Palliative and Supportive Care

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Researching children's perspectives in pediatric palliative care: A systematic review and meta-summary of qualitative research

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Review Article

Cite this article: Ghirotto L, Busani E, Salvati M, Di Marco V, Caldarelli V, Artioli G (2019). Researching children's perspectives in pediatric palliative care: A systematic review and meta-summary of qualitative research. *Palliative and Supportive Care* **17**, 107–118. https://doi.org/10.1017/S1478951518000172

Received: 7 November 2017
Revised: 15 March 2018
Accepted: 15 March 2018

Key words:
Meta-summary; qualitative research; pediatric palliative care; children's voice; neoplasm

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Abstract

Objective. Qualitative research is pivotal in gaining understanding of individuals' experiences in pediatric palliative care. In the past few decades, the number of qualitative studies on pediatric palliative care has increased slightly, as has interest in qualitative research in this area. Nonetheless, a limited number of such studies have included the first-person perspective of children. The aim of this article is to understand the contribution of previous qualitative research on pediatric palliative care that included the voices of children.
Method. A systematic review of qualitative studies and a meta-summary were conducted. MEDLINE, CINAHL, PsycINFO, PsycARTICLES, and ERIC were searched without limitations on publication date or language. Eligible articles were qualitative research articles in which the participants were children ranging in age from 3 to 18 years.
Result. We retrieved 16 qualitative research articles reporting on 12 unique studies, and we selected two mixed-method articles. The meta-summary shows eight themes: the relationship with professional caregivers, pain and its management, "living beyond pain," the relationship between pediatric patients and their families, children's view on their treatment and service provision, meanings children give to their end-of-life situation, consequences of clinical decisions, and the relationships among children in pediatric palliative care and their peers.
Significance of results. This meta-summary presents the "state of the art" of pediatric palliative care qualitative research on children and highlights additional research areas that warrant qualitative study.

Introduction

The World Health Organization defines palliative care as "an approach to care which improves quality of life of patients and their families facing life-threatening illness through prevention, assessment, and treatment of pain and other physical, psychological, and spiritual problems" (World Health Organization, 2002). "Palliative care for children (PPC) represents a special, albeit closely related field to adult palliative care" (World Health Organization, 1998, p. 8). It incorporates "total care" that addresses the child's body, mind, and spirit (World Health Organization, 1998), and it serves as a means of providing support to the family (Morgan, 2009). PPC is "recognised to be a specialised type of care requiring specific skills and knowledge" (Steele et al., 2008, p. 229); nonetheless, healthcare professionals can rely on a paucity of research on which to base care (Committee on Palliative and End-of-Life Care for Children and Their Families, 2003; Steele et al., 2008). As noted by the Committee on Palliative and End-of-Life Care for Children and Their Families, healthcare professionals and researchers are conscious of "shortfalls in clinical research involving palliative and end-of-life care for children" (Committee on Palliative and End-of-Life Care for Children and Their Families, 2003, p. 351). Because it can access and include patients' perspectives, qualitative research (QR) can contribute (Sackett & Wennberg, 1997) to PPC quality improvement. In the past few decades, the number of qualitative studies on PPC has increased slightly (Akard, 2013), as has interest in understanding the endeavor of QR in this area. Accordingly, the present article aims to understand the contribution of QR that has included children as participants.

Methods

Search strategy

We performed a systematic review and a meta-summary. A meta-summary is a method for aggregating findings that appear in exclusively qualitative reports and is suitable for

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https://doi.org/10.1017/S1478951518000172 Published online by Cambridge University Press

acknowledging the "preponderance of evidence" (Thorne et al., 2004) in QR and highlighting what the qualitative results reveal. We used both Preferred Reporting Items for Systematic Reviews and Meta-Analysis (for tutoring in retrieving papers (Moher et al., 2009) and the Enhancing Transparency in Reporting the Synthesis of Qualitative Research guidelines (Tong et al., 2012) to report the results.

We searched for QR articles on PPC involving children ranging in age from 3 to 18 years in the electronic databases MEDLINE, CINAHL, PsycINFO, PsycARTICLES, and ERIC, with no limitations on publication date or language. A generic search was also performed using Google Scholar, ResearchGate, and Academia.edu. We also performed a manual search using the references of systematic reviews we retrieved, and we included articles for both cross-checking and identifying other potential QR articles. We performed a systematic review from July 2016 to December 2016. We discussed the search strategy and agreed on using keywords from three primary domains: PPC-related terms, AND participant-related, AND QR keywords. We summarize the search terms in Table 1. We managed to work collaboratively using an online platform (i.e., Google Drive) to share progress regularly.

Search outcomes

We retrieved a total of 1,240 articles by searching the databases and checking the articles' references. We used an Excel file and inserted all titles and authors' names. We manually removed duplicates ($n = 504$) and then reviewed 736 articles by title. The articles had to meet the following inclusion criteria: report of a QR and study conducted with children ranging in age from 3 to 18 years and experiencing PPC. We eliminated 324 articles that were considered irrelevant according to their titles because they did not mention any methodology related to QR. There were 340 articles that, according to their abstracts, did not meet the inclusion criteria (i.e., QR articles involving children experiencing PPC and ranging in age from 3 to 18 years). LG and GA retrieved the full texts and shared the articles using the online platform. LG proposed an assessment framework to describe the aims, research questions, method, and ethical issues and to evaluate the inclusion/exclusion for each full text. We wrote a file for each article and uploaded the files using the online platform for sharing. We were able to check the full text of 72 articles, of which 56 were excluded because they did not meet the inclusion criteria. A total of 14 QR articles reporting on 12 unique studies and two mixed-method articles were selected. The Preferred Reporting Items for Systematic Reviews and Meta-Analysis flow chart of the search process is shown in Figure 1.

