her mother and titled it "Bella Bellissima":

> Here [looking at her first photocollage], I can see my anger. . . But from the perspective of time one can no longer be angry. It's a replica of it [my anger], or a memory of anger. Whoever keeps on being angry at their parents and blames them is wrong. [One] should come to a resolution. So . . . there's a picture of my mom above me here. And I call her "Bella Bellissima."

Sense of Contributing to Others

Another subtheme found among many participants captures their experience of helping other people, and how this gave them a sense of contribution and meaning in life. Itzhak, an 85-year-old Israeli man, selected a photograph showing two sets of hands, an adult's and a child's, cupping a luminous heart as the central image for his photocollage (**Figure 7**). He said he liked to sew for those in need and that helping others is something that gives him a sense of joy and meaning:

> Itzhak: In the center [of the photocollage]. This is it, this is it. Love, well [. . .] the heart. The heart that illuminates our future. Lots of hearts, one heart, one big light.
>
> Therapist: But from the things you have said [. . .] that there will be true love everywhere [. . .] Is there anything that helps these things happen?
>
> Itzhak: I have a sewing machine. That I help people, help those who need. I sew for them [. . .] I sew pillows, masks. Whatever comes [. . .] Whoever comes to me, "Something tore," and I say "Bring it!" [. . .] I love it, I love to be told "thank you very much." It is enough. It is joy in the heart. I never take [money]. I do not ask for a penny. I always help. From the heart. And I want more and more and more [. . .] If I'm healthy I'll do anything.

Giovanni, an 88-year-old Italian man, chose a personal photograph depicting himself as a musician along with other musicians and friends doing volunteer work in a nursing home. He positioned it next to another photograph of musicians and called it "Donating a smile":

> As my hobby, we often went to nursing homes to make them smile. . . Ever since I went back to music when I retired. It was in the nineties. We were a group of five people. And we went there to have them smile. . .If it wasn't for Covid, this would have been my future.

Caring for Humanity

The final subtheme under "connectedness with others" represents a sense of connectedness and respect for humanity more generally.

Rivka, an 80-year-old Israeli woman, related to COVID-19 when she chose a photograph of a tree growing at the edge of an abyss to represent her hope for humanity (**Figure 8**):

> This is also a beautiful picture, the roots. The picture with the tree and the roots in the rock. A beautiful and interesting picture. It's like I think the world is on loan, yes? That's what I say. The world is on the edge of an abyss, but our roots, humanity, perhaps the roots of the founders of the land are deep and strong [. . .] The roots that have grown throughout history are so deep that even on the edge of an abyss [. . .] [She titles the photo] "The world is on the edge of an abyss, yet the roots of humanity are deep."

Mary, a 92-year-old Israeli woman, chose a photograph of an airplane, which she titled "Openness to culture," to represent her

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FIGURE 5 | Lea's photographs and titles: "The first snow" and "Birds migrate but together."

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FIGURE 6 | Caterina's chosen photographs.

concern for humanity, her acceptance of other cultures, as part of her sense of universalism as her legacy.

> Yes. This one for me is important [the airplane] because it symbolizes travel. And, for me I think it's important that people, um, have an open mind about other people and other cultures. Because very often people think that yeah, my way is the only way forward. I think it's important for people to know how many different ways people live [. . .] I think people would get along better if they understood that other people have other ways of dealing with their life. Otherwise, I think a lot of misunderstandings and maybe arguments on being distanced from other people comes from the fact that they aren't open to see how other people live.

Both Mary and Rivka were prompted by the creative process to articulate profound feelings about the future of humanity. In so doing, the process also helped bolster their dignity by enabling them to pass on their wisdom at the end of life.

Connectedness With the Environment

The third main theme relates to feelings of connectedness with nature and with other living creatures.

Yossi, an 80-year-old Israeli man, selected an image of a man, seen from behind, gazing out over a wintry landscape (**Figure 9**) to represent the value of profound connectedness with the world. The image allowed Yossi to relate COVID-19 as a transient event:

> A person looks toward the horizon [. . .] In all this, here he is looking at the sky. He does not look at the sky, he looks at how the future changes every second [. . .] Look, he will be relaxed, sitting, looking at the view. This is a very, very worthwhile thing to learn in life. [. . .] He is looking toward the landscape. He not only looks at a landscape, he looks at how the world grows [. . .] He sees it all. This is a very beautiful thing. a full world he has. He has a full world [. . .] This Corona will be gone, but look at this landscape [. . .].

Many of our participants talked about nature and respect for all living creatures as a coping resource to deal with the COVID-19 pandemic. Miriam, an 82-year-old Israeli woman, put it this way:

> I think they should learn the intended lessons of the importance of respecting the environment, all of the live things that surround us, the animals, the trees, the people and the importance of remaining positive in your thinking and your actions [. . .] It's the only thing that will help cope with current situation [the pandemic]—identifying what caused it and what will fix it. There are lessons to be learned [. . .].

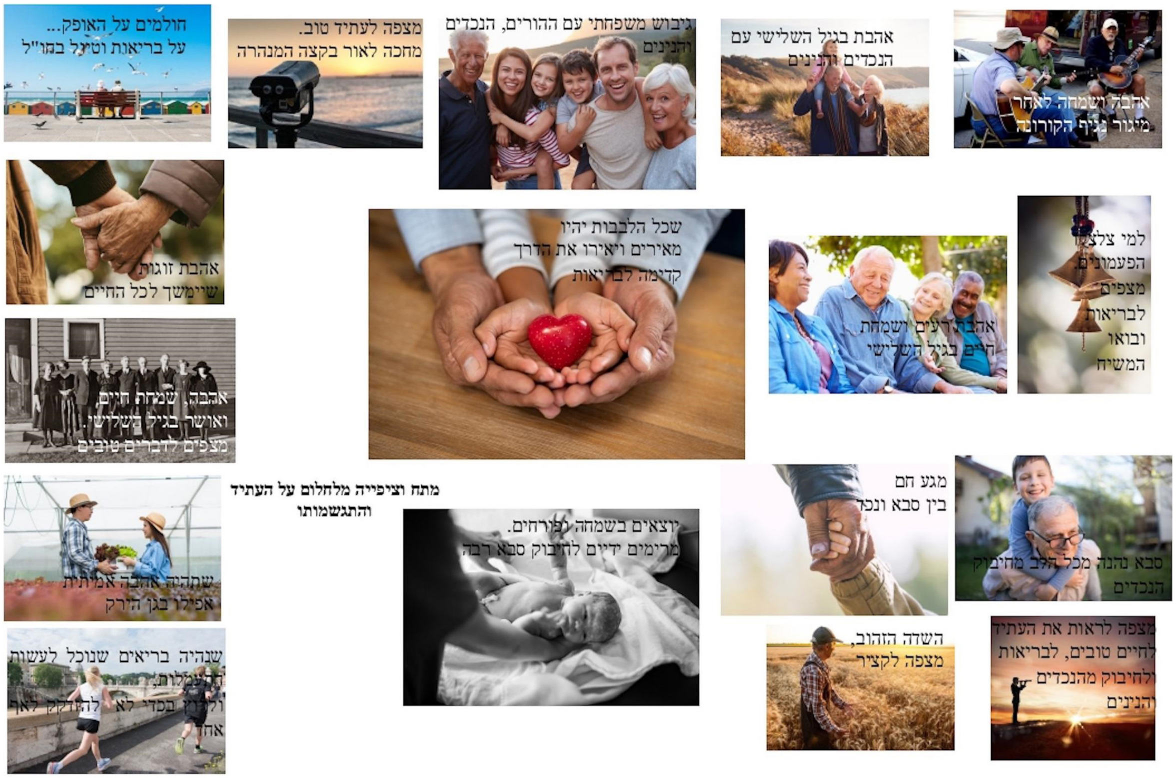

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FIGURE 7 | A photocollage by Itzhak: "Tenseness and anticipation while dreaming about the future and its fulfillment."

Nature can also symbolize connectedness to other generations and the deceased, as Tova, an 82-year-old Israeli woman, explained. We may note here again how the environmental domain overlaps with the communal. Just as connectedness with nature linked Lea to her memories of friendship, for Tova a connection with nature helps her cope with the loss of her beloved husband:

> I'll tell you a secret, I have a tree by my house, that we planted, me and my husband. And it's like, I wouldn't say a tombstone, but for me, it's my life with him, as if he is still alive within the roots of the tree. And when pigeons come, I speak to them, all the time, as if he is alive. As if he can hear me. And I ask for things. Maybe it is not normal, but it is what it is.

This theme also includes elements of gratitude and appreciation for the simplest things in life. Francesco, an 85-year-old Italian man, when asked to think of a title that would summarize the essence of the three photocollages he created, proposed "The sun gives you the strength to live":

> Because I can't have a future at my age [. . .] what's important—I always say—is that if I wake up in the morning, I'm already lucky. [. . .] Personally, I'm greatly in love with life. As a consequence when I see the sun, I'm joyful and I think for a person after 80, 85 years of age everyone's wish is to wake up in the morning and see a radiant day.

Connectedness With the Transcendent

The final theme represents spirituality as expressed in connectedness with a transcendent reality or power. The Italian and Israeli participants related differently to this theme. As nearly all of the Italian participants in this study were religious Catholics, many chose images that either had an explicit religious theme or could be interpreted as religious; and they positioned those images more centrally than the Israeli participants. As such, the Italian photocollages often related to faith and religiosity as something that one can cling to during times of adversity. Margherita, an 83-year-old Italian woman, chose a photograph of a church to represent a hope that both she herself and younger generations would keep faith as a coping resource. She called this photo "For a better future":

> You know what helps me [while coping with the COVID-19 pandemic]? [. . .] See, if I go to mass, I come home and I'm happy. I listen to the mass, I resonate, I join the singing [. . .] I'm happy, I don't know why, but I feel happy. I feel happy when I do that [. . .] I feel like I've done something so beautiful. Now I'm used to it but the church for me is everything. [. . .] Because when you have something [to worry about, like the pandemic], you must pray. Because praying gives comfort, even if it does not solve the situation, it comforts you.

Elena, an 87-year-old Italian woman, chose a photograph of a couple looking at the sunset to talk about how she and her

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FIGURE 8 | Rivka's chosen photograph and title: "The world is on the edge of an abyss, yet the roots of humanity are deep."

