

REVIEW ARTICLES

NEEDS AND CONCERNS OF SIBLINGS IN PEDIATRIC PALLIATIVE CARE: SCOPING REVIEW

NECESSIDADES E PREOCUPAÇÕES DOS IRMÃOS EM CUIDADOS PALIATIVOS PEDIÁTRICOS: SCOPING REVIEW

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ABSTRACT

Introduction: Understanding the needs of each family member in the setting of Pediatric Palliative Care is crucial. Sibling support is still an emerging area, with recommendations based on clinical experience and adaptation from other contexts. The aim of this study was to assess and describe the needs and concerns of siblings of children in Pediatric Palliative Care.

Methods: A literature search was conducted in Medline database. Only research articles with reports of siblings of children in Palliative Care were included. The main characteristics and results of studies included were summarized, and a narrative synthesis was performed.

Results: Nine studies were included showing that, although siblings' needs vary over the course of the disease, these generally include the need for self-support, access to information, and engagement in brother/sister care. Bereaved siblings are usually resilient, but poor-quality support can have a negative psychosocial impact.

Discussion and Conclusion: Siblings need informational, instrumental, appraisal, and emotional support. Quantitative prospective studies are needed, as well as accurate clinical practice recommendations taking into account the specificities of each child, family, and sibling.

Keywords: adolescent; child; emotional adjustment; palliative care; sibling

RESUMO

Introdução: Compreender as necessidades de cada membro da família em contexto de Cuidados Paliativos Pediátricos é crucial. O suporte aos irmãos ainda é uma área emergente, com recomendações baseadas sobretudo na experiência clínica e adaptadas de outros contextos. O objetivo deste estudo foi analisar e descrever as necessidades e preocupações de irmãos de crianças em Cuidados Paliativos Pediátricos.

Métodos: Foi efetuada uma pesquisa bibliográfica na base de dados Medline. Apenas artigos originais com relatos de irmãos de crianças em Cuidados Paliativos foram incluídos. As principais características e resultados dos estudos foram sintetizados, procedendo-se à sua descrição narrativa.

Resultados: Foram incluídos nove artigos, cuja análise demonstrou que, embora as necessidades dos irmãos variem ao longo do curso da doença, geralmente incluem a necessidade de suporte para o próprio, acesso a informação e envolvimento no cuidado ao irmão. Os irmãos enlutados são geralmente resilientes, mas um suporte de baixa qualidade pode ter um impacto psicossocial negativo.

Discussão e Conclusão: Os irmãos necessitam de suporte informacional, instrumental, de aprovação e emocional. São necessários estudos prospetivos quantitativos, bem como recomendações precisas para a prática clínica que considerem as especificidades de cada criança, família e irmão.

Palavras-chave: adolescente; ajustamento emocional; criança; cuidados paliativos; irmão

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INTRODUCTION

Pediatric Palliative Care (PPC) encompasses an active and global approach to the care of children and young people with life-limiting or life-threatening conditions (LLC or LTC), from the moment of diagnosis or recognition and throughout the child's life until potential death and beyond. It embraces physical, emotional, social, and spiritual elements and focuses on enhancing the quality of life of the child/young person and supporting the family.¹ Conditions in PPC can be grouped in four broad categories: (i) LTC for which there is curative treatment, but it can fail (e.g., cancer, irreversible organ failure of the heart); (ii) LLC with inevitable premature death (e.g., cystic fibrosis, muscular dystrophy); (iii) progressive LLC without curative treatment options (e.g., Batten disease); and (iv) non-progressive, irreversible LLC associated with severe disability, health complications, and premature death (e.g., cerebral palsy).¹ Pediatric conditions requiring palliative care are known to impact the family network.^{2,3} Coupled with the threat of premature death, the emotional impact on the family of a lifetime diagnosis in a child is profound.⁴ Furthermore, the demands of treatment may be highly disruptive, not only to parents but also to siblings at home.⁵ As a result, the family-centered approach in PPC should seek to maintain the integrity of each individual and family as a whole, providing guidance and support through the entirety of child medical care, from diagnosis to end of life and bereavement, and allowing time to prepare for impending challenges. For that purpose, understanding the needs of each family member, including siblings, is fundamental.^{3,6}

