

Review Article

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The role of spirituality in people with amyotrophic lateral sclerosis and their caregivers: Scoping review

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Abstract

Background. There are few studies evaluating the role of spirituality and the role of spiritually integrated interventions in people with amyotrophic lateral sclerosis (PALS) and their caregivers.

Objectives. A scoping review was conducted to examine the nature and breadth of peer-reviewed literature on the role of spirituality, interventions integrating spirituality, and outcomes for PALS and their caregivers.

Methods. A literature review was performed, following the methods from the Joanna Briggs Institute Reviewers, based on all articles published between January 2006 and April 2022, identified in the CINAHL Complete, MEDLINE Complete, MedicLatina, Psychology and Behavioral Sciences Collection, and SPORTDiscus with full-text databases using key terms. Extracted data included research aims, study design, population and characteristics, theme description, and measures or type of intervention.

Results. A total of 18 articles were included in this study: 14 qualitative, 3 quantitative, and 1 protocol of a quantitative study. Eight studies were based in Europe. The search identified different main themes related to spirituality for caregivers and patients, 2 spiritual measure scales, and one intervention. However, many studies were limited in sample size, generalizability, and transferability and used less sophisticated research designs.

Significance of the results. This scoping review illustrates the importance given to spirituality by caregivers and PALS and reveals a very heterogeneous response. Thus, experimental studies in the area of spirituality are needed to systematically explore the impact of spiritual interventions, and the results of these studies could advance practice and policy by enhancing the quality of life for PALS and their caregivers.

Introduction

Amyotrophic lateral sclerosis (ALS), also known as motor neuron disease (MND), is a terminal, heterogeneous inherited or sporadic neurodegenerative disease characterized by the degeneration of both upper (corticospinal) and lower (spinal and bulbar) motor neurons leading to motor and extra-motor symptoms. These symptoms typically spread into different body regions with progressive muscle weakness and loss of voluntary muscle control of the bulbar, limb, thoracic, and abdominal regions, usually leading to death from respiratory failure, on average, 2–5 years after symptom onset (Hardiman et al. 2017). The severity of the illness and uncertainties regarding the time course of disability demands clinical care centered around the patient and carers. Disease-focused expertise offer a multidisciplinary approach that leverages the experience of several health-care providers in order to control the symptoms and assist people with ALS (PALS) to reach their fullest potential, assist their routines and participation, and maximize physical, psychological, and emotional comfort during disease progression (Gonçalves and Magalhães 2022; Hogden et al. 2017; Paganoni et al. 2015). When faced with a terminal illness like ALS/MND, there is a tendency to question the meaning of life and death (Zheng et al. 2021).

Spirituality is a domain of supportive and palliative care in some national policies, and the *European Association for Palliative Care* has published a white paper encouraging multidisciplinary education on spiritual care for palliative care staff in order to provide this support as an integral part of palliative care (Best et al. 2020). PALS and their caregivers may seek out spirituality as a way to cope with disease progression, minimize their suffering, or provide hope.

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In this context, spirituality and religiosity must be distinguished as these terms are typically used as synonyms. Spirituality refers to personal attempts to understand final questions about life and their relation to the sacred and transcendent, including enhancement of meaning and purpose of life, relationship maintenance, provision of comfort, hope, and coping strategies, and a satisfying moral connection beyond the self and its relationship with others and a higher power (Evangelista et al. 2016; O'Brien and Preston 2015; Palmer et al. 2020; Zheng et al. 2021). Religion, in turn, corresponds to an organized system of beliefs, practices, rituals, and symbols aimed at facilitating closeness between individuals and the sacred, with religiosity being the ground level of religion, with which individuals believe, follow, and practice a given religion (Delgado-Guay 2014; Evangelista et al. 2016). Spirituality can lead, or not, to the development of religious practices (Selman et al. 2018; Zheng et al. 2021).

Despite the significance and prevalence of spiritual distress and needs in the context of advanced disease, these needs were often reported as neglected, and spiritual care was lacking in patients facing terminal illness and their caregivers.

Few studies evaluate the role of spirituality and the role of spiritually integrated interventions targeting PALS and their caregivers. Therefore, the value of understanding the nexus of PALS and their caregivers and spirituality is undermined. In order to address this gap, we conducted a scoping review to synthesize research on the intersection of these topics to answer the question: what has been published in peer-reviewed literature about the role of spirituality, interventions targeting programs to address spiritual needs, and their outcomes for PALS and their caregivers?

Methods

A scoping review of scientific literature was performed with the primary objective of mapping scientific knowledge, based on the guidelines of the Joanna Briggs Institute for this type of study (Peters et al. 2020). The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) model (Moher et al. 2009) was used to organize the information, and the recommendations described in PRISMA Extension for Scoping Reviews were followed (Tricco et al. 2018).