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FIGURE 9 | Yossi's chosen photograph and title: "He has a whole world—it is worth learning to observe the world."

husband discovered their spiritual dimension. She explained how faith helped her to cope when her husband was ill in cancer.

> Religion can be a sum of rites, of habits. We [my husband and I discovered faith…which brought us to experience a lived spirituality […] and this allowed us to go through a huge trauma when my husband was ill with cancer for a couple of years. […] This allowed us both to be strong and to create an environment of serenity around us and this I owe it, we owe it, to faith. […] The liturgy gives a certain lightness to the heart which is incredible. We derive our strength from it. The strength to live every day with serenity and with a smile. Even during hard times. Since I have been alone, this source of support has been fundamental, as it has allowed me to live my life with a smile.

She later added how her faith helped her to accept the difficulties and restrictions that followed the COVID-19 pandemic. She positioned a photograph of a cross above all the other photos in her photocollage, explaining that it was the most important one and that she wanted it to embrace all the others:

> I'm not worried [about COVID-19]. Because, you know, man's biggest fear is death. This is our destiny, there's nothing we can do about it. […] With this serenity, I must say, I live. And when the moment comes, I hope I will be able to accept it.

For the Israeli participants, traditional Jewish rituals and prayers served as a path to the transcendence domain, rather than religious faith itself, as presented by the Italian participants. Moshe, an 81-year-old Israeli man, positioned the photo depicting a candle (**Figure 10**) to describe how the Jewish ritual of lighting candles on Friday night helped him to cope with the absence of his wife. This is another example, too, of how the domains of spirituality sometimes overlap. For Moshe, a connection with the domain of transcendence created a path to the presence of his deceased wife.

> I think the candle [should be in the center]. I light the candles on Shabbat […] and I have a special candle for my wife. With even a special place [for it]. I always light it, in exactly the same color, and it looks the same. So, it gives me a feeling of not being alone […], it's not that she's really there or something like that, but I've been doing that for a long time, every Friday. Like it's part of a ceremony […] I'm doing it all by myself [crying]. The candle lighting, the "kiddush" [prayer over wine] [Crying.] […] It's only my voice inside the living room, when I do the kiddush […] but sometimes I feel her presence […]

Next to this photo, Moshe positioned the photograph of a man gazing out over a solitary landscape and added: "And the part where I sit there alone, put it [in the photocollage] […] next to the candle." He also asked us to include the poem by Holland (1910) in the collage, to further represent and reflect the continuous bond with his deceased wife.

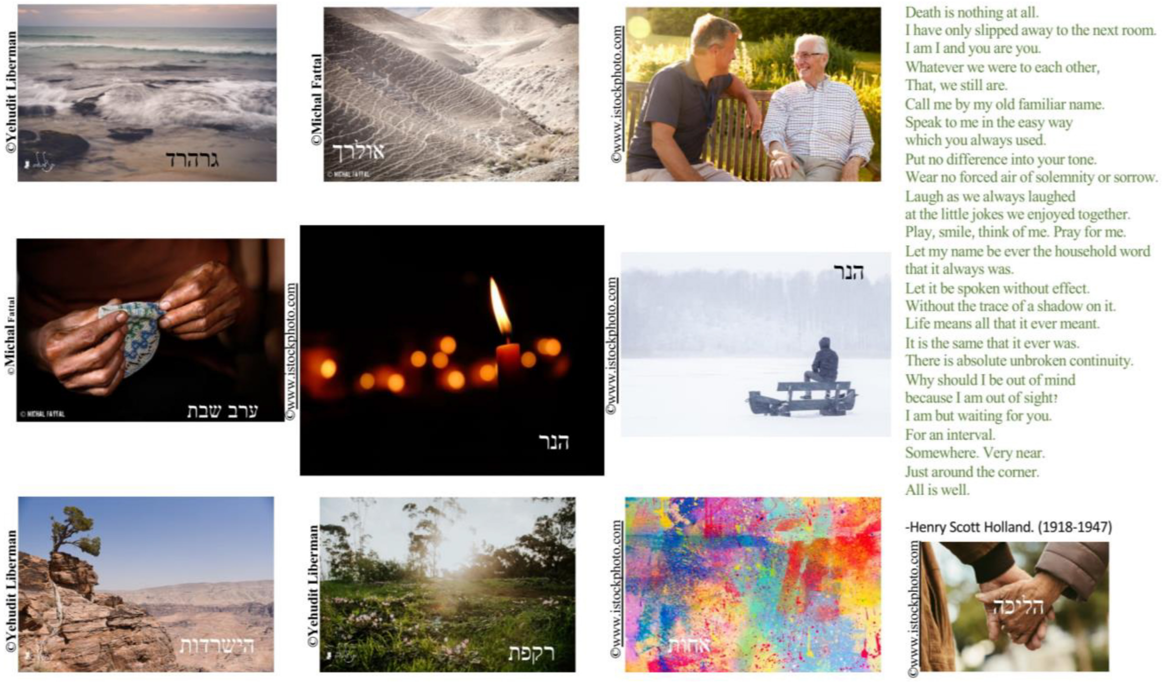
Noah, a 91-year-old Israeli man, chose a photograph of perching birds (**Figure 11**) to represent his appreciation of life and nature. He also linked this image to a Jewish prayer which expresses gratitude for being alive:

> Very beautiful, very beautiful. The birds, they don't ask themselves what tomorrow will be. Someone could come and shoot the bird and it would be gone […]. It's to appreciate being alive. […] Like in the Jewish morning prayers we say thank you to God for returning our souls to life. To be grateful.

DISCUSSION

This study developed and examined an online creative arts intervention aimed at helping older adults connect to their spirituality, and thereby support their sense of dignity and their coping resources at a time of social restrictions imposed by the COVID-19 pandemic. The intervention focused on the creation of photocollages integrated with narrative elements of dignity therapy (Chochinov, 2002). We examined how the creative process connected the participants to different domains of spirituality by exploring their life experiences, values, and future perspectives through the creation of three photocollages.

Four themes were generated, representing four domains of spirituality that were stimulated and expressed by the process (**Figure 12**): connectedness with the self, connectedness with



Death is nothing at all.
I have only slipped away to the next room.
I am I and you are you.
Whatever we were to each other,
That, we still are.
Call me by my old familiar name.
Speak to me in the easy way
which you always used.
Put no difference into your tone.
Wear no forced air of solemnity or sorrow.
Laugh as we always laughed
at the little jokes we enjoyed together.
Play, smile, think of me. Pray for me.
Let my name be ever the household word
that it always was.
Let it be spoken without effect.
Without the trace of a shadow on it.
Life means all that it ever meant.
It is the same that it ever was.
There is absolute unbroken continuity.
Why should I be out of mind
because I am out of sight?
I am but waiting for you.
For an interval.
Somewhere. Very near.
Just around the corner.
All is well.

-Henry Scott Holland. (1918-1947)

FIGURE 10 | Moshe's photocollage (untitled).

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FIGURE 11 | Noah's chosen photograph (untitled).

others, connectedness with the environment, and connectedness with the transcendent. We showed how each domain was stimulated and constructed through the visual images (photographs) used in the intervention, the narratives that were prompted by the photographs and the protocol's themes, and the creative process of selecting and positioning photographs to create a new art product (a photocollage).

The expressions of spirituality generated from the intervention point to the multidimensional nature of spirituality (Fisher, 2010; de Jager Meezenbroek et al., 2012; Puchalski, 2012). The spontaneous nature of the artistic process also revealed how the different domains of spirituality are interlinked. The projective visual stimuli of the photos elicited personal content as a form of "bottom-up" spontaneous thinking (Yaniv, 2018), through which participants' reflections in one domain prompted and nourished reflections in another (**Figure 12**). For instance, as we saw with Lea, the photo of the snowy landscape stimulated her connectedness to the environment through her memories of wonder at the snow as a child Holocaust survivor, and by extension it stimulated her connectedness to others when it aroused the memory of a good friendship at that time of adversity. In other cases, different domains of spirituality actually overlap, especially where the older person has lost a significant other, such as a spouse. For example, Tova and Moshe found that connectedness with the environment (Tova) or with the transcendent (Moshe) helped provide comfort and a sense of remaining connected with the absent other. This observation is consistent with other studies' findings of correlations between the domains of spirituality (Lifshitz et al., 2019).

The findings suggest that in most respects, the Italian and Israeli participants relied on the different domains of spirituality in similar ways, both when coping with the pandemic and when facing other struggles of life, including the challenges of aging. However, there were several differences between the two groups. In particular, the Italian participants in this sample were all Catholic, and most of them considered themselves to be religious. This was reflected in the Italian photocollages, in which religious images were more often positioned centrally. The Israeli participants, who tended to be more secular, were more likely to refer to their connectedness with and reliance on the self, and to point to positive solitude and self-care as coping resources.

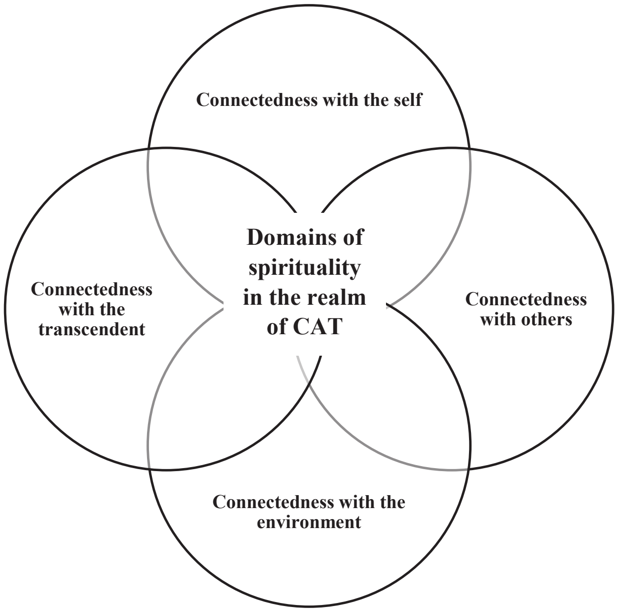

FIGURE 12 | Visual representation of the interconnected nature of the domains of spirituality in CAT.