According to previous studies, siblings of children with cancer do not consistently show elevated rates of psychopathology, but they do have psychosocial needs that should be recognized and addressed, such as loss of needed attention and threatened sense of security within the family.^{7,8} The demands of caring for a child with cancer often limit «parents' physical and emotional availability to fully attend the needs of other children». Consequently, recommendations indicate that the extended family, health care professionals, siblings' school staff, and relevant community members should consider the unique needs of siblings, in addition to the needs of the family in general and the health of the child with cancer.⁷ Moreover, studies investigating the psychological functioning of siblings of children with chronic illnesses also show a risk of negative psychological effects, demanding intervention programs.⁸

Accordingly, support for siblings in PPC setting is widely recommended.¹ This support should include the identification of increased needs and access to more specialized support when required, assuming that most siblings will cope with upcoming challenges if the appropriate support is given. Bereavement support should also be provided to all children and young people experiencing the death of a sibling.¹ However, sibling support is still an emerging area, and proposed recommendations are based on clinical experience and adaptation from specific settings, as Pediatric Oncology or chronic diseases.^{7,8} In fact, although a variety of tools

have been developed to assess the needs of caregivers of adult palliative patients, few are in place for siblings of patients in PPC.⁶ Overall, there is a lack of primary research on the needs and concerns of siblings of children in PPC. Additionally, no reviews on the topic have been found in a preliminary search in Medline.

The aim of this scoping review was to assess and describe the needs and concerns of siblings of children in PPC, as a greater understanding of this subject may lead to improved sibling support and, eventually, more specific clinical recommendations.

METHODS

A scoping review was performed based on the methodological frameworks proposed by Arksey and O'Malley⁹ and Joanna Briggs Institute.¹⁰ First, the research question was defined: "What should a young researcher in Pediatric Palliative Care know about the needs and concerns of siblings?", pinpointing participants (siblings), concept (needs and concerns), and setting (PPC).

The literature search was conducted in Medline database until December 31, 2020, using the following queries: 1) ("*Siblings*"[Mesh] AND ("*Hospice and Palliative Care Nursing*"[Mesh] OR "*Palliative Medicine*"[Mesh] OR "*Palliative Care*"[Mesh] OR "*Hospice Care*"[Mesh] OR "*Terminal Care*"[Mesh] OR "*Hospices*"[Mesh]); 2) *children palliative care siblings*. The search strategy was limited by publication date (2000-2020) and language (*English or Portuguese*).

The following inclusion criteria were used: 1) research articles; 2) studies related to PPC; 3) studies having siblings themselves as study participants (as the evidence shows that children's perspective on their experiences offers useful augmentation to parental proxy reports, which may obfuscate some of the more sensitive issues and opinions);² 4) studies with siblings in the pediatric age range at the time of diagnosis. Exclusion criteria applied comprised: 1) studies related to adult Palliative Care; 2) studies whose participants were not siblings themselves (but instead parents, health professionals, etc.); 3) studies not exploring the needs and concerns of siblings in PPC; 4) studies with no abstract available; 5) review articles.

Data about place and date, aim, participants, design/measurements, and main results were retrieved from studies included in the analysis and narratively described.

RESULTS

The literature search retrieved 151 citations. After exclusion of duplicates, 131 articles were screened for eligibility, resulting in the further exclusion of 122 articles. In the end, nine articles were fully assessed and included in the analysis.