Research method

To answer our research question, we selected the scoping review methodology. Peer-reviewed articles published until April 2022 were obtained from CINAHL Complete, MEDLINE Complete, MedicLatina, Psychology and Behavioral Sciences Collection, and SPORTDiscus. To identify relevant articles, search terms were focused on 2 distinct areas. First, keywords relating to "ALS" and "Motor Neuron Disease" were identified to capture the disease in question. Second, terms focusing on the relevant areas of "Spirituality" were identified for the current study. Combinations of descriptors were used, including medical subject headings, subject headings, and subject terms for each database, as well as using free-text terms and the "*" tool, which enhanced the search by including variations of the same word (("amyotrophic lateral sclerosis") OR (ALS) OR ("motor neuron disease*") OR (ALS/MND) OR ("Motor Neuron Disorder*") OR (MND)) AND ((Spirituality) OR (Spiritualism) OR ("spiritual need*") OR ("spiritual care") OR ("spiritual issue*") OR (Existentialism) OR ("Life Purpose") OR ("meaning*") OR ("spiritual value*")).

Inclusion criteria

To be included in this review, an article had to address spirituality in patients with ALS/MND and in their caregivers, namely, in the domain of meaning/purpose of life, throughout the disease course. The search methodology included all types of qualitative, quantitative, and mix-method studies, regardless of their design. Only articles written in English, Spanish, French, and/or Portuguese were selected from articles worldwide.

Exclusion criteria

Publications that did not target PALS or their caregivers' outcomes were excluded, such as health-care professionals providing services to the patients and families. In addition, publications were excluded if they only addressed religiosity or whose focus was not spirituality. Finally, publications that fulfilled the aforementioned criteria but reported studies other than primary research were also excluded, such as editorials, letters, concept papers, review articles, unpublished (gray) literature, dissertations, books, and book studies.

Selection and eligibility of articles

The results of each search in the different databases were imported into the bibliographic reference management software Mendeley®. Duplicate references were removed, and an initial selection by title and abstract was independently conducted by 2 researchers (F.G. and M.I.T.) using Rayyan® according to the outlined inclusion/exclusion criteria.

The full texts of the remaining references were obtained to determine their inclusion or exclusion in the final study based on the full text. Disagreements on the inclusion of an article were subject to discussion with a third researcher (B.M.) to reach a consensus. The PRISMA model was used to organize the information resulting from the article selection process.

Data extraction and analysis

Data were obtained using customized extraction forms. The following information was recorded for each study: (a) study ID, (b) author and year of publication, (c) country, (d) aim, and (e) population and characteristics. Two researchers performed the data extraction and synthesis processes independently (F.G. and M.I.T.). A third researcher (B.M.) resolved any disagreement. Finally, the data were sorted into tables through a narrative summary of the data extracted from each article.

Thus, the data related to spirituality identified in the studies with a qualitative component were recorded according to (a) author and year, (b) category, (c) subcategory, (d) descriptive theme, and (e) population. In studies with a quantitative component, the data were recorded according to (a) author and year, (b) measures and type of intervention, (c) findings, and (d) population. Two double-entry tables were constructed based on the dimensions evaluated for each study. Numerical referencing was used to identify the studies.

To map the knowledge obtained from the analysis of the themes and subthemes that emerged from the studies, a thematic construction and analytical structure was carried out, with the respective schematic and numerical representation of the included studies.