Table 1. Search domains and keywords

Domain	Key words
PPC	palliative care, early palliative care, simultaneous care, life threaten, chronic illness, disease, end of life, cancer, neoplasm, oncolog*, pain, pain management
Participants' type	pediatric, infant, child*, teen*, adolescent, young, youth
QR	qualitat*, qualitative research, interview*, observation, discourse, narrative, grounded theory, phenomenol*, ethno*, focus group

PPC, palliative care for children; QR, qualitative research.

Critical appraisal of the studies

Three authors (EB, MS, and LG) used Critical Appraisal Skills Programme (CASP, 2013) to highlight methodological strengths and weaknesses of the selected studies.

Meta-summary

LG, EB., MS, and VDM extracted the results/findings sections in each included study. All the text under the "results/findings" sections was extracted manually and entered in a table. LG performed the thematic analysis of findings to identify overarching themes (Sandelowski & Barroso, 2003; Sandelowski et al., 2007). The thematic techniques included (1) line-by-line coding, (2) the reduction of these codes into themes, and (3) calculation of the frequency (Sandelowski & Barroso, 2003), as shown in Table 4. The main topics were discussed among all the authors. GA and VC detailed the final version of the themes.

Results

We found 16 qualitative studies addressing children's experience of PPC. We report the studies' characteristics in Table 2.

Using the CASP, we found seven articles of high/moderate-to-high quality, in which authors openly discuss the research design and methods along with the methodological steps they have followed. There are three articles of moderate/moderate-to-low quality, as the description of the method lacks essential information (i.e., internal consistency, interview guide, data saturation). The main weaknesses of studies we assessed as low quality regard the ethical considerations and the data analysis process, which are not explicitly accounted. The appraisal is shown in Table 3.

The entire sample of the retrieved studies involved 212 children. For 105 children, the studies lacked detailed age-related information because the authors had indicated large age ranges, whereas, for the remaining children, the developmental stage can be sketched as follows: 29 preschoolers, 48 primary schoolers, and 30 teenagers. The reported conditions of pediatric participants were oncologic or hematologic diseases ($n = 60$), nononcological illnesses ($n = 63$), and unspecific life-limiting or life-threatening situations ($n = 89$).

The participating children were recruited within the services they attended and were mainly involved as interviewees in 10 of 16 articles. Additional data were derived from ethnographic observations (two articles), a focus group, a questionnaire with several free-response questions, a play-based workshop in action-research, and a personal diary.

Meta-summary results

We concentrated the analysis for the meta-summary on 14 articles because three articles refer to the same study. The QR with PPC children focuses on eight major themes, which are listed in Table 4.

Relationship with professional caregivers

The majority of QR with pediatric patients in palliative care (Amery et al., 2009; Carter, Edwards & Hunt, 2015; Coad et al.,

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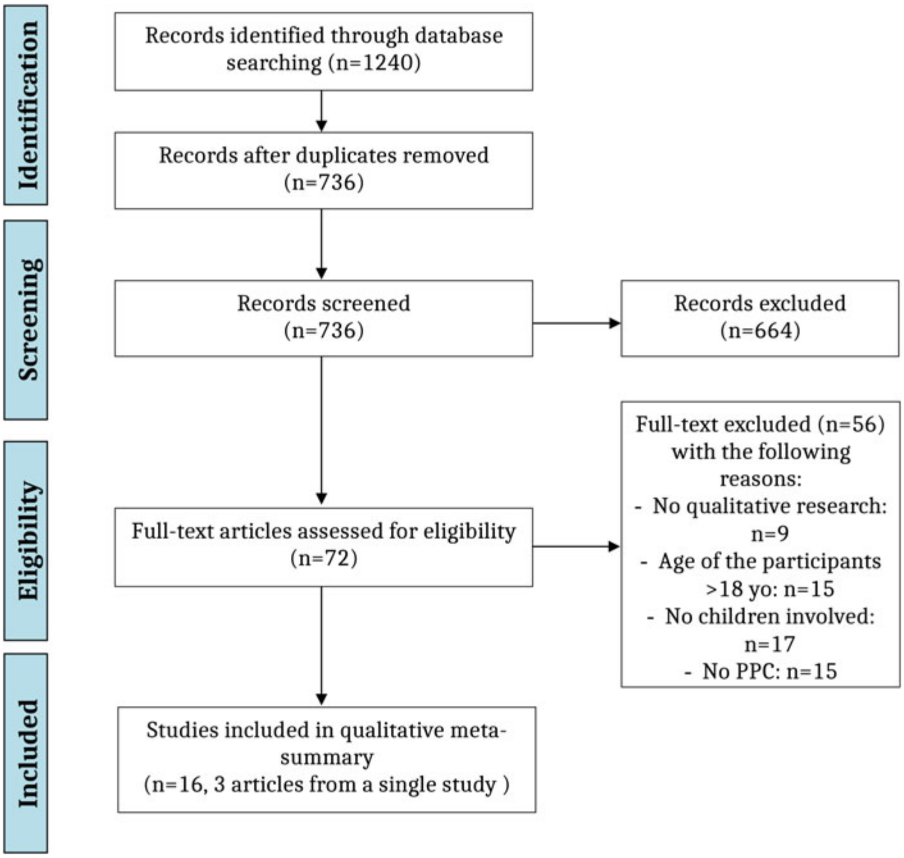

Fig. 1. Qualitative Preferred Reporting Items for Systematic Reviews and Meta-Analysis.