Data are summarized in **Table 1**. Studies included show that perceptions of the condition of the ill child and his/her symptoms, impact on daily life, emotional consequences, and way of coping

seem to be key issues for siblings of children in PPC.² Particularly, siblings report the need for their own support,¹¹ referring engagement in the exchange of information and in care of the brother/sister as relevant to them.^{6,11,12} Siblings also report insufficient or poor information regarding the ill child’s prognosis and psychological health outcomes, but also where to seek support for themselves.¹³ Siblings’ needs vary across the course of the disease: the most common problems are initially centered in deriving information to understand what is happening and why, in trying to keep up with self responsibilities (schoolwork) afterward, and in learning to cope with changes in the ill brother/sister in later stages. Regarding end-of-life, relevant concerns experienced by siblings include pain palliation, the ability to provide comfort to the brother/sister, the need to obtain information about death, preparing for death, and obtaining social support and family harmony.⁴ Bereaved siblings of

cancer patients are generally resilient and, although risky behaviors and psychological distress increase during the year after the brother/sister’s death, most return to baseline over time. Siblings who report dissatisfaction with communication, poor preparation for death, missed opportunities to say goodbye, and/or perceived negative impact of the cancer experience on relationships tend to have higher distress and lower social support scores.⁵ Furthermore, siblings’ perception of a nonpeaceful death and avoidance of physicians, poor medical information, and poor communication about the brother/sister’s death with family and friends predicted unresolved grief two to nine years post-loss.¹⁴ On the other hand, supporting the siblings of children with cancer throughout the cancer journey and afterward into bereavement has shown to have a positive buffering effect on their own endurance and personal growth, family cohesion, and social support.¹⁵

Table 1 - Characteristics and main results of studies included in the analysis

<i>Place and Date</i>	<i>Aim</i>	<i>Participants</i>	<i>Design / Measurements</i>	<i>Main Results</i>
<i>Freeman et al, 2003</i>				
USA 1999-2000	Identify the main concerns of children with brain or spinal cord tumors and siblings during different phases of illness. <i>Note: in this review, only results concerning siblings were considered.</i>	25 children with brain or spinal cord tumors and 32 siblings (mean age of siblings: 17 years [Standard Deviation (SD) 5.5]; average time from diagnosis to survey: 4.5 years [SD 3.0]; no information on gender).	Participants responded to a survey about health care provider interactions, medical information, health care utilization, and psychosocial aspects, rating individual items as helpful, a problem, and for importance.	The following problems (>30% of siblings) and helpful resources were reported: 1) at diagnosis: lack of information about etiology and prognosis and the manner physician and parents provided information; family and social support, as well as family harmony, were the two most commonly reported helpful resources; 2) during hospitalization/surgery: lack of information about prognosis, lack of help with schoolwork; in addition to family and social support, liberal visitation policies were reported as helpful and very important; 3) after hospital discharge: lack of help with schoolwork; support from friends, family, and religion were helpful and important; 4) during adjuvant treatment: lack of help with changes in sister or brother’s appearance, physical activity, mood, and information about the treatment; 5) at end of life: treatment of sister/brother’s pain, lack of information about dying, family harmony (although family and social support were reported as the leading helpful resource), support from friends, help with schoolwork, and preparation for the death; their ability to comfort their brother or sister was helpful and important.
<i>Malcom et al, 2013</i>				
UK 2009-2010	Report sibling experiences related to two rare degenerative and progressive conditions (Mucopolysaccharidoses (MPS) and Batten Disease).	8 siblings of children with MPS (n=7) and Batten Disease (n=1) (mean age: 10.5 years, range 7–12 years; 5 males and 3 females).	Semi-structured qualitative interviews were administered to participants and analysis was informed by grounded theory.	Four key themes demonstrated impact on siblings: 1) perceptions of the condition and its symptoms (siblings often had considerable knowledge of the condition, yet they protected parents from an awareness of how much they knew); 2) impact on daily life (while many spoke affectionately about their caregiving roles – supporting others to care, rather than taking full responsibility-, they also experienced limitations in social activities as a result of their sibling’s condition; this was described with acceptance and sadness); 3) emotional consequences (concern often dominated their thoughts, leading to difficulties in concentrating at school and also impacting on their social time with friends; they were also very aware of how worries could reverberate around the family system; siblings were affected by negative social attitudes towards disabilities, namely by peers); and 4) ways of coping (organised sibling support groups were mentioned as important; several siblings described their main sources of support as being family members and friends).