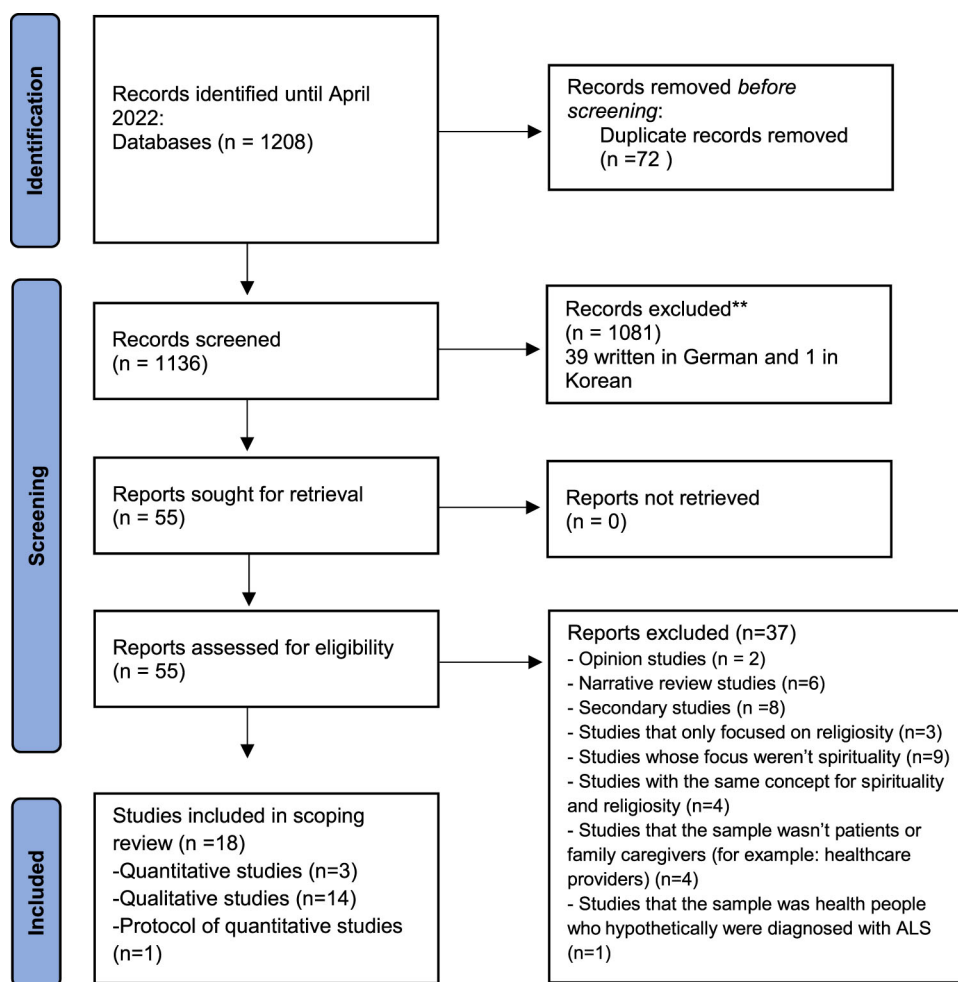


Fig. 1. Process of identification and inclusion of studies – PRISMA diagram flow.

Results

Selection of studies

A total of 1,208 articles were extracted from the initial search carried out in the different databases. After removing the duplicates, 1,136 articles were selected for the first analysis by title and abstract. The full-text analysis included 55 articles, of which 18 were considered for this review (Figure 1).

Characterization of the studies

Table 1 presents the summary information of the final studies analyzed, including authors, year, country, study aim, type of study, and population and characteristics. Most of the identified studies were qualitative ($n = 14$) (Akiyama *et al.* 2006; Cipolletta and Amicucci 2015; Dos Santos Costa *et al.* 2021; Fanos *et al.* 2008; Foley *et al.* 2007; Hamama-Raz *et al.* 2021; Locock *et al.* 2009; Madsen *et al.* 2018; O'Brien and Preston 2015; Ozanne *et al.* 2013, 2015; Rosengren *et al.* 2015; Warriar *et al.* 2020; Yuan *et al.* 2021) and used semi-structured interviews for data collection except 2 studies: one used biographies selected through a search by keyword (Rosengren *et al.* 2015) and the other used internet and print-published narratives written by people with ALS/MND (O'Brien and Preston 2015). Three were identified as quantitative

(Bentley *et al.* 2014; Fegg *et al.* 2010; Murphy *et al.* 2009): 1 was a cross-sectional study that utilized a single treatment group (Bentley *et al.* 2014) and the other 2 were cross-sectional studies with the application of scales (Fegg *et al.* 2010; Murphy *et al.* 2009). The protocol (Bentley *et al.* 2012) was the basis for a study (Bentley *et al.* 2014).

Characterization of the participants

The characteristics of the participants in the selected studies are summarized in Table 1. A total of 378 patients participated in the studies, 214 ALS patients and 164 caregivers. In one study, the sample was not the primary caregiver but all family members in contact with the patient and somehow care for them (Cipolletta and Amicucci 2015).

Three studies used a sample with ALS patients and caregivers (Bentley *et al.* 2012; Locock *et al.* 2009; Madsen *et al.* 2018). One of them was the protocol that recruited a total of 100 participants, of which half were ALS patients and the other half caregivers (Bentley *et al.* 2012).

From 10 studies that included ALS patients, 4 did not indicate the sex of the patients (Foley *et al.* 2007; Hamama-Raz *et al.* 2021; Locock *et al.* 2009; O'Brien and Preston 2015), 4 did not indicate the mean age (Foley *et al.* 2007; Locock *et al.* 2009; O'Brien and