Current Findings and the Literature

The expressions of spirituality stimulated by the creative process capture the multidimensional nature of spirituality as shown in the literature (Fisher, 2010; de Jager Meezenbroek et al., 2012; Puchalski, 2012). This is one of the first studies to use visual artistic methods to explore these dimensions with the older population. The findings show how photographs, as projective stimuli, allow for intense spontaneous expressions of spirituality in a manner that is flexible and responsive to different needs and perspectives. This is important, because other studies' findings show that maintaining a connection to different dimensions of spirituality is associated with better mental health and coping in times of adversity, both in general (Whitehead and Bergeman, 2012; Thauvoye et al., 2018; Lifshitz et al., 2019) and specifically during the COVID-19 pandemic (Ribeiro et al., 2020; Lucchetti et al., 2021; Keisari et al., 2022c). The ability of the intervention to connect the participants to the various domains of spiritually as coping resource is crucial, since studies have shown that the COVID social distancing restrictions caused psychological distress for many, including more symptoms of depression (Fountoulakis et al., 2022; Keisari et al., 2022a).

The current findings contribute to the growing body of studies that use photographs with older participants to prompt greater expression and deeper exploration of the participants' thoughts, feelings and experiences (Kohon and Carder, 2014; Tishelman et al., 2016; Platzer et al., 2021). While most studies in the field use the *photovoice* method, which refers to the participants' own production of photographs in relation to the research topic (Baker and Wang, 2006; Woda et al., 2018; Chui et al., 2020), or *photo elicitation*, which refers to the use of photographs in qualitative interviews to encourage participants to reflect on particular topics (Platzer et al., 2021), the current study with photocollage uses both ways: the photographs served as projective stimuli to elicit the personal content, and as raw material collected by the participants to make their own artistic product. This stimulated personal content, while also capturing and containing it in a concrete artwork that the participants could then create, observe and reflect upon (Keisari et al., 2022b).

The differences that were found between the Italian and Israeli participants in this sample mirror the familiar and well-established observation that within secular Western societies, people are likely to search for meaning in life within themselves, rather than on the basis of external rules or a connection with a higher entity (Heelas, 2000; de Jager Meezenbroek et al., 2012). These findings also point to the importance of considering intercultural differences in spirituality, and in the dominance of different spirituality domains (Thauvoye et al., 2018; Lifshitz et al., 2019), when designing interventions aimed at offering or strengthening spiritual care.

To the best of our knowledge, this is one of the first studies to develop an online creative arts intervention for older adults, which can be conducted during periods of social distancing (Kordova and Keisari, 2020; Feniger-Schaal et al., 2022). As such, the method goes beyond the current COVID-19 pandemic, as it can also be applied in other circumstances that entail social isolation—for example, among individuals who are homebound or have limited mobility (Milaneschi and Penninx, 2014). This creative online intervention can also be implemented in palliative care, as it is based in part on dignity therapy, known to make a key contribution to the wellbeing of individuals at the end of life (Chochinov, 2012; Fitchett et al., 2015; Testoni et al., 2019a). The current findings are also in line with studies indicating that interventions based on dignity therapy support the existential tasks faced by the majority of older adults, such as settling relationships, and preparing legacies of memory and shared values, as well as preparing for the end of life (Fitchett et al., 2015; Scarton et al., 2018).

Finally, the research methods and findings presented here can enrich narrative therapy protocols for the aging population, such as dignity therapy (Chochinov, 2002) and life review (Westerhof and Slatman, 2019). We found that both engaging with the individual images in the photographs, and the act of meaningfully positioning them in photocollages, helped participants find spiritual meaning in their personal narratives. Thus, narrative therapy protocols could benefit from being expanded to include the visual and creative elements of photocollage.

Limitations and Future Research

By definition this study consisted of a sample of older adults with access to the Internet and videoconferencing applications. This sample is thus not representative of other groups of older adults experiencing high levels of social isolation during the COVID-19 period, such as nursing home residents, individuals experiencing cognitive decline, or those lacking access to the Internet. The short-term nature of the intervention is also a concern. As mentioned, most of the older adults in the current study were living alone during the social restrictions of COVID, and a longer-term process might have been more beneficial for them.

Future research would benefit from replicating the current intervention with various aging populations, such as older adults with dementia, and in different care settings, such as nursing homes. It is also important to examine the experience of participating in a long-term intervention as a creative process that encourages learning and growth over time. Future studies might also examine the effect of longer-term creative online interventions on various wellbeing measures.

CONCLUSION

The objective of this study was to explore how an online creative arts intervention involving the creation of photocollages promoted spiritual care in older adults, and which domains of spirituality were stimulated and expressed. The findings highlight the importance of considering the multifaceted nature of spirituality when using online creative arts therapies to help older adults cope at time of adversity. More broadly, the findings suggest that online creative arts interventions are valuable for older adults during periods of social isolation.

DATA AVAILABILITY STATEMENT

The datasets presented in this article are not readily available because it contains potentially identifying and personal information. Therefore, the data will be kept confidential under the researchers' control. Requests to access the datasets should be directed to the corresponding author SK.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Ethics Committee for Experimentation, University of Padua (confirmation number 0581B1B9C39761AE3C03AD3D93EFDEE9) and by the Ethics Committee of the Faculty of Social Welfare and Health Sciences at the University of Haifa, Israel (confirmation number 366/21). The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

SK and IT conceived, designed the study, and supervised the research team. SK, SP, TE, and GM conducted the intervention, collected, and analyzed the data. SK and SP took the lead in writing the manuscript. HO and IT reviewed the thematic map and the manuscript and helped to improve it. All authors contributed to the article and approved the submitted version.

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SUPPLEMENTARY MATERIAL

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Death cafés as a strategy to foster compassionate communities: Contributions for death and grief literacy

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Introduction

The death-positive movement, the most recent manifestation of the death awareness movement, contends that modern society is suffering from a "death taboo" and that people should talk more openly about death (Koksvik and Richards, 2021). This movement is striving to shift the dialogue about (and place of) death and dying into community spaces (Breen, 2020).

People are dying at older ages and over longer periods of time, as a result of chronic disease trajectories and advances in medical interventions, generating new demographic, and epidemiological trends. In many circumstances, death and dying processes are over-medicalized due to aggressive treatments and practices in hospitals and residential eldercare facilities (Becker et al., 2014). Most deaths happen within such institutions, leaving communities frequently "in the dark" regarding processes of care and illness at the end of life (Breen, 2020). There is a widespread belief that community-based solutions in palliative care and support for the bereaved are needed (Richards et al., 2020). However, as argued by Park et al. (2022), scant attention has been given to community-level interventions for death, dying and grief, or to the public's readiness to fully participate in these interventions.

The COVID-19 pandemic highlighted the global need for communities to be prepared for illness, death and grieving. As a result, the general population and health and social care professionals became keenly aware of a variety of issues connected to mortality and the end-of-life, challenging tendencies to avoid discussions about death and dying. Two related concepts attempt to counteract this reluctance to consider or discuss death: death literacy and grief literacy.

Death literacy is defined as a set of skills and knowledge enabling people to learn about, understand, and act on end-of-life and death-care options (Noonan et al., 2016). People and groups with a high level of death literacy have a context-specific comprehension of the death system and can more easily adapt to it, becoming better equipped to provide care for others or to gain access to critical services necessary for high-quality end-of-life support (Noonan et al., 2016).

Grieving often accompanies loss, which is typically, but not always, tied to death. Grief literacy has been defined as knowledge, skills, and values that promote compassion for self and others in the face of loss (Breen et al., 2022). The aims of the grief literacy movement are to understand and normalize grief, improving everyone's ability to recognize and effectively respond to loss. Increased public awareness of loss will organically encourage greater emotional and practical support around grief, with clear benefits for the bereaved as well as frontline health and social care professionals, who often have limited time and resources to sufficiently attend to these issues. Grief literacy can be viewed as a natural extension of death literacy, and increasing individual capacities in both areas may help reduce the effects of death avoidance and death anxiety (McClatchey and King, 2015).

One approach that may enhance both death and grief literacies is the Death Café. Inspired by the work of Crettaz (2010), Death Cafes are a global social franchise, with locally-organized public events that encourage discussion of all aspects of death, dying and mortality. These discussion events, often taking place in cafes, restaurants, libraries or other public spaces and facilitated by volunteer hosts, adhere to one vital rule: cake or other culturally-appropriate celebratory foods should be enjoyed by participants to honor our precious "finite lives" (Death Café, 2022). The Death Café approach, currently used in more than 81 countries worldwide, allows individuals to discuss difficult topics, increasing their self-awareness, potentially reducing death anxiety, and augmenting compassionate connections through conversation and personal sharing (Miles and Corr, 2015; Fong, 2017; Chang, 2021).

We argue that the Death Café approach can be a useful strategy to improve both death and grief literacy levels and may help promote the burgeoning concept of compassionate communities as part of palliative care (Graham-Wisener et al., 2022).

Death cafés and compassionate communities

The Death Café movement, through its promotion of open discussion about the human realities of death and grief, has strong links to the compassionate community model. Public Health Palliative Care International (Compassionate Cities, 2022) defines a compassionate community as a community development initiative associated with global palliative care. A compassionate community consists of groups of neighbors/members of a community who come together to help people in their network living with a terminal illness, along with caregivers and the bereaved. Compassionate communities attempt to follow the 95% rule, wherein the dying person spends only 5% of end-of-life with formal medical services and the remaining time with their caregivers and connections in the community (Kellehear, 2013). Based on the premise that death and dying are shared communal concerns, care networks are created from within the existing community to support its members. Compassionate communities are framed as a public health response to death and bereavement, raising public awareness, and preparing people for death by discussing their wishes in advance and fostering compassionate behaviors toward those in the community who are nearing death or have recently experienced bereavement (Koksvik and Richards, 2021). Innovative community-based strategies, such as Death Cafes, may appeal to a wide variety of groups within the population, and can be adjusted to fit local culture and practices. The Death Café model allows for informal sharing of intimate stories and experiences within a social and convivial atmosphere (Leland, 2018). These kinds of activities may help community members better engage with the Compassionate Communities ethos (Liu et al., 2022).