2015; Flavelle, 2011; Hsiao et al., 2007; Spalding & Yardley, 2016; Swallow, Forrester & Macfadyen, 2012) describes the relationship with professional caregivers. This theme includes communication between children and doctors (Carter et al., 2015, Coad et al., 2015; Hsiao et al., 2007); the bond with family support workers (Carter et al., 2015); a reflection of the relationship between the home nurse and Ed, who is the protagonist in the phenomenological case study (Flavelle, 2011); and relations with professionals from a short-break service (Swallow et al., 2012) and a hospice (Amery et al., 2009).

Pain and pain management

Pain and pain management (Borghi et al., 2014; Flavelle, 2011; Kortesluoma et al., 2008; Kortesluoma & Nikkonen, 2004, 2006; Tamannai et al., 2015) constitute another dominant theme. Living with pain has an impact on self-image (Borghi et al., 2014) and may produce fatigue (Flavelle, 2011). Children can apply self-help strategies for pain (Borghi et al., 2014; Kortesluoma et al., 2008) such as physical actions (i.e., massage, caressing, deep breathing, moving or changing the body posture, application of heat or cold, resting and relaxation, immobilization or exercise, eating and/or drinking, loosening tight clothes, and defecation) (Kortesluoma et al., 2008). These actions offer them a sense of control over the pain. Besides, children follow what Kortesluoma et al. (2008) interpreted as cognitive and behavioral actions as they learn about what techniques may increase or reduce pain. Children can give meaning to their illness (Kortesluoma & Nikkonen, 2006) and can distinguish different pains and their origins (Tamannai et al., 2015), such as pain caused by medical and diagnostic procedures and basic nursing (Borghi et al., 2014; Kortesluoma & Nikkonen, 2004) and pain caused by accidents occurring in everyday activities. Children also experience pain they find inexplicable (Flavelle, 2011; Kortesluoma & Nikkonen, 2004), such as pain caused by aggressive actions of other people (Kortesluoma & Nikkonen, 2004). Kortesluoma and Nikkonen (2004) reported that one of the most intense types of pain children experienced was the physical pain and emotional suffering caused by being hit or verbally abused.

"Living beyond pain"

We titled this theme "living beyond pain" to indicate what children experiencing PPC consider important for them despite the pain and the disease (Borghi et al., 2014; Davies et al., 2005). This theme emerges from six articles (Amery et al., 2009; Borghi et al., 2014; Carter et al., 2015; Davies et al., 2005; Flavelle, 2011; Swallow et al., 2012). Children need to be engaged in activities such as sports and individual play (Borghi et al., 2014; Carter et al., 2015), video games, which authors consider an escape from illness (Flavelle, 2011), or group play (Amery et al., 2009; Carter et al., 2015; Davies et al., 2005). Playing is seen as a way to forget the disease for a while (Amery et al., 2009; Flavelle, 2011). In an assessment of a short-break service for children with life-limiting conditions (Swallow et al., 2012), teenagers appreciated better access to leisure activities (i.e., hide and seek, treasure hunts, playing on the computer/the Internet, singing and listening to music) compared with being at home.

https://doi.org/10.1017/S1478951518000172 Published online by Cambridge University Press

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Table 2. Studies' characteristics

First author (date) country	Aims	Study design	Sample (children's age)	Disease	Sampling strategy	Inclusion criteria	Data collection method(s)	Data analysis strategy
Spalding (2016) UK	– Explore perceptions of what medical students must learn to become "good doctors" among children, parents and staff in a hospice – Collaborate with children/parents and staff to develop educational materials based on their lived experiences for medical students – Assess feasibility of student-led action research in a children's hospice to develop research skills	Action research study	16 participants: 6 professionals, 3 parents, 7 children (3 children aged 8–12 years; 4 children aged 12–14 years)	Life-threatening/limiting conditions	Selective	– All children aged 8–19 years who had used hospice services in the past 12 months – Verbal communication skills and ability for basic understanding of the study aims	– Audiorecorded semistructured focus group (professionals) – Audiorecorded individual interviews (parents) – Two interactive multimedia workshops facilitated with a play specialist (children) – Research diary	Initial thematic framework
Carter (2015) UK	– Explore key aspects of the work of Family Support Workers in caring and supporting families from the families' perspectives	Range of methods, mainly qualitative	– 55 families (of which 12 bereaved) – 39 children (aged 2–18 years)	Life-limiting and disabling conditions	Purposive	Child having complex, life-threatening or terminal conditions receiving care from Family Support Workers	– Surveys – Interviews – Ethnographic observation	Thematic analysis
Tamannai (2015) Cameroon	– Gain a better understanding of the needs of Burkitt lymphoma patients and their families – Assess their perception of the PC outreach program – Identify issues that could be improved to better meet their needs	Qualitative study	12 participants among which 3 children (1 girl age 14 years, 1 girl aged 9 years, and 1 boy aged 10 years)	Burkitt lymphoma	Not declared	All patients and their caregivers who had been visited by the children's PC outreach program between 5 October and 29 November 2013	Semistructured interviews with open-ended questions	Thematic analysis
Borghi (2014) Brazil	– Understanding how children and adolescents under	Qualitative study	6 children (2 aged 6 years, 2 aged 13	– Epidermolysis bullosa – Osteogenesis	Purposive		Semistructured interviews	Text completely recreated, into 6 narratives