Table 1. Characteristics of the included studies

Study ID	Author (year)	Country	Aim	Study	Population and characteristics, patients (<i>n</i>), age, %female, time since diagnosis; caregivers (<i>n</i>), age, %female, relationship
S1	Akiyama et al. (2006)	Japan	Explain the experiences of caregivers of patients with ALS receiving invasive ventilation in Japan	Qualitative	Caregivers (<i>n</i> = 12), mean 56.1 years, 83% female, 9 spouses, 2 mothers, and 1 daughter
S2	Foley et al. (2007)	Ireland	Explore self-referent “meaning” of QoL as perceived by individual ALS patients and to explore how their experience of health care affected their subjective well-being	Qualitative	Patients (<i>n</i> = 5), 6 months–2 years of diagnosis
S3	Locock et al. (2009)	USA	Examine the relevance of biographical disruption and repair to the experience of MND	Qualitative	Patients (<i>n</i> = 35); caregivers (<i>n</i> = 11)
S4	Fanos et al. (2008)	USA	Explore hope in patients with ALS	Qualitative	Patients (<i>n</i> = 16), mean 54 years, 19% female, mean 3.2 years
S5	Murphy et al. (2009)	USA	Determine the contributions of social problem-solving, relationship satisfaction, spirituality, and religiosity in predicting the quality of life and psychological morbidity in a sample of ALS caregivers	Quantitative	Caregivers (<i>n</i> = 75), mean 58 years, 69.3% female, 85.3% spouses, 6.7% child, 1.3% sibling, 1.3% friend, and 5.3% other
S6	Fegg et al. (2010)	German	Investigate meaning in life in ALS patients and compare the findings with data from a representative sample of the German population	Quantitative	Patients (<i>n</i> = 46), 6.5%, 40–49 years, 15.2% 50–59 years, 50% 60–69 years, 28.3% >70 years, 50% females
S7	Bentley et al. (2012)	Australia	Assess the feasibility, acceptability, and potential effectiveness of dignity therapy to enhance the end-of-life experience for people with MND and their family caregivers	Quantitative (protocol)	Patients (<i>n</i> = 50), >18 years; caregivers (<i>n</i> = 50), >18 years
S8	Ozanne et al. (2013)	Sweden	Illuminate how people with ALS create meaning despite the disease	Qualitative	Patients (<i>n</i> = 14), mean 67.5 years, 50% female, 2–13 years
S9	Bentley et al. (2014)	Australia	Assess the feasibility, acceptability, and potential effectiveness of dignity therapy for family caregivers of people with MND	Quantitative	Caregivers (<i>n</i> = 18), mean 61 years, 72% female, 100% spouses
S10	Ozanne et al. (2015)	Sweden	Illuminate experiences of finding meaning in life among spouses of people with ALS	Qualitative	Caregivers (<i>n</i> = 13), mean 68 years, 38% female, 100% spouses
S11	O’Brien and Clark (2015)	United Kingdom	Understand how personal spirituality and religiosity might help those living with ALS/MND cope with their impending death	Qualitative	Patients (<i>n</i> = 54), age at diagnosis 20–71 years
S12	Cipolletta and Amicucci (2015)	Italy	Explore the experience of family members who have lived alongside ALS patients until their death	Qualitative	Family members (<i>n</i> = 13), mean 44 years, 77% female, 2 sons, 6 daughters, 3 spouses, 1 girlfriend, and 1 sister
S13	Rosengren et al. (2015)	Sweden	Describe patients’ experiences of living with ALS in the end-of-life situations	Qualitative	Patients (<i>n</i> = 4), 100% female
S14	Madsen et al. (2018)	Denmark	Gain insight into experiences and reflections of persons with ALS and relatives concerning the peer group rehabilitation programme “More Life – Less Illness”	Qualitative	Patients (<i>n</i> = 8), 25% female, age at diagnosis 50–79 years; caregivers (<i>n</i> = 10), 90% females, 80% spouses, and 20% close relative
S15	Warrier et al. (2020)	India	Explore the lived experience of spouses of persons diagnosed with MND	Qualitative	Caregivers (<i>n</i> = 2), 36 and 33 years, 100% spouses
S16	Hamama-Raz et al. (2021)	Israel	Gain a deeper understanding of hope as experienced by people living with ALS	Qualitative	Patients (<i>n</i> = 12), mean 60 years, mean 4.8 years
S17	Dos Santos Costa et al. (2021)	Brazil	Understand the existential transformations of the family caregiver of a person living with ALS	Qualitative	Caregivers (<i>n</i> = 12), 92% females, 18–71 years, 33% daughters, 25% spouses, 17% sisters, 9% father, 8% sister-in-law, and 8% affinity relative
S18	Yuan et al. (2021)	China	Gain a comprehensive understanding of the illness experience of ALS patients in China and the meaning they attach to those experiences	Qualitative	Patients (<i>n</i> = 20), 35% female, mean 51 years, mean 1.2 years

Note: ALS, amyotrophic lateral sclerosis; MND, motor neuron disease; QoL, quality of life.

Preston 2015; Rosengren *et al.* 2015), and 3 did not indicate the time since diagnosis (Fegg *et al.* 2010; Locock *et al.* 2009; Rosengren *et al.* 2015).

From the 8 studies that included caregivers, 6 presented spouses as being the most prevalent family related in the care of patients with ALS. Only 1 study showed incomplete information about the mean age, gender, and relationship with the patient (Locock *et al.* 2009).

Characteristics of studies with a qualitative component

Table 2 describes the resulting categories and subcategories of the studies. The main themes for caregivers and patients are represented in Figure 2.

Caregivers

Caregivers highlighted the meaning in caring by continuous processing of knowledge and the understanding of ALS, where caregivers intensified experiences living a shared meaningful everyday life among PALS (Madsen *et al.* 2018). For the family caregiver, the process of care was more than an act and assumed a sense, driven by love and a mutual exchange understood in the valorization of being, giving meaning to the life of both (Dos Santos Costa *et al.* 2021).

Caregivers reported changes in the marital relationship, intimacy, and communication between the couple, and in addition, they did not have time for themselves since caregiving became their primary focus and major responsibility (Locock *et al.* 2009; Warrior *et al.* 2020). However, they reported that relationships with others and the environment were illuminated as meaningful for their own life and must be preserved (Ozanne *et al.* 2015).

Spirituality involved the imminence of the loss process, as well as the need to adapt to a new reality in life, which included the addition of limitations of basic functions that fleetingly impact the routine and habits (Dos Santos Costa *et al.* 2021). The course of the disease alters normative life transitions of the caregivers, namely, with the increasing need for care, for example, when mechanical ventilation is requested to prolong the life of an ALS patient (Akiyama *et al.* 2006). A feeling of being supported by health-care professionals, self-help groups, and other people influenced finding meaning in prolonging life (Akiyama *et al.* 2006; Madsen *et al.* 2018; Ozanne *et al.* 2015).