Kellehear (2020) developed a model of person-centered care and the concept of Compassionate Cities for end-of-life patients. The person is surrounded by circles of care, namely (outward from the center): a closer internal network; supported by a wider external network; then the community, social and health services; and, lastly, the local and national authorities. When the Compassionate Community model is followed, family members and caregivers are more resilient and less exhausted during an individual's final days of life, and the dying person can have a better quality of life at home (Librada-Flores et al., 2020). Community support for patients and their caregivers is performed through tasks, such as spending time with the patient (e.g., reading a book, talking) or providing direct care, thus allowing the informal caregiver to rest or perform tasks outside their caregiver role. The care network may also assist by completing household chores, such as cooking, cleaning the house, pet care, shopping, etc. The idea is to create an intentional, but natural, external support network that enhances the direct care system for the individual (Librada-Flores et al., 2020). This intentional network, forming the circles of care around the dying and the bereaved, requires a high degree of comfort with intimacy and trust in the ability of community members to both offer and accept care from non-professionals during a vulnerable time in life. We believe that the personal sharing and interaction that takes place at Death Café events can help build the foundations of compassion and comfort that, over time, will allow this exchange of care to occur more naturally in groups and communities.

More compassionate connections within a community, augmented by initiatives like Death Cafes and Compassionate Communities strategies, should result in a better quality of life and death for all (Richards et al., 2020). Darwin suggested morality as the defining feature in the human-animal divide, and it can be argued that compassion within a human community is a powerful variable in survival (Wilson, 2019). Hypothetically, a person integrated into a more compassionate and generous community may survive longer and thrive within a high-quality social system. Opportunities to build compassion, and understanding and openness about death and grief within heterogeneous and constantly-evolving community structures are crucial to improving end-of-life and bereavement care for all (Abel, 2018).

Death cafes: Sharing stories and conversations about loss, death, and grieving

The Death Café model encourages the relaxed mutual sharing of personal memories, thoughts, beliefs, and feelings about mortality and grief (Chang, 2021). Participants tell stories about their experiences of losing loved ones and funerals they have attended, anticipate their own deaths, discuss preferences for their own death, beliefs in the afterlife and many other aspects of death, dying, and bereavement. Recalling these experiences, articulating a narrative from them, and listening to the narratives of others is a unique compassion-building benefit of Death Cafes (Mitchell et al., 2021).

According to Mroz et al. (2020), narrating adverse life situations within a supportive community is associated with increased subjective well-being and resilience. Following a loss, facing the experience may imply both positive and negative reframing, and the ability to integrate these experiences into one's life story will depend on how they are remembered (Neimeyer, 2001; Mroz et al., 2020). Individuals often retrospectively reconcile the stress of loss by retelling events that emphasize personal progress (Mroz and Bluck, 2018) and communion with others. We argue that participating in Death Cafes allows the sharing of life experiences connected with dying and grief, that can contribute to this increased sense of well-being. Narratives about loss tend to include more references to personal connections than those about other life issues, particularly fond memories and recollections of intimacy with a dying or deceased loved one (Bluck et al., 2008). Furthermore, recalling personal interactions with those involved throughout the process of loss can alleviate feelings of isolation (Mroz and Bluck, 2018, 2019).

The abrupt and unexpected death of a loved one can be extremely difficult for the bereaved, and this context of death is relevant given the present pandemic scenario (Gesi et al., 2020; Morris et al., 2020). Previous research has indicated that dealing with fatalities that are discordant with the natural life cycle, such as those caused by an accident or a sudden illness, is severely challenging (Shear, 2012; Keyes et al., 2014). The unexpected death of a loved one may worsen the sense of meaninglessness that bereavement can bring and heighten existential anxiety (Tang and Xiang, 2021). Due to isolation and social distancing during the pandemic, patients dying from COVID-19 had limited physical contact at their bedside and restricted emotional comfort and consolation at end-of-life, while the bereaved endured a lack of access to, or absence of, conventional culturally-acceptable rites and social or community resources (Cardoso et al., 2020; Laranjeira and Querido, 2021; Petry et al., 2021). These restrictions affected the dying person's care options, and fostered feelings of regret among family members, who missed the opportunity to "be there" in those final moments (Breen, 2020; Wallace et al., 2020; Laranjeira et al., 2022). By emphasizing personal sharing in an informal social environment, Death Cafes offer a space for people to talk about death and mortality and process the unprecedented challenges encountered during COVID-19. Recently, there has been a surge of interest in narrative strategies

TABLE 1 Strategies to promote grief literate societies and compassionate communities (Bartone et al., 2019; Breen et al., 2022; Hasson et al., 2022).

- Solidarity actions, such as illuminating landmarks during significant cultural or religious occasions.
- Dedicated support groups for grieving individuals and families by faith communities and social groups.
- Develop awareness on the nuances of grief among health and social professionals, so they can provide appropriate health education and, if necessary, refer patients to psychosocial care.
- Incorporate psychosocial education about loss, grief, and bereavement into employee wellness programs, providing additional opportunities to recognize and talk about loss.
- Broadcast stories of loss, anguish, and grief in the media, as well as stories of hope, healing, and recovery.
- Increase opportunities for creative exploration of grief, emphasizing an art and health approach to death, dying, bereavement and grief.
- Establish universal teaching and learning about death and grief at all educational levels, from primary school to university and adult education (lifelong learning).
- Consider the creation and implementation of a national, European or International Day of Grieving and Commemoration to acknowledge grief and raise global public awareness of death and grief literacy.
- Give special emphasis to unique contexts of death and grieving, including suicide, overdose, homicide, neonatal loss, miscarriage, stillbirth, etc.
- Create more opportunities for individuals to identify what kinds of support they would want at the end-of-life and during periods of grief. For example, advance healthcare directives can assist in promoting discussions around end-of-life care and bereavement.

for dealing with loss (Rolbiecki et al., 2021). Self-narrative framing around loss connects significant events and data into a series of components that span time and create a personal story (Ratcliffe and Byrne, 2022). Storytelling is what makes these events and facts intelligible; it gives them a function, place, and meaning, establishing the order of events in the past, present, and anticipated future. Death Cafes can offer this meaning-building activity of mutual storytelling and sharing at the community level.

We argue that death and grief literacies can better emerge within a population that engages with the Compassionate Communities concept and with social experiences such as Death Cafes. In the Death Café model, a person can remember and maintain a connection with a deceased loved one and share stories with other participants that can instill a positive reflection upon their own life, i.e., gaining a sense of personal growth, avoiding rumination, and focusing on positive social connection (Mroz et al., 2020). Therefore, Death Cafes are arguably aligned with both death and grief literacy and Compassionate Community efforts.

The pandemic has drawn attention to the need to develop grief literate societies and compassionate communities (Table 1). Although there is no single solution for assisting someone who is grieving, we provided some tips that may allow for mutual understandings and interdependent support both in the bereaved's day-to-day settings, as well as in broader society (Fang and Comery, 2021; Breen et al., 2022).

Final remarks

Community-level interventions are a crucial component of a multifaceted public health strategy regarding end-of-life, particularly given the increasing challenges imposed by the changing demography of death and COVID-19. Promoting death and grief literacy through education, health promotion, and community development strategies is essential to attain the skills and culturally appropriate values for a compassionate community. Achieving improved levels of these literacies for both the general public and health and social care professionals is a process that should be prioritized.

Researchers, health professionals, and social educators must collaborate with communities in the design of death and grief literacy projects. Rather than focusing on individual-level acute grieving, initiatives like Death Cafés can promote community-wide literacy around all facets of death and loss. We suggest a dual approach: developing specialist resources while also investing in community capacity to understand grief, give empathetic care and reduce the stigma of death and bereavement.

Finally, there can be unforeseen consequences of making bereavement care a societal responsibility. Presently existing services in government, religious, and charitable sectors may decide to dispense with grief care, if they view the community as the sole source of such assistance. Ultimately, effective joint procedures and true community partnerships among professionals and the public are vital to developing death and grief literacy for all.

Author contributions

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

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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.

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The impact of the COVID-19 pandemic on perinatal loss among Italian couples: A mixed-method study

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Background: Perinatal bereavement is an event that greatly impacts the emotional, psychological, and psychosocial aspects of those who want to have a child.

Objectives: Since there are few studies on the psychological impact of the COVID-19 pandemic on couples grieving for perinatal loss, this research aimed to survey this experience.

Participants: Between 2020 and 2021, in Italian provinces highly affected by the COVID-19 pandemic, 21 parents participated: 16 mothers (76%; mean age 36.2; SD: 3.1) and 5 fathers (24%; mean age 40.2; SD: 3.4), among which there were 4 couples.

Methods: A mixed-method design was used through self-report questionnaires and in-depth interviews. Accompanied by a sociodemographic form, the following questionnaires were administered: Prolonged Grief-13, the Parental Assessment of Paternal Affectivity (PAPA) (to fathers), the Parental Assessment of Maternal Affectivity (PAMA) (to mothers), the Dyadic Adjustment Scale short version, the Daily Spiritual Experiences Scale, and the Impact of Event Scale-Revised. The texts obtained through the in-depth interviews underwent thematic analysis.

Results: Fifty per cent of participants suffered from Post-Traumatic Stress Disorders (PTSD) symptoms and 20% suffered from relational dyadic stress. Four areas of thematic prevalence emerged: psychological complexity of bereavement, the impact of the COVID-19, disenfranchisement vs. support, and spirituality and contact with the lost child. Participants interpreted their distress as related to inadequate access to healthcare services, and perceiving the pandemic restrictions to be responsible for less support and lower quality of care. Furthermore, they needed psychological help, and most of them were unable to access this service. Spirituality/religiosity did not help, while contact with the fetus and burial did.

Conclusion: It is important to implement psychological services in obstetrics departments to offer adequate support, even in pandemic situations.