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	palliative care manage pain in their daily lives – How they describe its intensity and quality		years, and 2 aged 17 years)	imperfecta – Xeroderma pigmentosum – Human T-cell lymphotropic virus – Hereditary spastic paraparesis		– Children and adolescents from 6 to 17 years; – Agreed to participate in the interview – Chronic diseases, with no possibility of cure – Under palliative care – Receiving pain treatment		analyzed with Piaget's theory of cognitive development
Coad et al. (2015) UK	Answer the question: "How well are the palliative care needs of children with life-limiting and life-threatening conditions and their families met by services?"	Appreciative inquiry	– 59 adults – 8 children and young people (aged 0–25 years)	Life-threatening/ life-limiting conditions	Purposive	– Children and young people up to 25 years – Diagnosis of a life-limiting/life-threatening condition – Receiving health care – In receipt education at schools serving needs up to 18 years	– Semistructured interviews – in-depth interviews	Framework analysis
Gaab (2013) New Zealand	– Child-focused research – Describe self-identified factors that affect 9–18 years patients and their siblings during the process of receiving PPC	Ethnographic study	– 7 children patients – 3 brothers – 6 sisters (participants aged 9–18 years)	– Cancer – Heart conditions – Dystrophies	Not declared	Participants attending PPC facilities	– Diaries – Recordings	Inductive thematic analysis
Noyes (2013) UK	– Develop and evaluate the "My choices booklets" for use by parents and children – Facilitate thinking and engagement with future care planning	Qualitative study	– 15 parents – 10 professionals – 11 children (aged 6–15 years)	Life-threatening/ life-limiting conditions	Convenience	– Children involved in PPC at National Health Service, social services, and not-for-profit organizations – Multiagency palliative care professionals	– Interviews (parents, children, professionals) – Questionnaires (professionals) – An open web-based consultation	– Qualitative interviews: framework analysis; – Descriptive questionnaire: descriptive statistics using SPSS – Web- based feedback: summarized using survey monkey
Flavelle (2011) Canada	Provide a unified description of an adolescent's experience of living with and dying of cancer	Qualitative analysis using phenomenological methods (case study)	1 adolescent (aged 15 years)	Cancer-osteosarcoma	"Before Ed died, he graciously offered his journal for the benefit of others. He	Not applicable	Analysis of his 90-page journal that spanned 3 months just before death	– Line-by-line approach by Van Manen

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					clearly stated in his journal that it should be made available for ‘anyone who wants to read’” [27] (p28)			
Swallow et al. (2012) UK	Determine teenagers’ and parents’ views of a short-break service for children with life-limiting conditions	Qualitative study	– 20 parents – 5 teenagers (aged 14–18 years).	Life-limiting condition	Convenience	Teenagers and parents using hospice service	– 5 individual interviews – 4 focus groups with parents – 2 focus groups with teenagers	Framework analysis
Amery (2009) Uganda	Evaluate a children’s palliative care service designed specifically for a resource-poor sub-Saharan African setting	Mixed quantitative and qualitative methodology: quantitative, retrospective, comparative survey and cross-sectional, noninterventional survey	– 11 children (unreported age) – 12 parents – 10 hospice and hospital professionals	Cancer	Convenience	– Children and parents using hospice service or attending hospital cancer wards – Hospice and hospital professionals at research settings	Semistructured interviews	Thematic analysis
Kortesluoma (2008) Kortesluoma (2006) Kortesluoma (2004) Finland	– Describe interventions young children use themselves – Describe their expectations of other’s help, when managing the pain experienced during hospitalization – Identify the children’s preferences and expectations – Gather children’s descriptions of their experiences of pain and views regarding the purpose of pain – Increase understanding of the phenomenon – Describe the pain experiences of hospitalized children	Qualitative study	44 children (aged 4 to 11 years)	– Problems of locomotive system; – Internal-surgical problems – Tumors – Hematologic diseases – Diabetes – Burns	Purposive	The inclusion criteria expected the child to have an ailment or disease that caused hospitalization and that the child had complained of pain during hospitalization	Interviews with open-ended questions	Inductive qualitative content analysis

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Hsiao (2007) USA	Identify the aspects of physician communication that children with life-limiting illnesses and their parents perceived to be facilitative or obstructive in pediatric palliative care	Qualitative study (part of a larger mix-methods study)	– 20 children (aged 9–21 years) – 20 parents	– Oncological diseases – Cardiac issues	Not declared	– Children with life-limiting conditions having a <20% chance of survival beyond 3 years – English speakers – Mentally competent parents	Semistructured interviews	Grounded theory approach
Davies (2005) Canada	– Assess what children find helpful or what they think about care facilities – Grasp ill children and siblings voices – Contribute to the knowledge of pediatric palliative care; – enable health care providers to develop optimal palliative care programs.	Qualitative study based on the principles of participatory an action research	– 36 parents; – 30 ill children (aged 6 to 19 years); – 51 siblings (aged 3–19 years)	Life-threatening conditions	Randomly selected	Children in pediatric palliative care programs	– Face-to-ace interview (36 parents, 4 ill children, and 10 siblings) – Questionnaires (26 ill children and 41 siblings) with several free-response questions	Inductive thematic analysis
Hinds (2005) Australia	– Identify the preferences of children and adolescents with advanced cancer about their end-of-life care – Identify the factors that influenced their decisions	Descriptive study	– 20 children (aged 10–20 years) – 19 parents – 14 physicians	– Refractory solid tumor – Brain tumor – Leukemia	Purposive	– English speakers – Patients between age 10 and 20 years, directly involved in the end-of-life decision – Consent to participate	Open-ended interviews	Thematic analysis with code dictionary

PC, palliative care.

