Instruments to Measure Outcomes in Pediatric Palliative Care: A Systematic Review

Marie Friedel, PNP, MPH,[a,b] Isabelle Aujoulat, PhD,[a] Anne-Catherine Dubois, PNP, MPH,[a] Jean-Marie Degryse, MD, PhD[a,c]

abstract

CONTEXT: Pediatric palliative care (PPC) is intended to promote children's quality of life by using a family-centered approach. However, the measurement of this multidimensional outcome remains challenging.

OBJECTIVE: To review the instruments used to assess the impact of PPC interventions.

DATA SOURCES: Five databases (Embase, Scopus, The Cochrane Library, PsychInfo, Medline) were searched.

STUDY SELECTION: Inclusion criteria were as follows: definition of PPC used; patients aged 0 to 18 years; diseases listed in the directory of life-limiting diseases; results based on empirical data; and combined descriptions of a PPC intervention, its outcomes, and a measurement instrument.

DATA EXTRACTION: Full-text articles were assessed and data were extracted by 2 independent researchers, and each discrepancy was resolved through consensus. The quality of the studies was assessed by using the *Standard Quality Assessment Criteria for Evaluating Primary Research Papers From a Variety of Fields* checklist.

RESULTS: Nineteen of 2150 articles met the eligibility criteria. Researchers in 15 used quantitative methods, and 9 were of moderate quality. Multidimensional outcomes included health-related quality of life, spiritual well-being, satisfaction with care and/or communication, perceived social support, and family involvement in treatment or place-of-care preferences. PPC interventions ranged from home-based to hospital and respite care. Only 15 instruments (of 23 reported) revealed some psychometric properties, and only 5 included patient-reported (child) outcome measures.

LIMITATIONS: We had no access to the developmental process of the instruments used to present the underlying concepts that were underpinning the constructs.

CONCLUSIONS: Data on the psychometric properties of instruments used to assess the impact of PPC interventions were scarce. Children are not systematically involved in reporting outcomes.

---

[a]Institute of Health and Society, Université Catholique de Louvain, Brussels, Belgium; [b]Institut Parnasse-Institut Supérieur d'Enseignement Infirmier, Haute Ecole Léonard de Vinci, Brussels, Belgium; and [c]Department of Public Health and Primary Care, Katholieke Universiteit Leuven, Leuven, Belgium

Ms Friedel conceptualized and designed the study, searched all studies in the databases, screened all studies on the basis of titles and abstracts, screened all full-text articles, designed the extraction tables, extracted all data from the included studies, drafted the initial manuscript, and reviewed and revised the manuscript; Dr Aujoulat contributed to the conceptualization of the study, screening of all full-text articles, design of the extraction tables, extraction of data from the included

To cite: Friedel M, Aujoulat I, Dubois A-C, et al. Instruments to Measure Outcomes in Pediatric Palliative Care: A Systematic Review. *Pediatrics.* 2019;143(1):e20182379

The purpose of pediatric palliative care (PPC) is to enhance the quality of life (QoL) of children and their families when facing life-limiting or life-threatening illnesses. According to the World Health Organization, palliative care for children is the active total care of a child's body, mind, and spirit, and it also involves giving support to the family. It begins when illness is diagnosed and continues regardless of whether a child receives treatment directed at the disease.[1] It is estimated that although ~21 million children worldwide would benefit from a palliative care approach, 8 million children are in need of specialized palliative care.[2]

Measuring outcomes in PPC is considered to be essential to improve clinical care, evaluate the quality of services, and secure funding for programs, and it has been identified as a priority on the research agenda, especially in PPC.[3–8] However, there is currently little evidence of the effectiveness of PPC because it is difficult to define appropriate outcome measures in this field.[9–11] Several obstacles have been identified regarding outcomes research in PPC: small sample sizes, the difficulty of identifying a relevant comparison group, and the broad heterogeneity in children's diseases and ages. Moreover, the relevance of assessing QoL in children within a palliative context has been criticized by some consequentialist authors, who argue that in the face of inevitable death, measuring the impact on QoL would no longer be a priority.[12] These assumptions are often linked to false representations of PPC that are largely restricted to the end-of-life moments.

A systematic review conducted by Coombes et al[13] revealed that there is currently no ideal outcome assessment measure available yet for use in PPC. This finding is in agreement with the conclusions of Knapp and Madden[12] and Huang et al,[14] who found none of the generic QoL measurement instruments to be valid for use in a PPC context.

Measuring QoL has become a growing interest and an end point in many clinical settings. However, in studies of palliative care, QoL may become the principal or only end point of consideration.[15] QoL outcomes are now also commonly called patient-reported or person-reported outcomes to more clearly reflect the broad spectrum of dimensions that are measured (such as pain, fatigue, depression, and observable physical symptoms, such as nausea and vomiting), which are included in the assessment. Measuring children's QoL in a PPC context remains challenging for at least 3 reasons. First, additional dimensions have been suggested by some researchers, such as the ability to cope with illness, the spiritual dimension, and satisfaction with life.[16] Second, depending on the nature of the disease and age of the child, proxy assessments by a relative or other close observer are often used. Third, the diversity of diseases and contexts dealt with by those in PPC often make it difficult to disentangle the impact of disease severity and treatment from the impact of PPC interventions.