<i>Gaab et al, 2014</i>				
New Zealand 2010-2011	Identify the concerns of siblings of PPC patients.	18 siblings of PPC patients (aged 9 to 22 years, including bereaved (six months to two years after their sibling's death) and nonbereaved children; no information on gender).	Semi-structured interviews were administered to participants and analysed using qualitative inductive thematic analysis.	Participants held two main concerns: 1) Most siblings felt it was important to discuss the impending death, because it increased their understanding of the situation (this knowledge gave them a greater appreciation of their sibling; a few mentioned anger at being 'left in the dark' or expressed confusion and fear of siblings' symptoms without having them explained; siblings generally wanted to be informed of their ill siblings' health statuses but did not want death/mortality to be the topic of every conversation); 2) Most siblings expressed the importance of helping the ill child (with practical support such as transporting wheelchairs or fetching medications, but also with games, schoolwork, etc; they helped in order to worry less, provide comfort, keep things positive and normal; they recognized the ill children's needs were greater than their own and most expressed gratitude that the ill children received extra attention).
<i>Rosenberg et al, 2015</i>				
USA 2008-2009	Describe the prevalence of risky health behaviours, psychological distress, and social support among bereaved siblings and potentially modifiable factors associated with poor outcomes.	58 bereaved siblings (mean age at survey: 25.6 years [SD 7.8]; mean age at sibling's diagnosis of cancer: 10.9 [SD 6.2]; mean age at sibling's death: 13.8 [SD 7.3]; on average 11.8 years had passed since their sibling's death [SD 3.2]; 40 females and 18 males).	Survey-based study; linear regression models identified associations between personal perspectives before, during, and after the family's cancer experience and outcomes (health behaviours, psychological distress, and social support).	Anxiety, depression, and illicit substance use increased during the year after the brother/sister's death but then returned to baseline. Siblings who reported dissatisfaction with communication, poor preparation for death, missed opportunities to say goodbye, and/or a perceived negative impact of the cancer experience on relationships tended to have higher distress and lower social support scores. Almost all siblings (88%) reported that their loss still affected them; half stated that the experience impacted current educational and career goals (for example, 12% reported that their experience had negatively impacted their work or career, whereas 45% reported a positive impact on work or career). Personal growth was reported in subsets of cases (36% reported that they were better communicators, 43% more mature, 45% more kind-hearted, and 17% more confident than others their age). None of these responses (personal growth, impact on education/career) were related to current distress or social support.
<i>Lövgren et al, 2016</i>				
Sweden 2009	Explore bereaved siblings' advice to healthcare professionals (HCP) working with children with cancer and their families.	108 bereaved siblings (mean age at time of death: 18 years [SD 3.7]; mean age at data collection: 24 years [SD 3.7]; mean time since loss 6.3 years [SD 2.2]; 69 females and 39 males).	Participants answered an open-ended question about what advice they would give to HCP working with children with cancer and their families; responses to this single question were analysed using content analysis.	Six categories of advice were constructed: 1) <i>Siblings' Wish for Own Support</i> (56%; included support from someone to talk to, support groups, or other kinds of activities for siblings; support in daily life, for example, with homework, school activities, and hobbies); 2) <i>Siblings' Wish for Information About Their Brother's or Sister's Disease and Care</i> (31%; siblings felt that information should be given continuously during all the different phases; explanations given in a playful way, such as with illustrations using cartoons and emojis, were appreciated more than those given in a more formal way); 3) <i>Siblings' Wish to Participate in the Care of Their Brother or Sister</i> (7%; these siblings wished HCP involved them more); 4) <i>Support and Information to Parents</i> (3% of siblings suggested that parents should receive information from HCP about how to talk to, help, and involve siblings in their brother's or sister's care); 5) <i>Advice About the Brother's or Sister's Care</i> (12%; siblings emphasized the importance of letting the ill brother or sister have a normal life, and of asking them how they wanted to be cared for); 6) <i>Psychosocial Aspects to Consider in Relation to the Affected Family</i> (44%; common suggestions were related to positivity, hope, and happiness, but also to realism and honesty). No differences between sexes or age groups in relation to the type of advice were found, except that significantly more women reported advice associated with the wish for own support.