Table 3. Critical appraisal of the included articles

First author (date), country	Strengths (+) and weaknesses (-)	Quality appraisal (CASP)
Spalding (2016) UK	+ Relevance of the research + Listening to children, parents, and hospice staff – Action-research approach is not clearly developed (i.e., research questions and aims are not consistent with the approach) – Research design and methods are not properly discussed + Recruitment is well documented – Researchers did not clearly define how they have reached data saturation – Relationship between researcher and participants, especially children, has not been adequately considered + Each child's agreement was requested, over parent's consent – No focus group and interview guide provided – How thematic analysis was conducted is insufficiently described – There is no explicit description about how triangulation or combination of the different type of data was performed	Moderate-to-low
Carter (2015) UK	+ Relevance of the research + Research design is appropriate + Ethical consideration is well described + The findings are explicit and discussed – Recruitment strategy is not documented	High
Tamannai (2015) Cameroon	+ Relevance of the research + Research design is appropriate – Ethical consideration is not well described – The examined sample is small – The data analysis is not sufficiently described	Low
Borghi (2014) Brazil	+ Children approved the final text before analysis – No children assents requested – Piaget's theory reduced and narrowed data interpretation – participants' quotations are inconsistent with the themes	Low
Coad et al. (2015) UK	+ Relevance of the research + Listening to children, parents and hospice staff + Research design and methods are properly discussed + The recruitment strategy is appropriate to the aim of the research + The inclusion and exclusion criteria in the study are expressed clearly + Ethical consideration is well described – Relationship between researcher and participants, especially children, has not been adequately considered – The researcher has not discussed the credibility of their findings (respondent validation)	Moderate-to-high
Gaab (2013) New Zealand	+ Relevance of the research + Research design is appropriate + The adopted research methodology is explained – Ethical consideration is not well described + The data analysis is sufficiently described + The recruitment strategy is documented	High
Noyes (2013) UK	+ Relevance of the research + Research design is appropriate + The findings are well described and discussed + Ethical consideration is not well described + New areas of research are identified	High
Flavelle (2011) Canada	+ A single case study can provide valuable information in a field such as pediatric palliative care in which the patient's perspective may be difficult to access or ascertain – Anyhow transferability is not assessed as it is a single case + Relevance of the research + Research design is appropriate + Ethical consideration is well described	Moderate-to-high

(*Continued*)

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Table 3. (*Continued.*)

First author (date), country	Strengths (+) and weaknesses (-)	Quality appraisal (CASP)
Swallow et al. (2012) UK	+ Relevance of the research + Listening to teenagers with life-limiting condition and parental consideration – Small sample – Action-research approach is not clearly developed (i.e., research question is not consistent with the approach) + Research design and methods are properly discussed + Recruitment is well documented – Researchers did not clearly define how they have reached data saturation – Relationship between researcher and participants, especially teenagers, has not been adequately considered + Each teenager's agreement was requested, over parent's consent + Focus group and interview guide provided – How thematic analysis was conducted is insufficiently described – There is no explicit description about how triangulation or combination of the different type of data was performed	Moderate
Amery (2009) Uganda	+ The statement of the aim of the research is clear + Listening to children, parents and hospice staff + The qualitative methodology is appropriate – Research design and methods are not properly discussed – Recruitment is not well documented – Relationship between researcher and participants, especially children, has not been adequately considered + Each child's agreement was requested, over parent's consent + The ethical issues have been taken into consideration – No focus group and interview guide provide + The findings are explicit	Moderate
Kortesluoma (2008) Kortesluoma (2006) Kortesluoma (2004) Finland	+ Relevance of the research + Research design is appropriate + Ethical consideration is well described + The findings are explicit and discussed – It was only examined a sample hospital	High
Hsiao (2007) USA	+ Relevance of the research – Ethical considerations are not well described – Validity is not discussed – Small sample	Low
Davies (2005) Canada	+ Relevance of the research + Research design is appropriate – Recruitment strategy is not documented – The data analysis is not described and is not clearly – Ethical consideration is not well described	Low
Hinds (2005) Australia	+ Relevance of the research + Research design is appropriate + The setting is appropriate + The inclusion and exclusion criteria in the study are expressed clearly + The ethical consideration is described + The data analysis is sufficiently described	High

Relationship with the family

The relationship between pediatric patients and their families was mentioned in six qualitative studies (Borghi et al., 2014; Flavelle, 2011; Gaab et al., 2013; Hinds et al., 2005; Kortesluoma et al., 2008; Kortesluoma & Nikkonen, 2006). The relationship with the family is interlinked with the relationship with healthcare professionals and peers (Kortesluoma et al., 2008). Children describe their family as the primary source of support (Borghi et al., 2014; Gaab et al., 2013; Hinds et al., 2005). Studies (Flavelle, 2011; Gaab et al., 2013; Kortesluoma & Nikkonen, 2006) have found that the diagnosis affects the relationships among family members. Children often note that their feelings have changed from before the diagnosis (Gaab et al., 2013; Hinds et al., 2005). On the one hand, the clinical situation makes them feel closer to their parents, whereas, on the other hand, it augments children's dependency on family members (Flavelle, 2011; Gaab et al., 2013).