KEYWORDS

perinatal grief, COVID-19, pandemic, mixed-method research design, psychological support

Introduction

Perinatal loss, comprised of experiences of miscarriage, stillbirth, and neonatal death, is a highly traumatic experience characterized by stressful conditions aggravated by the psychological dissolution of expectations regarding raising a child and developing a parental role (Obst et al., 2020; Testoni et al., 2020). Experiences of loss are similar at every stage of gestation up to the first month of the child's life (Callister, 2006) and have a deeply negative impact on mothers' psychological and physical wellbeing (Kersting and Wagner, 2012). Typical feelings are sadness and despair, anxiety, guilt, anger, and longing for the child, which are accompanied by physiological changes, such as sleep disturbances and lack of appetite, and psychiatric symptoms, including complicated grief (Kersting et al., 2011). Guilt mainly occurs, especially in the presence of ambivalent feelings toward the pregnancy (Kersting and Wagner, 2022), the conviction of having made mistakes, and the perception of failure of the body (Jones, 2014). This condition may persist for several months or even years (DeMontigny et al., 2017), and the couple's relationship is often undermined because of feelings of guilt, mutual attribution of responsibility, resentment, and the perceived failure of the parental project (Kersting and Wagner, 2012; Cena and Stefana, 2020).

The suffering of loss may be exacerbated and prolonged due to lack of social or partner support (Lasker and Toedter, 2000; Burden et al., 2016), an unsatisfactory relationship with healthcare staff (DeMontigny et al., 2017), lack of funerals or other rituals (Kersting and Wagner, 2012), and perceived social delegitimization of grief (Capitulo, 2005). On the contrary, protective factors are social support (Toedter et al., 2001), membership in religious communities (McIntosh et al., 1993), having children, and a satisfactory relationship with healthcare professionals (DeMontigny et al., 2017; Cena et al., 2021a). The literature on the effects of COVID-19 on maternity is contradictory, as is the information from the media. It seemed that pregnant women affected by infectious diseases did not appear to report more severe symptoms than non-pregnant women and, in most cases, were asymptomatic or mildly symptomatic (Delahoy et al., 2020). However, studies have also shown that adverse outcomes following maternal COVID-19 infection were infrequent (Delahoy et al., 2020), and others have evidenced that pregnant women were more likely to be intubated and develop composite morbidity (DeBolt et al., 2021; Kotlar et al., 2021) or die (Zambrano et al., 2020). The risk of COVID-19 transmission from mother to fetus is very low (Egloff et al., 2020); however, some studies point to an increase in premature births (Blitz et al., 2020; Lokken et al., 2020; Allotey et al., 2021), low birth weight, cesarean sections (Knight et al., 2020; Savasi et al., 2020), and maternal and neonatal mortality, and extrauterine pregnancies compared with the pre-pandemic period (Khalil et al., 2020; Chmielewska et al., 2021). As infodemia had a negative effect on pregnant women (Ennab et al., 2022), it is possible to hypothesize that some concerns inherent in COVID-19 could have impacted pregnant women and their partners who lost their babies. Since the literature is still scarce on this issue, this study wanted to consider this particular group of persons.

The research

Objectives and participants

The aims of the research were to investigate the experiences related to the impact of the COVID-19 pandemic on mothers and fathers' grief who experienced a perinatal loss between 2020 and 2021. Changes in social and couple relationships, maternal/paternal affectivity and satisfaction, trauma, grieving strategies, and un/helpful factors were detected.

They were recruited at healthcare centers throughout Italy (counseling centers, hospitals, etc.) by healthcare professionals (psychologists, psychotherapists, psychiatrists, midwives etc.), collaborating with the Observatory of Perinatal Clinical Psychology at University of Brescia.

Psychotherapists collaborating with family counseling centers identified potential participants and indicated them to the researchers of this study. The inclusion criteria were as follows: the loss occurred from March 2020 to March 2021; being able to speak the Italian language. Mental health was the main exclusion criterion: participants were not undergoing psychiatric or psychopharmacological treatment. Recruitment of participants ended when the topics brought in by the research participants became repetitive and the data reached theoretical saturation (Morse, 2015).

Twenty-one parents participated: 16 mothers (76%; mean age 36.2; SD: 3.1) and 5 fathers (24%; mean age 40.2; SD: 3.4). Among them, 86% had Italian nationality; 62% had a university degree, 33% had a high school diploma and 5% had a middle school license; 95% is employed; 52.4% had a medium-high economic condition, 33.4% a modest one and 14.2% some economic problems; 95% are married or cohabiting. Healthcare professionals and participants (mothers and fathers) took part in the study voluntarily and without compensation. The recruited subjects were given an Information Note with a description of the aims of the study and those who agreed to participate were asked to sign an Informed Consent Form. To protect their privacy, a code was assigned to participants, with which they became part of the study (they could authorize the communication of their names to the research centers). The research followed APA Ethical Principles of Psychologists and Code of Conduct. Participants were explained in detail all the objectives of the research and the methodology of analysis used. The study was approved by the Ethics Committee for Experimentation of the University of Padua (n. 53FB052AA456203CE7F4E9C76EBAFAEE).

Methods and instruments

A mixed-methods design was adopted through the use of a self-report protocol and the implementation of an in-depth interview (Cena et al., 2022).

The quantitative protocol

A sociodemographic form was used to collect age, nationality, education, profession, marital status, pregnancy data (gestational week, referral facility, number of pregnancies, any previous abortions), and perceived level of support from family, friends, and healthcare services.

Prolonged Grief-13 (PG-13) (Prigerson et al., 2008) comprised of 13 items (2 dichotomous, 11 assessed on a 5-point Likert scale) was used to investigate the presence of prolonged grief symptoms. The result is calculated on the basis of five criteria: the event of loss; separation anxiety; the duration criterion; cognitive, emotional, and behavioral symptoms; and significant functional impairment after 6 months. A prolonged grief diagnosis is given when all five criteria are met. However, the instrument can also serve as a continuous measure by summing up the symptom items and excluding the two items concerning duration and impairment of functioning (Prigerson et al., 2008).

The analysis of internal consistency confirms that the single factor structure is highly satisfactory, with Cronbach's α coefficient equal to 0.93. Therefore, the extraction of the only factor and good reliability analysis leads to the conclusion that the PG-13 can be considered a valuable instrument for the evaluation of the PGD in clinical practice (De Luca et al., 2015). In current study Cronbach's alpha of PG-13 is 0.83 for female participants and 0.68 for male participants.

The Parental Assessment of Paternal Affectivity (PAPA) (Baldoni and Giannotti, 2020) uses a 10-point Likert scale to investigate paternal affectivity and the corresponding version for mothers. *The Parental Assessment of Maternal Affectivity* (PAMA) for mothers consisted of 10 items that investigated 8 dimensions: anxiety, depression, perceived stress, irritability/rage, relational problems (in a couple, with friends and at work), alterations in illness behavior (somatizations, functional medical disorders, hypochondriac complaints), physiological disorders (sleep, appetite, or sexual desire-related disorders), addiction disorders, and behavioral acts. The instrument thus makes it possible to identify fathers/mothers' perinatal affective disorders, respectively.

Preliminary findings concerning the prenatal period showed significant association between PAPA total scores ($P = 0.05$) and single scale scores with many scores on CES-D, SCL-90-R, ASA, PSS, and DAS. Preliminary data of this Italian validation study confirm the PAPA as a useful tool for the screening of paternal affective disorders in the perinatal period (Baldoni et al., 2018). In the study described here, Cronbach's alpha is 0.68 for PAMA and 0.66 for PAPA.

The Dyadic Adjustment Scale short version (DAS-4) (Sabourin et al., 2005b) was used to assess couple satisfaction. It consists of four items, three of which are on a 6-point Likert scale ranging from 0 (all the time) to 5 (never), while the final item is on a 7-point scale, ranging from 0 (extremely unhappy) to 6 (perfect).

The DAS-4 proved to be informative at all levels of couple satisfaction. Compared with the 32-item version of the DAS (DAS-32), it was as effective in predicting couple dissolution and was significantly less contaminated by socially desirable responding (Sabourin et al., 2005a). In the present research, the Cronbach's alpha of the DAS-4 corresponds to 0.61 for female participants and 0.62 for male participants.

The Daily Spiritual Experiences Scale (DSES) (Currier et al., 2012) (16 items on a 6-point Likert scale) to examine an individual's perception of transcendence and their interaction with it in everyday life.

The DSES evidenced good reliability across several studies with internal consistency estimates in the 0.90 s (Underwood and Teresi, 2002). In this study, the Cronbach's alpha of the DSES for women is 0.93, while it is 0.90 for men.

The Impact of Event Scale-Revised (IES-R) (Weiss and Marmar, 1997) (22 items on a 5-point Likert scale) was used to measure the subjective response to the pandemic impact, assessing the presence and extent of post-traumatic stress disorder symptoms: intrusiveness, avoidance, and hyperarousal. The IES-R is a revised version of the IES and was developed because the original version did not include a hyper-arousal subscale. Both versions have shown good psychometric properties. Test–retest reliability (r = 0.89–0.94) and internal consistency (Cronbach's α) for each subscale (intrusion = 0.87–0.94, avoidance = 0.84–0.97, hyper-arousal = 0.79–0.91) are acceptable. Correlations have been found to be high between those of the IES-R and the original IES for the intrusion (r = 0.86) and avoidance (r = 0.66) subscales, which supports the concurrent validity of both measures (Craparo et al., 2013). The Cronbach's alpha of the IES-R in this study is 0.88 for female participants and 0.81 for male participants.

Self-report questionnaires were uploaded on an online platform. Data analysis was carried out using SPSS software.

Qualitative analysis

The data collection was carried out through in-depth interviews, which allowed the researchers to understand the participants' points of view on the investigated topic and draw on narratives about their experiences, attitudes, and perceptions. The interviews, lasting about 60 min each, were conducted *via* the Internet, recorded, and transcribed. The semi-structured interview investigated the following issues: history of pregnancy, communication of the loss, experiences related to grief, rituals, changes in the relationship with the partner, support received, the psychological impact of COVID-19, and the role of spirituality in the management of grief.

As required by the thematic analysis (Braun and Clarke, 2012), three phases were conducted. First phase: repeated listening to the audio recordings of the individual interviews and reading of their transcription to retrieve information about participants' experiences, leading to the recognition of the main themes of their narratives. Second phase: interpretation of the meanings of the main themes and their links with the whole discourse and recognition of the perspectives of each individual participant, as well as the similarities and differences between them. Third phase: definition of the thematic dimensions (Testoni et al., 2019).

Thematic analysis is very flexible and is suitable for studying participants' experiences and perceptions of a given phenomenon (Braun and Clarke, 2012) through an inductive process of coding and creating themes from the data without referring to any previous theory (Boyatzis, 1998).