Even in the end stage, when patients cannot react, caregivers reported that they find other meaning in the situation to continue caring for them (Akiyama *et al.* 2006). Shortly after the partner's death, a shift is highlighted in the wishes for their own life to change, and thoughts of a continued life arose (Ozanne *et al.* 2015).

Patients

Patients expressed 2 different ways of living with the disease: either to lie down, be angry, and let the disease take over the everyday life or to live right here and now, the time that is left. In Rosengren *et al.*'s (2015) study, female patients highlighted that they took control of their life situation by choosing to live in the moment. While trying to make sense of life, some patients search for a positive aspect to emerge as a tranquil composure and hope, acquiring strength from turning misfortune into an advantage (Bentley *et al.* 2014; Hamama-Raz *et al.* 2021). They argue that the feeling gets stronger to utilize all moments regarding a limited lifetime where every second counts (Rosengren *et al.* 2015).

Several participants stated that their perspectives had changed. Appreciation of the new life was interpreted differently by patients:

some, instead of enjoying participating in activities with their loved ones, were now able to enjoy watching them (Fanos *et al.* 2008). Other perspectives included gratitude for past experiences, appreciation of residual time and function, living in the present, appreciation of the natural world, and the importance of getting things done (Foley *et al.* 2007; Ozanne *et al.* 2013). Others hoped that the disease would stop or at least not become much worse and survive over a particular time (Hamama-Raz *et al.* 2021; Ozanne *et al.* 2013; Yuan *et al.* 2021). Many kept their mind occupied by focusing on daily activities or hobbies like continuing to work while still able to do. However, some chose to stop work to concentrate on other more valued aspects of their lives, make holiday trips as a distraction, as an assertion that life was not yet over and was there to be enjoyed, writing to reflect on their lives, and meditation to help in their search for giving meaning to their situation (Fanos *et al.* 2008; Locock *et al.* 2009; Yuan *et al.* 2021).

Computer technology accessibility and augmentation played an increasingly important part in this new way of life by providing opportunities for virtual socializing through online forums, access to information to enable people to manage their own condition, and, crucially, voice software for augmentative and alternative communication to restore a voice to those unable to speak (Locock *et al.* 2009).

A sense of community that supported the internal relationships between PALS and their relatives occurred, building up a sense of fighting ALS together (Madsen *et al.* 2018; Yuan *et al.* 2021). This sense of community was reported as meaningful in different studies, where the process of sharing the experience of living with ALS among patients and caregivers was very important and self-described by patients as "openness, understanding, joy of life, and humor." Sharing laughs can also be important and helps to disarm the tragedy of the disease; a good sense of humor and calm with caregivers and health-care professionals helped to deal with problems when something went wrong and feel being accepted as an individual (Fanos *et al.* 2008; Ozanne *et al.* 2013; Rosengren *et al.* 2015).

Characteristics of studies with a quantitative component

Bentley *et al.* 2012 (S7) created a study protocol with the potential to provide a precise intervention based on Dignity therapy to ameliorate psychosocial and existential distress and improve the quality of care provided to people with ALS and their family caregivers. The cross-sectional study S9 (Bentley *et al.* 2014), developed by the same author in 2014, showed that the individual results on anxiety and depression are encouraging and suggest that dignity therapy has the potential to decrease anxiety and depression in family caregivers who are experiencing moderate to high levels of distress. Family caregivers felt that the therapy provided a benefit to their family members and that the document would help them in bereavement, describing the experience as satisfactory and recommending it to others. Whether a family caregiver was directly involved in the therapy or not had little impact on the acceptability or feasibility of the intervention. However, family caregivers' level of acceptance of their partner's imminent death, or the quality of the relationship between family caregiver and partner, may be negatively impacted, as suggested by the comments that were provided on the feedback questionnaire, which can lead to dignity therapy having a potentially negative impact on family caregivers at the time of the intervention.

Murphy *et al.* (2009) used different scales to study the relationship between several variables in caregivers; for example,