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Table 4. Meta-summary: themes and subthemes

Themes	Subthemes	Articles	Frequency
Relationship with professional caregivers	Communication with doctors	Coad et al., 2015; Hsiao et al., 2007; Spalding & Yardley, 2016	(7/16) 43.75%
	Connection with family support workers	Spalding & Yardley, 2016	
	Home nurse	Flavelle, 2011	
	Short-break service staff	Swallow et al., 2012	
	Hospice staff	Amery et al., 2009	
Pain and pain management	Impact on self-image	Borghi et al., 2014	(6/16) 37.50%
	Fatigue	Flavelle, 2011	
	Strategies	Kortesluoma et al., 2008	
	Discrimination of pains	Borghi et al., 2014; Flavelle, 2011; Kortesluoma & Nikkonen, 2006; Kortesluoma & Nikkonen, 2004; Tamannai et al., 2015	
"Living beyond pain"	Sports and individual play	Borghi et al., 2014; Carter et al., 2015;	(6/16) 37.50%
	Video game	Flavelle, 2011	
	Group play and leisure activities	Amery et al., 2009; Carter et al., 2015; Davies et al., 2005; Swallow et al., 2012	
Relationship with the family	Source of support	Borghi et al., 2014; Gaab et al., 2013; Hinds et al., 2005; Kortesluoma et al., 2008	(6/16) 37.50%
	Relationship change	Flavelle, 2011; Gaab et al., 2013; Hinds et al., 2005; Kortesluoma & Nikkonen, 2006	
	Dependency enhancement	Gaab, Owens & MacLeod, 2013; Flavelle, 2011	
Treatment	Comprehension	Flavelle, 2011; Hinds et al., 2005; Kortesluoma & Nikkonen, 2004; Tamannai et al., 2015	(4/16) 25%
Evaluation of the service provision	Future care planning tool	Noyes et al., 2013	(4/16) 25%
	The whole service	Amery et al., 2009; Davies et al., 2005; Swallow et al., 2012	
Death and consequences of end-of-life decisions	"Having limited time"	Flavelle, 2011; Gaab et al., 2013	(4/16) 25%
	Consequences	Hinds et al., 2005; Kortesluoma & Nikkonen, 2004	
Relationship with peers	Communication with peers	Borghi et al., 2014; Gaab et al., 2013; Swallow et al., 2012	(3/16) 18.75%
	Feeling understood/ discriminated	Gaab et al., 2013	

Treatment

A fifth theme is the view children have regarding treatment (Flavelle, 2011; Hinds et al., 2005; Kortesluoma & Nikkonen, 2004; Tamannai et al., 2015). Children feel and comprehend both the adverse effects of medications (Flavelle, 2011) and the benefit of pain relief medications (Tamannai et al., 2015). Children are aware of the effects treatment may induce (Kortesluoma & Nikkonen, 2004). Moreover, children can recall all of their past treatment options (Hinds et al., 2005).

Evaluation of the service provision

Another theme emerging from the articles is the children's evaluation of service provision (Amery et al., 2009; Davies et al., 2005; Noyes et al., 2013; Swallow et al., 2012). A tool for future care planning has been assessed by children, parents, and healthcare professionals (Noyes et al., 2013). Similarly, children and parents had the opportunity to evaluate the equipment and, generally, the atmosphere of a short-break service (Swallow et al., 2012), the strengths and weaknesses of children's palliative care service in Africa, including staff attitudes (Amery et al., 2009), and a Canadian free-standing hospice programme (Davies et al., 2005).

Death and consequences of end-of-life decisions

The meanings children give to their end-of-life situation (Flavelle, 2011; Gaab et al., 2013) and the perception of consequences that the decisions or treatments may bring (Hinds et al., 2005; Kortesluoma & Nikkonen, 2004) emerged as a theme. Children mentioned death and indicated that they were aware of having limited time (Gaab et al., 2013). Ed wrote about the end in a humorous way (Flavelle, 2011). A child affirmed that he knew he would die if he lost blood (Kortesluoma & Nikkonen, 2004); other patients identified their death as an outcome of their decision (i.e., getting sick from an experimental medication, delaying death, and dying) (Hinds et al., 2005).

Relationship with peers

The last theme addressed in QR with PPC patients involves peer relationships (Borghi et al., 2014; Gaab et al., 2013;

https://doi.org/10.1017/S1478951518000172 Published online by Cambridge University Press

Swallow et al., 2012). It includes critical communication with friends (Borghi et al., 2014), the desire and the difficulty of speaking and having a "normal" conversation with healthy friends (Gaab et al., 2013) and the ability and willingness to talk freely with peers who share the same condition (Swallow et al., 2012). If allowed to confront others, children are more likely to feel understood (Gaab et al., 2013). Nonetheless, relationships with others may cause feelings of discrimination (Gaab et al., 2013).

Discussion

A meta-summary of findings was conducted to understand the contribution of QR on PPC in which the voices of patients were solicited. We retrieved 16 qualitative studies concerning eight major themes (and 22 related subthemes). The relationship with professional caregivers, pain and its management, "living beyond pain," and the relationship between pediatric patients and their families are dominant themes in the literature. These themes are consistent with research on children's perspectives regarding their treatment and service provision. Finally, four studies discussed the meanings children give to their end-of-life situation and the consequences of clinical decisions, whereas three articles mentioned the relationships children in PPC have with their peers.