Analyses were performed by a trained researcher under the supervision of an experienced qualitative analysis researcher. This made it possible to have two coders (inter-rater reliability) who clearly defined coding rules in advance, thus enabling the creation of a shared analysis codebook. There was total agreement between the two researchers. After they reached agreement on the interpretation, another researcher discussed the procedure and the results obtained. Finally, after modifications were agreed upon by the three researchers, the final structure of the report was defined.

The paper-and-pencil analysis operations were then integrated using the computer program qualitative analysis software Atlas.ti (Muhr, 1991) which has been precisely designed to aid researchers in qualitative data interpretation, allowing an analysis that is as objective as possible.

Results

Quantitative analysis

With respect to participants, there were four couples: Isabella–Antonio, Cristina–Ernesto, Laura–Giulio, and Valeria–Pietro. Five participants did not attend the interview (Barbara, Claudia, Elisa, Silvia, and Teresa).

Results for female participants

With respect to the sociodemographic variables, the average gestation period was about 19 weeks (M = 18.7, SD = 12.4, Range = 2–42), the average number of previous pregnancies is one (M = 1.4, SD = 1.5, Range = 0–5), 43% had previous experience of miscarriage and for almost half of participants it was the first child. One-third of the group did not perceive sufficient support from family members, their network of friends, and especially the healthcare service. Half of the mothers stated that they received good support from their partners, while half defined it as absent or scarce.

IES-R: Seven participants (44%) present symptoms usually similar to post-traumatic stress disorder.

PG-13: All participants met criterion A, as all had experienced bereavement; 31.3% met criterion B, relating to separation distress, while criterion C, relating to the duration of bereavement symptoms, was met by 43.8%. Criterion D, relating to the presence of cognitive, emotional, and behavioral symptoms, was met by only 6.3%, and criterion E, relating to significant functional impairment 6 months after loss, was met by 31.3% of participants. However, none of the participants met all five criteria necessary for establishing the diagnosis, so none of the mothers showed complicated grief.

The PAMA: Those who had a score above 40 (Barbara = 40, Elisa = 56, Flavia = 60, Laura = 50) were at risk of an affective parental disorder.

The DSES: 50% of the participants had a high perception of transcendence, 19% had a slight perception, and the remaining 31% had little or no perception.

The DAS-4: 3 participants (19%) had a score below 13 and suffered from couple dissatisfaction.

Results for male participants

The sociodemographic form showed that, for one, this was the first experience of pregnancy. Regarding the analysis of social support, for four of the fathers, the support received from family was good; in relation to the friendship network, three described it as good, while two described it as sufficient. Concerning the health services, two felt that the support was absent, one described it as poor, one as sufficient, and one as very good. Four noted the lack of psychologists and assistants. Four fathers pointed out that they could not access hospitals or have contact with doctors and paramedics. The lack of support was blamed on the COVID-19 restrictions.

IES-R: Three (60%) present symptoms usually similar to post-traumatic stress disorder.

PG-13: All participants met criterion A because they had suffered from grief. None met criterion B relating to separation anxiety. Criterion C, the duration of symptoms related to grief, was met by one. None met criterion D, relating to the presence of cognitive, emotional, and behavioral symptoms. None met criterion E, relating to a significant functional impairment 6 months after the loss. Thus, none of the participants fulfilled all five criteria necessary for the diagnosis, and none of the fathers showed a significant picture of prolonged bereavement.

PAPA: All participants scored below 40; thus, none of the participants were at risk of developing an affective parental disorder.

DSES: Two of the participants reported a slight perception of transcendence, while another two described it as moderate; one reported a high perception.

TABLE 1 Correlations of distress measures with maternal/paternal affectivity, couple satisfaction, spirituality and with socio-demographic, pregnancy, and perceived support variables ($N = 21$).

	Distress measures				
	Avoidance	**Intrusiveness**	**Hyperarousal**	**IES-R total**	**PG-13 total**
Distress measures					
Avoidance	−−				
Intrusiveness	0.45*	−−			
Hyperarousal	0.46*	0.55**	−−		
IES-R_Total	0.77***	0.87***	0.79***	−−	
PG-13 Total	0.52*	0.57**	0.55**	0.68***	−−
Maternal/Paternal affectivity					
PAMA/PAPA Total	0.38	0.44*	0.04	0.38	0.55**
Couple satisfaction					
DAS-4 total	−0.46*	−0.43	−0.31	−0.50*	−0.43
Spirituality					
DSES total	0.64**	0.08	0.24	0.37	0.39
Socio-demographic variables					
Gender (Female = 1; Male = 0)	0.08	0.01	0.02	0.04	0.29
Age	−0.21	−0.06	−0.09	−0.14	−0.11
Education (university degree = 1; lower level = 0)	−0.54*	−0.04	−0.21	−0.30	−0.13
Economic condition (medium-high level = 1; lower level = 0)	−0.44*	−0.01	−0.31	−0.28	−0.28
Previous children (Yes = 1; No = 0)	0.36	0.29	0.53*	0.46*	0.39
Pregnancy variables					
Gestational week	0.17	0.28	0.71***	0.45*	0.43
Number of pregnancies	0.11	0.04	0.40	0.19	0.40
Previous miscarriages/stillbirth (Yes = 1; No = 0)	−0.25	0.11	0.29	0.05	0.24
Precived support variables					
Support from family	−0.53*	−0.24	−0.26	−0.41	−0.31
Support from friends	−0.44*	−0.08	−0.41	−0.35	−0.40
Support from health services	−0.29	−0.04	−0.05	−0.15	0.10

$^{*}p < 0.05$; $^{**}p < 0.01$; $^{***}p < 0.001$.

DAS-4: No participants scored below the clinical cut-off, indicating the absence of clinical distress related to their relationship with their partner.

Results of correlations

Correlation analysis between all distress measures and between distress measures and other variables—maternal/paternal affectivity, couple satisfaction, spirituality, socio-demographic variables, pregnancy variables, and perceived support variables -, were performed using the Pearson r coefficient. All distress measures are significantly correlated: the PG-13-total show high positive correlation with the subscales of the IES-R avoidance (r = 0.52), intrusiveness (r = 0.57), hyperarousal (r = 0.55), and with the total (r = 0.68). The PAMA/PAPA total correlates positively with the PG-13 total score (r = 0.55) and IES-R intrusiveness (r = 0.44). The DAS-4 total correlates negatively with IES-R total (r = −0.50) and IES-R avoidance (r = −0.46). The DSES total positively correlates with IES-R avoidance (rho = 0.64). Considering socio-demographic variables, high education and high level of economic conditions correlate negatively with IES-R avoidance (r = −0.54 and r = −0.44, respectively) while the presence of previous children correlates positively with IES-R hyperarousal (r = 0.53) and with IES-R total (r = 0.46). Furthermore, considering the pregnancy data, gestational week correlates positively with IES-R hyperarousal (r = 0.71) and with IES-R total (r = 0.45). Finally, considering perceived support, high support from family and high support from friends correlate negatively with IES-R avoidance (r = −0.53 and r = −0.44, respectively). Spirituality was significantly correlated only to IES-R Avoidance (r = 0.64) (**Table 1**).

Results of participant's gender differences

Gender differences on all variables—distress measures, maternal/paternal affectivity, couple satisfaction, spirituality, socio-demographic variables, pregnancy variables, and perceived support variables—were analyzed using the test t for independent samples. No gender differences were found except for age of participants (t = 2.44, df = 19, p = 0.012) and support of health services (t = −2.31, df = 19, p = 0.016). Male participants are older than female participants (M = 40.2, SD = 3.4 and M = 36.3, SD = 3.1, respectively), while male participants perceived less support of health services than female participants (M = 2.4, SD = 1.7 and M = 3.9, SD = 1.2, respectively).

Gender differences on correlations were analyzed by calculating the r-to-z transformation to compare correlations across men and women. No gender differences were found except for education with IES-R avoidance correlation (z = −2.53, p = 0.006) and previous children with PG-13 total (z = 2.45, p = 0.007). Male participants showed highest negative correlation between education and IES-R avoidance than female participants (r = −0.98 and r = −0.36, respectively), while they showed highest positive correlation between previous children with PG-13 total (r = 0.98 and r = 0.41, respectively).

Qualitative analysis

Four areas of thematic prevalence emerged from the analysis of the interviews:

- Psychological Complexity of Bereavement: where the main grief-related experiences described by the participants were reported.
- The Impact of COVID-19: the thematic area refers to the difficulties experienced by parents related to the concomitance of the Covid-19 pandemic with loss.
- Disenfranchisement vs. Support: refers to the presence or absence of external support.

Relationship with Spirituality and contact with the lost child: the area concerns the role played by spirituality/religion in the elaboration of grief.

All the names cited are pseudonyms. In the brackets, only the significant results are reported the first time each name is quoted (**Table 2**).

Psychological complexity of bereavement

The first theme refers to grieving. The most frequent feelings were sadness, fear, anger, envy, guilt, and the sudden dissolution of expectations about the imminent construction of a family. This area was composed of two subareas.

The first subarea was inherent to the grieving, which involved both mothers and fathers in different ways. The body was the first dimension denouncing the loss. Sofia (IES-R = 34; DSES = 33) stated: "I have hallucinated pains, and this and this prevents me from returning to normal. My body and I are still quarreling." The sense of an empty and arid womb was particularly painful, as described by Roberta (DSES = 40): "After the curettage, what you feel is a sense of emptiness, because you perceive that your body was changing, your abdomen swelled, and then a great sense of emptiness overwhelms you." According to her, "My husband was worried, first of all, whether I would recover, and that's why he didn't want to grieve, hoping to support me." Marianna said: "The body can resist physical pain, what is unbearable is the internal suffering. Something breaks inside me." Valeria (IES-R = 49; DSES = 31), who also had

TABLE 2 Thematic areas and sub-areas emerging from the qualitative analysis.