Eilertsen et al, 2018				
Sweden 2009	Explore cancer-bereaved siblings' positive and negative memories and experiences of their brother's or sister's illness and death.	123 bereaved siblings (between 12 and 25 years old when their brother or sister died and between 19 and 33 at the time of data collection; no further age or gender data).	Participants responded to two open-ended statements, which focused on siblings' positive and negative memories and experiences of illness and death. The data was analyzed using systematic text condensation.	Bereaved siblings have many positive memories and experiences, even though the death of a brother or sister is a distressing and grievous situation; sick siblings, as well as parents, seem to play important roles in the shaping of their healthy siblings' experiences. The bereaved siblings' responses were categorized into four different themes: 1) <i>endurance versus vulnerability</i> (endurance was expressed as the influence that the ill siblings' strong willpower, good mood, and stamina in their difficult situation had on healthy siblings, whereas vulnerability was expressed as the feeling of emptiness and loneliness involved with having an ill and dying sibling); 2) <i>family cohesion versus family conflicts</i> (family cohesion was expressed as the bonds being strengthened between family members, whereas family conflicts often led siblings to feel invisible and unacknowledged); 3) <i>growth versus stagnation</i> (most siblings expressed the feeling that they grew as individuals in the process of their brother's or sister's illness and death, whereas others experienced stagnation because of the physical and mental distress they bore throughout this time, often feeling forgotten); 4) <i>professional support versus lack of professional support</i> (most siblings perceived physicians and staff at the hospital as being warm, kind, and honest, while some siblings had negative experiences).
Lövgren et al, 2018				
Sweden 2009	Identify modifiable or avoidable family and care-related factors associated with unresolved grief among cancer-bereaved siblings two to nine years post loss.	174 cancer-bereaved siblings (mean age at time of death: 18 years [SD 3.7]; mean age at data collection: 24 years [SD 3.8]; 101 females and 73 males).	Participants answered 29 close-ended questions on grief, family and care-related factors, as well as the Hospital Anxiety and Depression Scale. A multivariable prediction model was built.	Several predictors for unresolved grief were identified: siblings' perception that it was not a peaceful death [odds ratio (OR): 9.86, 95% confidence interval (CI): 2.39–40.65], limited information given to siblings the last month of life (OR: 5.96, 95% CI: 1.87–13.68), information about the impending death communicated the day before it occurred (OR: 2.73, 95% CI: 1.02–7.33), siblings' avoidance of the doctors (OR: 3.22, 95% CI: 0.75–13.76), and lack of communication with family (OR: 2.86, 95% CI: 1.01–8.04) and people outside the family about death (OR: 5.07, 95% CI: 1.64–15.70). Depressive symptoms (OR: 1.27, 95% CI: 1.12–1.45) and time since loss (two to four years: OR: 10.36, 95% CI: 2.87–37.48 and five to seven years: OR: 8.36, 95% CI: 2.36–29.57) also predicted unresolved grief. Together, these predictors explained 54% of the variance of unresolved grief.
Wallin et al, 2020				
Sweden 2009	Explore cancer-bereaved siblings' advice to peers with a brother or sister with cancer.	125 cancer-bereaved siblings (mean age at time of brother's / sister's death: 17.9 years [SD 3.6]; mean age at data collection: 24 years [SD 3.8]; 74 females, 51 males).	Participants answered an open-ended question about what advice they would give to peers with a brother or sister with cancer; responses to this single question were analysed using content analysis.	Siblings gave 257 pieces of advice, presented in four categories: 1) <i>Be together</i> (n=131; a majority of the advice from cancer-bereaved siblings to peers related to being with the ill brother or sister, participating in his/her care, cherishing the time together, staying friends, doing fun things together, saying how much the brother or sister means, saying goodbye, and letting him/her know that he/she will never be forgotten); 2) <i>Communicate openly</i> (n=81; communicate openly with the ill brother or sister, but also with parents, peers, in school and with HCPs, asking for help and information); 3) <i>Let go of guilt</i> (n=27; siblings also advised peers not to blame themselves, stressing the importance of making themselves heard and showing their feelings in order to avoid loneliness); 4) <i>Live life as usual</i> (n=18; keep taking part in everyday activities like going to school, after-school programs and other things that would make life go on as before; the siblings advised peers to occasionally take a break from illness and death, as a way of taking care of themselves).