According to this systematic review and the meta-summary results, the voice of children, when they are able to talk and express themselves, appear to be misrepresented within the scientific panorama (Akard, 2013; Hinds, et al., 2007). Emerging PPC research priorities (Baker et al., 2015; Steele et al., 2008) would be adequately met by conducting more QR "with" (and not only "on") pediatric patients (Committee on Palliative and End-of-Life Care for Children and Their Families, 2003; Stevenson et al., 2013). Further QR is needed to investigate what matters most for children receiving PPC (Snaman et al., 2018; Steele et al., 2008), assess evidence-based practice guidelines in PPC (Baker et al., 2015), and comprehend the children's role in making treatment decisions about their palliative and end-of-life care (Baker et al., 2015). A recent example of dealing qualitatively with those emerging priorities is a study protocol about specialized outpatient palliative care, which has been conceived to provide information through qualitative narrative interviews with pediatric patients on what matters most to them receiving specialized outpatient palliative care (Ulrich et al., 2018).

Accessing the voices of children through QR may also contribute to helping parents communicate with their child about the child's life-threatening illness and the likelihood of death (Baker et al., 2015). These research implications are consistent with recently published articles (Badarau et al., 2017; Snaman et al., 2018; Weaver et al., 2016), which list topics of urgent consideration. Among them, effective communication (Snaman et al., 2018), and shared decision-making (Badarau et al., 2017; Weaver et al., 2015) are pressing subjects.

We also recommend that researchers and clinicians conduct QR about how to take advantage of children's spontaneous pain management to define its effectiveness, how the relationship with professional caregivers can make important differences in the provision of PPC, the role of play and leisure for children experiencing PPC, the way intrafamiliar relationships change during the care process (Snaman et al., 2018) and how to address the interdependency among PPC patients and family members.

Strengths and limitations

Among the limitations of the systematic review, we note that, although we followed a rigorous process in retrieving the articles, articles may have been missed. To address this limitation, we searched five comprehensive databases and performed a generic search using academic social networks (i.e., Google Scholar, ResearchGate, and Academia.edu) in addition to checking references in articles. The systematic review was limited to articles from peer-reviewed journals and those that were published in English. Many studies appear in different languages and in non–peer-reviewed journals (i.e., dissertations, doctoral theses, and gray literature), which we have not included. We understandably did not include qualitative PPC articles investigating the experience of children who were not able to communicate for themselves; therefore, we attempted to avoid bias in reviewing and appraising the studies using more than two members in each of the review stages, and the online platform that we used helped the team in cross-validation and checking tutoring. Regarding the meta-summary, we decided not to differentiate the findings according to children's age because of the scarceness of the number of items and detailed age-related information in most of the retrieved articles. Nonetheless, we are aware that children have particular physical, cognitive, and social development trajectories that change over time; therefore, when the QR body is increased, there will be a need to distinguish children according to age and related needs. Thus, we performed a thematic analysis that led us to collect themes from the included qualitative studies. Although the themes were discussed and validated by all of the authors, they might not represent all possible interpretations. To the best of our knowledge, this study is the first meta-summary of the current contribution of QR on PPC in which the voices of children were solicited and considered.

Conclusion

Conducting this systematic review and meta-summary revealed that QR with children about their experience in PPC is an emerging and promising field. We support the need for more QR, especially regarding the research relaunches we highlighted. It is our understanding that conducting QR with children in PPC will be pivotal for comprehending patients' psychological, social, and spiritual needs (World Health Organization, 2002) and for improving PPC interventions and, consequently, children's quality of life (Huang et al., 2010; O'Quinn & Giambra, 2014). In fact, QR findings may make the first-person perspective on PPC emerge and concur with respect to incorporating "total care" that addresses the child's body, mind, and spirit (World Health Organization, 2002).

Acknowledgments. The authors received no financial support for the research, authorship, and publication of this article.

LG and GA conceived and designed the systematic review and the meta-summary. All authors discussed the search strategy and agreed on final terms list for the search. All authors revised the article critically and approved the release sent to the journal.

Conflicts of interest. None.

https://doi.org/10.1017/S1478951518000172 Published online by Cambridge University Press

References

Akard TF *et al.* (2013) From qualitative work to intervention development in pediatric oncology palliative care research. *Journal of Pediatric Oncology Nursing* **30**(3), 153–160.

Amery JM *et al.* (2009) The beginnings of children's palliative care in Africa: Evaluation of a children's palliative care service in Africa. *Journal of Palliative Medicine* **12**(11), 1015–1021.

Badarau DO *et al.* (2017) Decision making in pediatric oncology: Views of parents and physicians in two European countries. *AJOB Empirical Bioethics* **8**(1), 21–31.

Baker JN *et al.* (2015). Research priorities in pediatric palliative care. *The Journal of Pediatrics* **167**(2), 467–470.

Borghi CA *et al.* (2014) Living with pain: the experience of children and adolescents in palliative care. *Revista da Escola de Enfermagem* **48**, 67–73.

Carter B, Edwards M, and Hun A (2015) 'Being a presence': The ways in which family support workers encompass, embrace, befriend, accompany and endure with families of life-limited children. *Journal of Child Health Care* **19**, 304–319.

Coad J *et al.* (2015) Exploring the perceived met and unmet need of life-limited children, young people and families. *Journal of Pediatric Nursing* **30**(1), 45–53.

Committee on Palliative and End-of-Life Care for Children and Their Families, Board on Health Sciences Policy (2003) *When children die: Improving palliative and end-of-life care for children and their families.* Washington DC: The National Academies Press.