	First theme	Second theme	Third theme	Fourth theme
	Psychological complexity of bereavement	**The impact of COVID-19**	**Disenfranchisement vs. support**	**Relationship with spirituality and contact with the lost child**
First subarea	Bereavement-related experiences	The loneliness experienced by both partners due to restrictions	Disenfranchisement	Role of spirituality/religion in grieving
Second subarea	Confusion related to one's parental identity	The difficulty in accessing services	Social support	Contact with the fetus and funeral rites

a previous miscarriage/stillbirth, and the new baby was born dead after 9 months due to dystocia, narrated: "I was petrified, I couldn't say anything, think about anything, or cry. Even today, at times, I deny what happened." She tried to understand her partner Pietro": "He's a racer, so he's raced a lot, worked a lot, outlined technical projects. I understood that this was his way to face the loss." The participants also suffered from intense anger. Flavia (IES-R = 53; PAMA = 60; DAS-4 = 10), who had experienced a previous miscarriage, reported: "I'm absolutely angry because, very nicely, the gynecologist told me, 'Well madam what do you want me to tell you, there is no longer the baby's heartbeat."' Her relationship with her husband went into crisis: "Because of constant guilt and anxiety. In the end, I had to give him an ultimatum." Miriana stated, "I am angry. If they had realized before that the fibroma was already big, what would have happened?" And Flavia said: "I am angry with karma and with God. Two very dear friends of mine found out they were pregnant, but they did not accept it. Seeing them not happy and me instead who would have strongly wanted it disturbs me profoundly." Pietro, Valeria's husband, similarly said: "I had never felt an emotion like this, I think the right name is anger; it was explosive at the beginning, I felt this energy, I was not willing to tolerate this loss'." The common emotion between mothers and fathers was a sense of guilt. Additionally, fathers showed an intense feeling of guilt, as in the case of Ernesto (IES-R = 41; DSES = 29), who had to make a difficult choice when, with his wife Cristina, he discovered that the fetus had a severe malformation at the fifth month: "It was a terrible choice that I didn't expect to make. I still live with this choice in my mind. I have regret, and the doubt persists. Did we get it right, or did we get it wrong?" Fathers also felt intense existential suffering, as in the case of Antonio (IES-R = 41), who affirmed that "I always ask me what I have lived, what happened, what it could have been. Now there is a feeling of sadness, of emptiness, of a physical lack, because I wanted another child." However, the most important feeling was the sense of responsibility and a need to be useful to recovering normalcy, as affirmed by Alberto (IES-R = 41): "She prefers to talk about it to get over that moment, while my personal protection is to deflect the subject. [...] I also had to take care of our son, and it wasn't easy to explain to him what had happened"; and Pietro "My reaction is to do, to act, my wife on the other hand needs time, to stay calm, to put her thoughts in order, to try to accept this loss."

The second sub-area was inherent to confusion related to one's parental identity, especially among mothers who have had previous miscarriages or stillbirth. The participants were confused about their role, they did not know whether they should consider themselves mothers or not. Sofia, who had already previously had two similar experiences, said, "I wasn't able to manage pregnancy. Ambivalence characterized my entire pregnancy; I was pregnant and I drank a glass of wine, I was pregnant and I started to smoke again, I was pregnant and I took a drug to sleep, always with the thought that I was doing damage, that I was not an adequate mother. Moreover, I didn't want to see the baby during expulsion, and I said, 'what kind of mother are you? Why don't you want to see your child?' So finally, I chose to see him." She also said, "It is not the events themselves that upset me the most, but seeing becoming a mother fade away, which is becoming more and more impossible by now; all this gives strong frustration." Similarly, Marianna, who had a previous miscarriage, affirmed, "I want a child, so I asked myself what the meaning of my life was. I thought there wasn't one; I wasn't working and, above all, I wasn't a mother." Gioia also had the same feeling: "For a while, I feel like a mother who has a child. On these days, I would wonder, if 1 day I will have children and they ask me how many children you have had, will I have the courage to count the miscarried ones? I'm really confused." Cristina (DSES = 47) said: "I had the perception that I was not a mother because I had abandoned my child, both after the birth and after the burial."

The impact of COVID-19

The second thematic area was related to the concomitance of pandemic and miscarriage and stillbirth. It was divided into two subareas: the loneliness experienced by both partners due to restrictions and the difficulty in accessing services.

The first subarea considered the experiences of mothers and fathers, who could not be near their partner during visits, thus fueling a sense of loneliness and exclusion. The couple also did not have the opportunity to confront each other when making important decisions as the restrictions did not allow fathers to attend the various visits. Flavia, who had problems related to the infections, tells: "I was terrified of being pregnant, alternating with happiness at the news after the COVID nightmare, but given everything that was happening, I was terrified that something might happen." The other participants reported that the main consequence of the pandemic was that they had to make their visits alone and be hospitalized and operated on in the absence of their partner or parents, as well as having to make important choices alone, as in the case of Antonio's wife Isabella, who said, "The phone, the video call maybe, were not enough, and maybe the most fundamental part for me was the emergency room, when you really had to realize what was happening, and my husband was out there. It made me so sad." Fathers also tell, who, on the other hand, suffered from an inability to attend the visits and participate in their partners' hospitalizations, exacerbating their sense of helplessness, as described by Antonio: "I was just there waiting for my wife for 48 h, I did not have the ability to access the hospital. My biggest regret is not having seen my son and not having been with my wife in that terrible moment."

The pandemic restriction also exacerbated the distress, as explained by Cristina: "It was difficult to go for a walk, it seemed to me that the house was closing in on me, as if the walls were collapsing and the pain was becoming even more vivid," and by Roberta: "Without meeting people, I had too much time to think, and this had broken me down. I felt that my head would explode because I was suffocating."

A huge sense of protection toward the partner derived from this situation, as described by Giulio (DSES = 40) with respect to his wife Laura: "Mainly I was worried, not so much for the loss of the child, but for what was happening to my wife. I was worried about her physical and psychological conditions," and by Antonio: "A huge feeling of protection toward my wife took over, because I wanted her to feel well since the beginning, regardless of what could have happened with the pregnancy."

The second subarea considered the difficulty in accessing services and medical examinations. Moreover, it was very difficult to access the necessary controls, as described by Marianna: "My gynecologist told me that she couldn't receive any more patients for examinations. Trying to call the hospital, I received only negative answers. Everything was blocked; the answer I was given was to go to a private professional, and this upset me further."

Furthermore, as stated by Ernesto: "There was a lack of concrete support: finding the doctors, going to the hospital. The exams were continuously postponed, extensive waiting times, calling and not finding anybody," and by Alberto: "My wife was not assisted at all, and the operators did not even let me stay in the waiting room."

Disenfranchisement vs. support

This area is divided into two subareas: disenfranchisement and social support.

The first subarea concerns disenfranchisement, which caused great suffering. Participants did not always feel their grief and bereavement was acknowledged from the outside and sometimes received superficial or devaluing comments about the experience from acquaintances, friends and family members and even from their partners, a factor that exacerbated the pain of the moment. Marianna perceived her partner as absolutely absent and detached: "He didn't suffer as much as I did, and he didn't talk about it, because it was an uncomfortable subject. For him, this is not a problem." In Flavia's opinion, "People don't give importance to this experience and think that I already have a daughter and so I must be satisfied! For this reason, I prefer to avoid people who know me." Similarly, Marianna did not want to tell anyone about this loss: "I decided not to talk to anyone about my condition for fear of not being understood. However, I needed someone to ask me if I needed help." According to Ernesto, "You can only talk about it if someone shows interest. This wound is very painful and the others don't understand it, so they can't help me." Similarly, Pietro said, "Some people feel the need to ask questions, disrespecting you. Their language goes faster than their thoughts because they are curious. There is a terrible level of emotional ignorance and lack of empathy." The medical staff also proved unable to recognize the mothers' suffering, as Cristina continued: "The doctors don't recognize your pain. They are cold, detached, even when they give you the bad news."

The second subarea concerned the importance of support. If, on the one hand, participants did not always feel understood by others, receiving comments from outside aimed at minimizing the loss, on the other hand, they always found figures within their own circle of friends, family or care team who supported them. For many of them, social support was an important protective factor in the grieving process, as described by Isabella: "I was lucky because the nurse was very close to me and allowed me to go out to see my husband," and Miriana: "I met a fantastic team. Everyone tried as much as possible to overcome the limitations of the pandemic to make me feel more comfortable. Their sense of humanity prevailed." Those who were supported by mutual-aid groups had a great advantage, as Valeria emphasized: "Participating in these groups allowed us to find out that many other couples had the same experience, and we found great support in these people especially, because they understand."

Most fathers and mothers, however, considered the support offered by family and friends as adequate and comforting.

Relationship with spirituality and contact with the lost child

Religiosity/spirituality was not particularly effective in supporting the grief experience. Sofia (IES-R = 34; DSES = 33) stated, "I don't have a religion to take refuge in. I think things are the way they are, and I deal with them the way they are. I consider myself unlucky and do not consider any existence of this baby." In the same vein, the fathers also have a low level of spirituality. According to Giulio (DSES = 40), "'It had to be like this,' it is a scenario that is contemplated when you think about having a child, it is a natural thing that can happen." Similarly, Ernesto (IES-R = 41; DSES = 29): "I don't find support in religion and I don't feel like a particularly unfortunate case and I'm not desperate. I feel very lucid because I believe that this is a human thing, that it can happen." Both lower and high levels of spirituality are accompanied by a positive experience of contact with the fetus and the funeral ritual, as in the cases of Cristina (DSES = 47): "I'm so pleased, I wanted to see it because now it's not nebulous; I have a concrete face to remember, which at the beginning occupied an immense space in my mind and is now slowly getting smaller, taking the right proportions," and Valeria (IES-R = 49; DSES = 31): "I needed to touch, to see, to give a face, to give a weight, even holding her in my arms helped me. I would have lacked contact with reality if I had not seen her. [...] In this way I closed the circle," as well as Miriana: "Being able to touch and hold him allowed me to understand what happened," and Flavia (DSES = 51), who also treasured the funeral: "In this way I was able to say goodbye to him, as we do to those who have lived with us."

For the fathers, it was easier not to celebrate any funeral, as in the case of Ernesto (DSES = 29): "The funeral makes sense if you can remember who died, if you do it for someone you lived with. I knew he was going to be stillborn because we had chosen to have him stillborn because of his severe malformation, so it was better not to proceed further," and Alberto (DSES = 58): "I suffered a lot for this loss. I preferred not to have any funeral because if I had to think about the burial, it would have been much harder."