Lövgren <i>et al.</i> , 2020				
Sweden 2018-2019	Explore how families in pediatric oncology experienced illness-related information and communication with HCP and within the family. <i>Note: in this review, only results concerning siblings were considered.</i>	118 family members, representing 27 families. 38 siblings participated (20 females, 18 males; mean age 12 years [SD 5.2]).	Participants responded to open and closed questions; those aged ≥13 years also filled the Family Adaptability and Cohesion Scale IV (FACES IV) Family Communication. Descriptive statistics and content analysis were applied.	All siblings reported that someone had told them about the cancer illness, but 45% of the siblings wanted to know more about the illness. A vast majority of the siblings aged 13 years and older (n=14) reported that they had not received information or that they wanted more information regarding several areas: prognosis (64%), how the cancer and its treatment could affect the child's physical (64%) and psychological health (71%), where or whom they could turn to if they have questions about the ill child's care (57%), and where or whom they could turn to for own support (50%). The siblings described that they did not dare talk to HCP because "they talk around things," which resulted in even more uncertainty. Moreover, the siblings reported that HCP interpreted everything as negative criticism, which led the siblings to keep silent. 52% of siblings reported that they had feelings or thoughts that they did not want to share with their family, related to their school situation, sadness at night, feeling neglected, and anger/disappointment at the parents. Conversely, 24% siblings reported that they wanted to reveal more about how they felt to someone in the family.

HCP, health care professionals; MPS, mucopolysaccharidoses; PPC, Pediatric Palliative Care; SD, standard deviation

DISCUSSION

Results of this study indicate that there is room for improvement in the support to siblings of children in PPC^{11,13,15} in various dimensions (informational, instrumental, appraisal, and emotional) and throughout the course of the disease.⁴ Informational support should be tailored to siblings in a developmentally targeted manner⁵ and include the description of the disease and possible side effects of treatment that may involve changes in the appearance and level of activity of the affected child. Most importantly, health care providers should emphasize that siblings had no role in causing the disease.⁴ Additionally, siblings may benefit from being prepared for the death of the brother/sister and from having the opportunity to say goodbye.⁵ In fact, the International Society for Paediatric Oncology guidelines for the support of siblings of children with cancer¹⁶ advise health care professionals and parents to involve siblings from the time of diagnosis, keeping them informed.^{4,11,16} As shown by Roseberg *et al.*, a period of great vulnerability seems to exist during and immediately after the illness (or death) experience. Sharing information during this time may be challenging for parents, with most seeking to protect their children from difficult information.⁵ Additionally, staff overidentification with parents' needs to protect the sibling often leads to a lack of information. Consequently, these siblings often have mistaken ideas regarding the disease,⁴ which may ultimately hamper the bereavement process,⁵ leading to unresolved grief.¹⁴ Actually, the consequences of talking to siblings about sensitive issues are likely to outweigh the costs of remaining silent.⁶ Caregivers who are apprehensive about involving siblings should be explained that being involved in the care of the ill child and having conversations about his/her general health status are generally viewed as important by the siblings themselves.⁶ In the