Table 2. Characteristics of studies with a qualitative component

Study ID	Author (year)	Category	Subcategory	Theme description	Population
S1	Akiyama et al. (2006)	“Finding meaning in prolonging life”	–	The motivation to continue caring for patients was related to whether caregivers seemed to have found meaning in this decision	Caregivers
			“Feeling of being supported”	Finding meaning in prolonging life was described by caregivers when they received support from people around them. This led to a feeling of being supported and continued caring for patients	
			“Uncertainty of the future”	Caregivers hoped that the situation would improve if patients received invasive ventilation. When they faced the fact that the disease progressed even after receiving invasive ventilation, they could not find any meaning in receiving invasive ventilation	
			“Communication”	Good communication represented the motivation to continue caring for patients. After ventilation began, many of the cases in which finding a meaning in prolonging life was related were those in which caregivers experienced good communication with patients	
			“Maintaining their own life”	Even if the disease progressed to the final stage and patients were unable to react, caregivers find other meaning in the situation	
			“Support”	At all stages, support from visiting nurses, members of self-help groups, and other people around provided the motivation for continuing to care for patients and had an influence on finding meaning in prolonging life	
S2	Foley et al. (2007)	“Importance of family”	–	Was evident from participants’ recognition of their dependence on family and from their appreciation of family support	Patients
			–	Importance of family, social, and health services support and about the welfare of others	
			–	Desire to maintain independence, adopting healthy behaviors or conversely worrying less about other health issues, seeking to maintain normalcy, perception of health as independent of ALS, and desire for a cure	
			–	All participants shared a strong appreciation of life. Uncertainty regarding the future prompted participants to value the present and display gratitude for past experiences	
S3	Locock et al. (2009)	“Biographical repair”	“Keeping hold of the old normality”	People sought to restore a sense of normality in different ways, at different stages in their illness. Doing the same things for as long as possible was a common focus	Patients and caregivers
			“Creating a new normality”	To keep a “normal life” going and to distract them from thinking about the future. Some people found new activities they loved	
			“Living life to the full – a heightened normality”	Confronting a dramatically shortened life span led many people to start cramming in all the things they had wanted to do in the rest of their lives, telescoping a previously envisaged normality into a few months or years	
			“Finding new meaning”	Participants reflected on their changed attitude to life, the new value they attached to just being and living for the moment, and the preciousness of each day	
S4	Fanos et al. (2008)	“Hope”	“Social support”	Participants discussed the importance of social support from friends, family, the medical team, and even their pets	Patients
			“Search for information”	Participants actively sought the latest research concerning ALS. Some of them were happy to participate in various research trials, knowing that a cure may not be possible in their lifetime	
			“Adapting to changing capacities”	The process of mourning lost abilities and taking pleasure in new ones appeared to be very important in maintaining hope	
			“Living in the moment”	Participants tried to live each day to the fullest, attempting neither to dwell on their illness nor look too far into the future	
			“Self-transcendence”	Participants expressed acceptance and altruistic concern for others	

(Continued)

Table 2. (Continued.)

Study ID	Author (year)	Category	Subcategory	Theme description	Population
S8	Ozanne et al. (2013)	“Finding meaning despite the illness”	“Family and friends give strength”	After the disease developed, families and friends were sources of meaning and strength	Patients
			“Giving and receiving help”	Making life meaningful for participants implies feeling safe in knowing that they could trust to receive the help they might need and feeling that they were needed and could help others	
			“Having one’s own life”	An active life created a feeling of freedom and meaning appeared when participants could fulfil their own interests	
			“Accepting the present”	This acceptance implied that participants found important things in life here and now	
			“Life perspectives grow from shallow to deep”	It was no longer so important to spend energy on things that had previously seemed irritating, and material things. The disease brought another dimension of what was important in life	
S10	Ozanne et al. (2015)	“Finding meaning despite the proximity of death”	“Cherishing one’s own life”	For the spouses, meaning in life arose from having time for recreation, relaxation, and reflection	Caregivers
			“Gaining strength from fellowship”	The family, friends, and the possible well-being of the ill spouse was a great source of happiness and meaning in life	
			“Accepting the present”	Once some time had passed after the diagnosis, it was possible to enjoy the sun and rejoice with others again	
			“Believing in meaning after a partner’s death”	Experiences of a lost belief in the future and no wish of having one’s own life after the partner’s death changed after a while, and thoughts of a continued life arose.	
S11	O’Brien and Clark (2015)	“Spirituality”	–	Spiritual life involving one’s interior journey, thinking, meditating, and arranging one’s own feelings and views of the world.	Patients
S13	Rosengren et al. (2015)	“Experiences of a Limited life”	–	Was described by women as a constant battle finding meaningfulness within while suffering	Patients
			“Meaningfulness”	Meaningfulness was described by women as strength, joy, and the ability to live in the moment	
S14	Madsen et al. (2018)	“Sense of Community Building”	–	Built up a shared social support network based on the sharing of experiences from an everyday life context	Patients and caregivers
			“Facing Facts”	Facing facts about existing in a state of limbo was transformed to less of a hard reality in which the participants could exist on their own terms and emphasize individual needs in obtaining meaningfulness	
			“Retaining Normality”	Live a meaningful life with a feeling of being able to do things as they used to, feeling included and valued as a person, and not being restricted about living a fulfilling life	
			“Understanding my ALS?”	In terms of making sense of their life situation, programme participation enabled the participants to form a kind of individual lay understanding of ALS pathology, and this acted as personal illness perception and explanation	
S15	Warrier et al. (2020)	“Relationship”	–	Change in the marital relationship and in time for themselves	Caregivers
		“Adaptation”	–	Adapt and readjust their life to the new normal. It’s a process of frequent challenges	
		“Life without the loved one”	–	Attempted to adapt to the absence of the partner by trying to get engaged in normal family routine	
S16	Hamama-Raz et al. (2021)	“The second voice: hope as a resource”	–	Hope as a source of meaning/purpose of life	Patients
S17	Dos Santos Costa et al. (2021)	“Being the caregiver of the person living with ALS”	–	Care contemplates the positive way of caring for participants’ loved ones, understanding that it is important and not as a synonym for kindness	Caregivers
		“Spirituality influencing the transformation process”	–	Spirituality helped to relieve the daily burden of stress for family caregivers through potential mechanisms of being a dimension of transcendence and an individualized experience	

(Continued)

Table 2. (Continued.)