Discussion

This study adopted a mixed-method research design. The quantitative analysis exhibited that a large number of participants presented most of the symptoms attributable to post-traumatic distress, moreover, several participants showed low levels of couple satisfaction and disturbances in the affective parental sphere. Correlational analysis revealed significant relationships between the presence of symptoms of post-traumatic stress disorder and the dimensions of hyperarousal and intrusiveness. Although all participants are suffering intently from the loss, none could be diagnosed as having complicated grief, and the total PG-13 score correlated with all the dimensions of the IES-R, showing that a higher level of distress corresponded to a greater difficulty in processing the loss. These results are in line with the literature emphasizing that miscarriage is an extremely traumatic experience (Farren et al., 2016). The results indicate that greater difficulties in grieving are positively correlated with emotional difficulties in the parenting role, having had children previously, and when gestational age was longer. With respect to gestational age, the results are in line with Cuisinier et al. (1993) but in contrast with some studies that underline that the suffering is the same in all the phases of pregnancy (Callister, 2006; Kersting and Wagner, 2012). This issue is less studied, and the limited results are inconsistent, so it would be useful to conduct further research.

From the qualitative analysis, four areas of thematic prevalence emerged, with some subareas. Psychological complexity of bereavement was the most important because participants preferred to describe their psychological experience. It was composed of two subareas: the grieving experience and the couple relationship. The second area, the impact of the COVID-19, was composed of two subareas: the experiences of mothers and of fathers. The third, disenfranchisement vs. support, considered the two experiences in two different subareas. Finally, spirituality is positively correlated with avoidance, and this result indicates the ambivalent role of this dimension, that did not seem to help parents in grieving. Indeed, this dimension did not signify the contact with the lost children and whether the relationship with transcendence influenced the relationships with them. With regard to grieving, it was confirmed that there were different ways of dealing with loss between mothers and fathers. The women considered it necessary to have time for introspection and reflection, and it seemed to help to have positive conversations. Conversely, the male tendency was to adopt elusive strategies, avoiding talking about it or engaging in other activities that allowed them not to focus on the problem (Obst et al., 2020). Specifically, while women described a great deal of grief, demonstrating the presence of all the stages of mourning described by the Kübler Ross model (Corr, 2019), as already described in the literature (Testoni et al., 2020), men preferred silence and did not share their grief with their partners (Miller et al., 2019), probably due to culturally established gender roles and stereotypes (Rinehart and Kiselica, 2010), which also limited them in seeking support in family and friendship networks (Saunders and Peerson, 2009). This imbalance may partly explain why the DAS-4 results reported that several participants suffered from relationship problems.

The second thematic area was divided into the negative impact of COVID-19 on both maternal and paternal bereavement experiences because of indirect effects related to the difficulties in accessing healthcare services and follow-up visits, the lack of adequate support from social and healthcare personnel, and the inability of fathers to assist and offer support at the time of their wives' hospitalizations. None of the participants, in fact, felt that Covid-19 had a direct effect on their pregnancy, even in cases where the women were infected. The main consequence of the pandemic was found in the fact that mothers were forced to make their visits alone, as well as being hospitalized and operated on. In fact, all of this took place without the presence of their partners or parents: this context burdened their experience and exacerbated their sense of loneliness and abandonment. Moreover, in some cases, the women found themselves making important decisions alone, without being able to communicate except by telephone. Participants felt, precisely because of the COVID, that they had lost an important piece of their experience, namely sharing with their partner. Finally, they felt very frightened about not being able, in some cases, to undergo check-ups on time.

Finally, the presence of Covid also weighed on the grieving process; the restrictions due to the infection prevented many from going out, distracting themselves, resuming work and having contact with the outside world.

The results are in line with the literature (Burki, 2020; Roberton et al., 2020), which shows that the negative effects of the pandemic on perinatal health are not limited to morbidity or mortality caused directly by the virus, but also by the restrictive measures that undermined social relationships, and thus support, especially inhibiting access to health services. Indeed, the literature has already shown that the pandemic was very stressful for fathers (Baldoni and Giannotti, 2020) and especially for pregnant women, who reported higher levels of anxiety and depression compared to cohorts analyzed in the pre-pandemic period (Dennis et al., 2017; Woody et al., 2017; Cena et al., 2020, 2021b; Yan et al., 2020), partly due to the lack of support from the healthcare system (Thayer and Gildner, 2020). The negative effects of the pandemic on maternal and perinatal health include restrictive measures, disruption of health services, and fear of using these services, which are among the main factors that compromised the physical, psychological, and social wellbeing of participants, similar to those facing pregnancy during the height of the pandemic (Burki, 2020; Roberton et al., 2020). Linked to this is an additional factor: the altered family and social relationships due to estrangement from loved ones and friends (Khalil et al., 2020; Chmielewska et al., 2021). Social support is important, but most participants perceived the disenfranchisement of their grief, on the part of both family and friends, and especially by health professionals (Lang et al., 2011). Perinatal grief was, in fact, misunderstood because if there were already children in the family, people said that they could be enough and, in any case, that there remains the possibility of trying to have more. Unfortunately, these perceptions do not consider that, in the representation of the mother and sometimes also of the father, the fetus is already personified and considered a child. This phenomenon has also been found to be widespread in the literature: according to Lang et al. (2011), many couples report that most friends and relatives are not even aware of important dates, such as the birth of the child or the anniversary of the loss; furthermore, it appears that health professionals often adopt a depersonalizing attitude toward the couple and the child, which conveys a feeling of devaluation toward this type of loss. This lack of support contributes to exacerbating the feelings of isolation and mourning experienced by parents.

The interviews also reveal that men feel they have to silence their pain, as their role is to accept and listen to their partner's suffering. This is in line with the literature according to which men perceive that they cannot share their pain with their partner (Miller et al., 2019); furthermore, the freedom to openly manifest this suffering is also influenced by the lack of recognition of the man's loss, based on social expectations that men should be strong and impassive and have the sole responsibility to act as a support for their partner (Rinehart and Kiselica, 2010; Due et al., 2017). This also seems to be linked to less support-seeking among both the friendship and family network (Saunders and Peerson, 2009).

Despite spirituality/religiosity being widely considered very helpful in managing grief (Park and Halifax, 2011), in this group, this dimension did not result in support. All the mothers who did not participate in the interview (Barbara, Claudia, Elisa, Silvia, Teresa) and those who obtained higher scores in PAMA (Barbara, Elisa, Flavia, Laura) were characterized by the highest spirituality in parallel to high levels of posttraumatic symptoms and low couple satisfaction. This suggests that not only is the couple's relationship important but also that there may be a spiritual conflict with respect to the loss of the child (cf. Testoni et al., 2021) and that all this could have made it very difficult to talk about the experience. The inauspicious and often traumatic outcome of pregnancy has often led to a break, albeit temporary, with one's spiritual dimension, especially among those who have always entrusted their lives and destinies to the divine plan. On the other hand, it was highlighted by participants in the interviews that neither the religious nor spiritual dimension informed the desire to see or touch the miscarried or stillborn fetus or mourn it through a funeral rite. Indeed, the literature shows that religiosity helps to overcome distress through the social support of the religious community, where grievers can share languages and symbolism to process the sense of loss (McIntosh et al., 1993; Arshad and Hafeez, 2016). This is in line with the study by McIntosh et al. (1993), according to which religious communities can be a supportive resource, just as greater religious participation seems to be correlated with a greater perception of social support, helping to decrease distress related to parental bereavement. It is possible that the restrictions imposed by the pandemic reduced the positive effect of this aspect.

Conclusion

The results of the study show how the pandemic negatively impacted couples' experience of miscarriage due to indirect consequences related to difficulties in the healthcare environment and restrictions imposed to prevent contagion. The restrictions exacerbated the negative effects of the trauma by making relationships with healthcare professionals more difficult and limiting contact with friends and family. In addition, decreased contact with participants' religious communities diminished the power of that social support. The need for support was not compensated for by adequate psychological services either. It is imperative to equip gynecology and obstetrics departments with at least one staff psychologist because this professional could have provided the necessary help, even in the most difficult moments of the pandemic.

The most important limitation of this study is the fact that the majority of fathers did not want to participate, so it is possible that the results obtained by those who did participate are not indicative of a specific male method for dealing with this loss. In addition, The group of participants was small due to the fact that some parents who were contacted by the healthcare professionals did not agree to participate in the study because the perinatal loss event was traumatic and painful and they did not want to talk about it again. Therefore, the small number of participants did not allow for more in-depth statistical analysis, and results cannot be generalized. Furthermore, most of the sample is Italian, has a university degree, and medium-high economic conditions. Then, results are not representative of a larger population.

Additional studies could further survey the relationship between grieving and gestational age and with religiosity/spirituality.

The present study has as its future objective the administration of the instruments at a distance of 6 months: it would be useful to carry out a follow-up with the aim of verifying the occurrence of significant changes, also in relation to the evolution of the pandemic context. Furthermore, another future objective is to extend the research to a larger number of participants, who have also suffered perinatal losses beyond the time period considered so far; connected with this objective, extending the number of participants could make it possible to recruit a substantial number of couples, which would allow an analysis not only of individuals, but also between partners.

The participants also pointed out that social support was crucial for them in the process of processing the loss but that in some circumstances the social network was not able to recognize the underlying suffering of the loss m Considering mothers' need to express their grief and to receive strong support, together with the need to mentally and psychologically reorganize the way they represent their deceased child, further research should focus on what professional interventions could be particularly helpful in supporting the grieving process, especially by social workers who can improve social support. Finally, future research should investigate the reactions of siblings and fathers to perinatal loss, since this type of bereavement is still poorly recognized by society, in order to develop a more appropriate support system for them as well.

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 Ethics Committee for Experimentation of the University of Padua. The patients/participants provided their written informed consent to participate in this study.

Author contributions

IT and LC: conceptualization, methodology, writing—review and editing, and supervision. EI and AT: software. IT, LC, AT, GL, LR, and EI: analysis. IT, LC, GL, EI, and AT: investigation. GL: resources. IT, LC, EI, GL, AT, and LR: data curation. IT, LC, GL, EI, AT, NT, and LN: writing—original draft preparation. IT, LC, EI, and AT: project administration. All authors have read and agreed to the published version of the manuscript.

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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.

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