study by Freeman *et al.*, one of the most helpful resources identified by siblings was the ability to visit the hospitalized child when desired. According to the authors, visiting allows the sibling to directly observe how the ill brother or sister is doing medically and the type of treatment and care provided, promoting the reality of the situation and positioning the sibling to interact with health care providers for the acquisition of information. Additionally, visiting likely involves other family members, which may foster feelings of family cohesion.⁴ It is also important to be aware of how siblings engage in protective buffering. Consequently, professionals need to assess siblings' level of knowledge of the condition and its impact directly from the child, rather than from parental proxy reports, which may underestimate the impact on siblings.² Siblings also need instrumental support, especially as the disease progresses and they tend to return to their own concerns, requiring parental attention.⁴ In the study by Lövgren *et al.* (2016), more than half of siblings suggested advice related to their own need for day-to-day support from diagnosis to several years after bereavement.¹¹ Also, in the study by Wallin *et al.*, siblings advised peers to occasionally take a break from illness and death as a way of taking care of themselves.¹² Accordingly, studies in pediatric cancer setting show that minimal gestures, such as asking healthy siblings how they are doing (instead of asking about the child with cancer) or providing them with individual attention concerning their interests, may be beneficial and appreciated.⁷ It may be important to increase support for siblings from their extended family, school, and community members, by raising awareness of the situation of the healthy sibling in these groups.^{7,11} Siblings may also benefit from appraisal support, including instruction in coping strategies to deal with changes of the affected child and engagement in his/her actual care and comfort.^{4,15} With open, transparent, directive instructions on how to care for their brothers/sisters and family in general, siblings may engage in helping behavior, fulfilling their cognitive and

active coping styles.⁶ Additionally, this may in turn promote strong family cohesion, thus contributing by helping bereaved siblings to create more positive experiences with and memories of the sick sibling.¹⁵ This has been previously recommended for siblings of children with cancer, with advice for the Oncology team to include siblings in treatment, as appropriate (e.g. giving tours of the hospital ward; explaining tests, procedures, and treatments), as this may help siblings feel more included and less isolated.⁷ The need for emotional support can be addressed with support groups during hospitalization, throughout treatment⁴ and, importantly, after the child's death.⁵ Health professionals have an active role in this domain, since they must mediate hope in a realistic and honest way.¹¹ It is important to adopt a systemic approach to better understand the mutually reinforcing relationship between the family and wider environment on sibling adjustment.² This review parallels previous findings in siblings of children with cancer, where higher levels of distress were more common within two years after diagnosis, with most siblings responding well with minimal support. As in Oncology setting,⁷ it seems reasonable to recommend that those who display significant distress should be referred to evaluation and treatment by mental health care specialists.

Although seven studies included in this review refer to the Oncology setting, siblings' needs may vary substantially,⁶ as PPC patients have a great diversity of medical conditions, with very different trajectories and prognosis, making generalizations from disease-specific studies inappropriate. For example, for siblings of children with progressive LLCs, the ongoing deterioration of the child's condition requires that support be flexible enough to respond to changes, symptoms, and relationships, to provide the best care to siblings.² In fact, previous studies show that chronic illnesses with daily treatment regimens are associated with negative effects compared to chronic illnesses that do not affect daily functioning.⁸

Five of the nine studies included in this review focused on the bereavement stage. However, as previously described, PPC encompasses a far broader and earlier approach than solely bereavement support.¹ Therefore, future research should focus on the needs and concerns of siblings in PPC using an earlier and longitudinal assessment. It should also be noted that the exclusion of studies in which participants were not the siblings themselves, although informed by evidence showing that siblings' own perspectives provide beneficial augmentation to proxy reports,² may have omitted research articles concerning younger siblings - a population that requires special attention and care.

CONCLUSION

Siblings of children in palliative care have the need for information, engagement in brother/sister's care, and psychosocial support. In the future, quantitative studies of siblings' wishes may enable a more effective assessment.⁶ Prospective studies in which siblings are

interviewed as they go through the different stages of disease should be performed in order to evaluate their perspectives, experiences, and outcomes.^{4,5} Clinical practice recommendations should also be developed, taking into account the general principles of palliative care but also leaving room to include the specificities of each disease course and, most importantly, the uniqueness of each child, family, and sibling.

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