Study ID	Author (year)	Category	Subcategory	Theme description	Population
S18	Yuan et al. (2021)	“Family self-help: ‘we kept an eye on each other’”	“Family warmth and hardship”	Due to the illness, patients and their families spent more time with each other than before	Patients
		“Reconstruction of life: ‘What was the meaning of my life?’”	“Learning to accept”	Learning to accept reality and death was the most crucial first step to embark on a journey of meaning-making. Patients could reconsider their value and rediscover the meaning of life	
			“Rebuilding self-worth”	Participants rebuilt their self-worth to make the meaning of life investing their energies in activities to feel valued and needed	
			“Resetting the priority list”	The meaning of life can be found by resetting the priority list; now, spending time with family has become the most important thing for them	
			“Living in the moment”	Patients chose to live in the moment to liberate themselves from the disease temporarily and reduce worries to a manageable level	

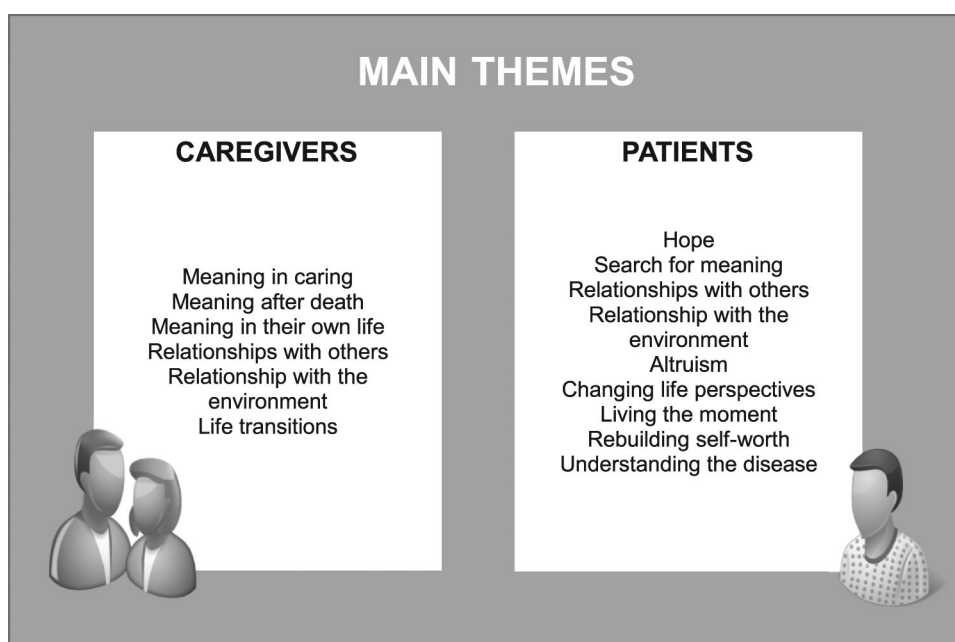


Fig. 2. Main themes for caregivers and patients, based on the qualitative studies.

the relationship between spirituality, measured by the Spiritual Perspective Scale (SPS), with quality of life (QoL), measured by the Quality of Life Inventory, Overall Life Satisfaction Score (QoLI), and psychological morbidity, measured by the Brief Symptom Inventory, General Severity Index Score (BSI). Spirituality was considered a significant predictor of QoL (0.321; $p < 0.01$) but not a significant predictor of psychological morbidity.

Fegg et al. (2010) used the Schedule for Meaning in Life Evaluation Scale (SMiLE), a measure to assess the meaning in life (MiL), in PALS comparing with a representative sample of the German population. Subjects of the representative sample showed a significantly higher SMiLE index (indicating the overall importance of the respondent’s MiL areas) compared to PALS ($p < 0.001$). This was due to a significantly higher satisfaction in the listed MiL areas ($p < 0.001$). However, importance ratings between importance and satisfaction did not differ significantly.

The results of the studies are described in Table 3.

Discussion

This scoping review broadly examined the nature and extent of research on the relevance of spirituality and the role of spiritually integrated interventions targeting PALS and their caregivers. Most literature is based on qualitative approaches. These are useful to identify experiences and self-related perceptions, especially in the light of spirituality, a domain where open questions and narratives can be a useful method to understand and narrow the path of the interventional approaches (Fanos et al. 2008; Foley et al. 2007; Madsen et al. 2018; O’Brien and Preston 2015; Ozanne et al. 2015; Rosengren et al. 2015). Furthermore, qualitative studies highlight the importance of differential approaches according to the stage of the disease and its progression (Akiyama et al. 2006; Dos Santos Costa et al. 2021), including the availability to speak and communicate and to use augmentative and alternative strategies that allow PALS to express their insights during the course of the disease and