Critical Appraisal Skills Programme (CASP) (2013) *Qualitative research checklist*. Available from: http://media.wix.com/ugd/dded87_951541699e9edc71ce66c9bac4734c69.pdf (accessed 26 July 2017).

Davies B *et al.* (2005) Children's perspectives of a pediatric hospice program. *Journal of Palliative Care* **21**(4), 252–261.

Flavelle SC (2011) Experience of an adolescent living with and dying of cancer. *Archives of Pediatrics & Adolescent Medicine* **165**(1), 28–32.

Gaab EM, Owens RG, and MacLeod RD (2013) The voices of young New Zealanders involved in pediatric palliative care. *Journal of Palliative Care* **29**(3), 186–192.

Hinds PS *et al.* (2007) Patient-reported outcomes in end-of-life research in pediatric oncology. *Journal of Pediatric Psychology* **32**(9), 1079–1088.

Hinds PS *et al.* (2005) End-of-life care preferences of pediatric patients with cancer. *Journal of Clinical Oncology* **23**(36), 9146–9154.

Hsiao JL, Evan EE, and Zeltzer LK (2007) Parent and child perspectives on physician communication in pediatric palliative care. *Palliative and Supportive Care* **5**(4), 355–365.

Huang I-C *et al.* (2010) Measuring quality of life in pediatric palliative care: Challenges and potential solutions. *Palliative Medicine* **24**(2), 175–182.

Kortesluoma RL and Nikkonen M (2006) 'The most disgusting ever': children's pain descriptions and views of the purpose of pain. *Journal of Child Health Care* **10**(3), 213–227.

Kortesluoma RL and Nikkonen M (2004) 'I had this horrible pain': The sources and causes of pain experiences in 4- to 11-year-old hospitalized children. *Journal of Child Health Care* **8**(3), 210–231.

Kortesluoma RL, Nikkonen M, and Serlo W (2008) "You just have to make the pain go away"--children's experiences of pain management. *Pain Management Nursing* **9**(4), 143–149.

Moher D *et al.* (2009) Preferred reporting items for systematic reviews and meta-analyses: The PRISMA Statement. *PLOS Medicine* **6**(7), e1000097.

Morgan D (2009) Caring for dying children: Assessing the needs of the pediatric palliative care nurse. *Journal of Pediatric Nursing* **35**(2), 86–90.

Noyes J *et al.* (2013) Planning ahead with children with life-limiting conditions and their families: development, implementation and evaluation of "My Choices." *BMC Palliative Care* **12**, 5.

O'Quinn LP and Giambra BK (2014) Evidence of improved quality of life with pediatric palliative care. *Journal of Pediatric Nursing* **40**(6), 284–288.

Sackett DL and Wennberg JE (1997) Choosing the best research design for each question. *British Medical Journal* **315**(7123), 1636.

Sandelowski M and Barroso J (2003). Creating metasummaries of qualitative findings. *Nursing Research* **52**(4), 226–233.

Sandelowski M, Barroso J, and Voils CI (2007) Using qualitative metasummary to synthesize qualitative and quantitative descriptive findings. *Research in Nursing & Health* **30**(1), 99–111.

Snaman JM *et al.* (2018) Pediatric palliative oncology: The state of the science and art of caring for children with cancer. *Current Opinion in Pediatrics* **30** (1), 40–48.

Spalding J and Yardley S (2016) 'The nice thing about doctors is that you can sometimes get a day off school': An action research study to bring lived experiences from children, parents and hospice staff into medical students' preparation for practice. *BMJ Supportive & Palliative Care* **6**, 459–464.

Steele R *et al.* (2008) Research priorities in pediatric palliative care: A Delphi study. *Journal of Palliative Care* **24**(4), 229–239.

Stevenson M, Achille M, and Lugasi T (2013) Pediatric palliative care in Canada and the United States: A qualitative metasummary of the needs of patients and families. *Journal of Palliative Medicine* **16**(4), 566–577.

Swallow V, Forrester T, and Macfadyen A (2012) Teenagers' and parents' views on a short-break service for children with life-limiting conditions: A qualitative study. *Palliative Medicine* **26**(3), 257–267.

Tamannai M *et al.* (2015) An evaluation of a palliative care outreach programme for children with Burkitt lymphoma in rural Cameroon. *International Journal of Palliative Nursing* **21**(7), 331–337.

Thorne S *et al.* (2004) Qualitative metasynthesis: Reflections on methodological orientation and ideological agenda. *Qualitative Health Research* **14**(10), 1342–1365.

Tong A *et al.* (2012) Enhancing transparency in reporting the synthesis of qualitative research: ENTREQ. *BMC Medical Research Methodology* **12**, 181.

Ulrich L-R *et al.* (2018) Study protocol: evaluation of specialized outpatient palliative care (SOPC) in the German state of Hesse (ELSAH study) – work package II: Plliative care for pediatric patients. *BMC Palliative Care* **17**(1), 14.

Weaver MS *et al.* (2016) Establishing psychosocial palliative care standards for children and adolescents with cancer and their families: An integrative review. *Palliative Medicine* **30**(3), 212–223.

Weaver MS *et al.* (2015) Palliative care as a standard of care in pediatric oncology. *Pediatric Blood & Cancer* **62**, S829–S833.

World Health Organization (1998) *Cancer pain relief and palliative care in children*. Geneva: WHO.

World Health Organization (2002) *WHO definition of palliative care*. www.who.int/cancer/palliative/definition/en (accessed 29 September 2017).

https://doi.org/10.1017/S1478951518000172 Published online by Cambridge University Press