Table 3. Studies with a quantitative component

Study ID	Author (year)	Measures and type of intervention	Findings	Population
S5	Murphy et al. (2009)	Scale: SPS: Spiritual Perspective Scale	Spirituality was correlated with, and was a predictor of, caregiver QoL (0.321; $p < 0.01$) but was not a predictor of psychological morbidity (-0.034 ; $p > 0.05$).	Caregivers
S6	Fegg et al. (2010)	Scale: Schedule for Meaning in Life Evaluation' (SMiLE)	In conclusion, the SMiLE reflects different components of MiL, which are differentially affected. The areas listed by a patient and the importance ratings describe the framework of how an individual finds meaning. This aspect of MiL appears sustained during the illness. Where ALS patients suffer a progressive loss, however, is with regard to the fulfilment of MiL. This aspect is reflected in the diminished satisfaction ratings.	Patients
S7	Bentley et al. (2012)	Intervention: Dignity therapy study protocol	Dignity therapy offers people with terminal illness the opportunity to create a generativity document. In a recorded interview guided by a counsellor or health-care professional, the participant is invited to recount aspects of their life they want remembered, find meaning and purpose to their life, and express final words or advice.	Patient and caregivers
S9	Bentley et al. (2014)	Intervention: Dignity therapy	There were no significant pretest posttest changes on the group level, but there were decreases in anxiety and depression on the individual level. Baseline measures indicate that 50% of family caregivers had moderate to severe scores for anxiety prior to dignity therapy. MND family caregivers saw benefits to the person with MND and to themselves after bereavement, but acceptability of dignity therapy at the time of the intervention was mixed, with some family caregivers indicating it was helpful, some indicating it was harmful, and many expressing ambivalence. Dignity therapy involving MND family caregivers is feasible, and the involvement of family caregivers has minimal impact on the therapy.	Caregivers

Note: ALS, amyotrophic lateral sclerosis; QoL, quality of life; MiL, meaning in life; SMiLE, Schedule for Meaning in Life Evaluation Scale.

to actively communicate with their family and caregivers (Locock et al. 2009).

The quantitative instruments adopted by studies on the spiritual dimension is relevant, and the research summarized by the present scoping review considers only a small number of them: SPS and SMiLE (Fegg et al. 2010; Murphy et al. 2009). Nonetheless, others can be found in the literature and can be used in further studies to evaluate and to monitor PALS and caregivers' progression (Blaber et al. 2015), with some common instruments that can be used in a palliative care setting to measure QoL, in general, and spiritual well-being, in particular, as well as different domains of spirituality (relation with themselves, other people, environment, or transcendental). When selecting the appropriate instrument in PALS, researchers and health-care providers should consider the clinical and cultural traits of the population, as well as the validation of their psychometric properties (Long 2011). This study highlights the importance of integrative care that goes beyond the biologic aspects of interventions and that considers the spiritual needs. Therefore, it can encourage professionals to consider this dimension when delivering care to PALS and caregivers and consider this dimension a useful and indispensable component of palliative care (Kang et al. 2012). Accordingly, this increases the need for increased investment in health-care professionals' education, training, and capacitation for times when healing therapies can no longer control the disease and spiritual support can help manage distress, meaningfulness, and QoL (Kang et al. 2012; Long 2011). The importance of spiritual care is highlighted, especially in a situation like ALS that affects a person's sense of meaning in life due to its impacts on physical activity, social participation, self-perception deterioration, and loss of

autonomy. The evidence included in this review suggests the need to address the loss of hope or meaning in life for PALS and their caregivers during the course of the disease and after the bereavement and continuous life transitions. Furthermore, this includes strategies targeting the rebuilding of the self-worth, changing life perspectives, and continuous capacitation and promotion of the ability to establish efficient relationships with others and the environment.

This scoping review indicates several directions for future investigation, especially systematically including spirituality in palliative care teams and interventions aimed at PALS and their caregivers.

Some limitations of the present study include the exclusion of studies in languages other than English, Spanish, Portuguese, or French. Second, as it was a scoping review, we did not systematically assess the scientific rigor of our literature sample.

It is also important to highlight an additional limitation resulting from the current state of the literature based on the nature and categorization of spirituality. The unclear delimitation of the boundary and definition of spirituality is also a finding in and of itself. Nevertheless, defining thematic subcategories in spirituality as we did in this scoping review can be a useful tool to guide health-care providers to better structure their intervention and assessment of the spirituality domains and their evolution along different stages of the disease or even after with regards to caregivers' self-identification and rebuilding their paths after bereavement.

More intervention should be empirically developed in order to test spirituality and spirituality care on the impact over QoL, coping, and readjustment to life at the different stages of PALS and

their caregivers. In addition, there is a clear need to raise awareness of this among health-care professionals involved in providing this care.

Conclusion

This scoping review illustrates the importance given to spirituality by caregivers and PALS with respect to 2 principal domains: (1) searching for meaning and purpose of life; (2) the relationships between them, others, and the environment. Overall, this review found support for spiritual-integrated care interventions for PALS and their caregivers. Spirituality is considered to be a unique part of everyday interactions and life, and spiritual care should always be considered as a way to target and improve patients' and their caregivers' QoL by addressing psychosocial and spiritual suffering and capacitation that can make the difference throughout the whole disease course, and the constant search for meaning was, in the most of times, the strength to continue the journey on its different domains.

Therefore, practitioners in contact with PALS and their caregivers must have proper training, formation, and capacity to assess and address the spiritual needs and to explore and put in practice ways of intervening and evaluating their applicability, being able to recognize the signs of distress experienced and prove an effective continuous follow-up and way to communicate with PALS.

Thus, it is of great importance to carry out experimental studies in the area of spirituality in order to systematically explore the impact of spiritual interventions and their results.

Conflicts of interest. The authors report no conflicts of interest.

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