QoL is seldom measured directly but rather is explored through a combined assessment of several aspects, which are labeled as dimensions, and although there is disagreement about the aspects that should be included, there is a consensus that QoL should be considered to be a multidimensional construct. Thus, regardless of the instrument used, items of different natures used to target different dimensions will be included.

Commonly, instruments used to measure health-related outcomes rely on 1 of 2 theoretical frameworks depending on the relationship between the items and the construct to be measured.[17] The distinction between reflective and formative models in the field of QoL was introduced by Fayers and Machin.[15]

In a reflective model, the construct manifests itself, and the items are effective direct indicators of that construct (eg, on a scale intended to measure anxiety, all items will reflect a certain degree of anxiousness). In a formative model, the items form or build the construct and are called causal items (eg, in an instrument intended to measure stress, the amount of stress is measured by assessing many items that all contain stress-evoking events). In the field of QoL measurements, many instruments used in PPC settings have a hybrid nature and contain reflective and causal items. They most often are conceptualized as multidimensional scales and contain direct observable, self-reported, and proxy-reported items. Thus, it is challenging to assess their psychometric characteristics.

In this systematic review, we aim to identify and describe the instruments that have been used to assess the impact of PPC interventions and assess their psychometric properties.

METHODS

Search Strategy

This study is in compliance with the Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) guidelines.[18] The methods were prespecified and documented in a protocol.

We conducted primary systematic literature searches using a combination of keywords, including "child," "infant," "pediatric," "adolescent," "young adult," "palliative care," "comfort care," "supportive care," "bereavement care," "respite care," "organization," "standards," "planning," "outcomes," "results," and "effects" (see Supplemental Table 6 for the full search strings). We used broad keywords and Medical Subject Headings terms to maximize inclusiveness and searched 5 databases (Embase, Scopus, The Cochrane Library, PsychInfo and PubMed [Medline]) for studies published from January 1, 2006, to June 1, 2016.

The year 2006 for starting the inclusion of studies was chosen because the first International Meeting for Palliative Care in Children took place that same year in Trento, Italy, leading to the first publication of standards for PPC in Europe in 2007.[19]

Study Selection

All types of PPC interventions and programs were eligible for inclusion, such as supportive care, respite care, and bereavement care.

Furthermore, all types of outcomes (not restricted to QoL but focused on the child, siblings, or parents) were taken into account.

A study was included if all of the following criteria were fulfilled: (1) the full text was written in English, French, German, Italian, or Dutch; (2) the study sample included a clear description of infants, children, and/or adolescents ranging in age from 0 to 18 years; (3) children's diseases were included in the directory of life-limiting diseases[20] or were labeled as life-limiting or life-limited diseases or complex chronic conditions; (4) the study included empirical data; (5) the study presented a combined description of a PPC intervention, outcome, and measure instrument; and (6) a minimal definition of PPC was presented in the study.

In contrast, a study was excluded if any of the following criteria were fulfilled: (1) sickle cell disease, diabetes, obesity, perinatal death, or chronic pain were included; (2) patient age was >18 years; and (3) being restricted to a specific molecule or treatment assessment, to pain as the single outcome, or to an economic assessment.

Children with sickle cell diseases are rarely referred to PPC teams. The management of pain alone was not a criterion of inclusion. Perinatal deaths were also excluded because they concern a specific population with particular PPC needs for which the literature would need to be searched independently.

Data Extraction and Analysis

After retrieving all records, the duplicates were removed. All studies were initially screened on the basis of titles and abstracts and then on the basis of the full text. Three authors (M.F., I.A., and J.M.D.) independently assessed the eligibility of the studies. Any discrepancy was discussed and resolved by consensus. The quality of studies included was assessed by using the *Standard Quality Assessment Criteria for Evaluating Primary Research Papers From a Variety of Fields* checklist.[21] Supplemental Table 7 includes detailed information on the data extraction and analysis.

The Reliability of the Measurement Instruments

There are several ways to interpret a reliability coefficient of a given value depending on the type of characteristic measured and the method of obtaining the estimate of reliability. In classic test theory, a reliability coefficient can be interpreted as the proportion of the observed variance that is "true" rather than the "error variance." However, 1 difficulty with expressing the reliability coefficient as a dimensionless ratio of variances is that it is difficult to interpret in terms of an individual score.[22] The standard error of measurement (SEM) is defined in terms of the SD (σ) and reliability as $\text{SEM} = \sigma \times \sqrt{1 - \text{reliability}}$. Knowledge of the SEM makes it possible to construct a 95% confidence interval (CI) ($95\%\ \text{CI} = \pm 1.96 \times \text{SEM}$) around a person's observed score so that the amount of measurement error around that score can be quantified in a meaningful way.[17] Thus, we decided to compute the SEM whenever possible using either the reported reliability index or any other reliability figures available in the literature.

RESULTS

Our electronic search was performed on June 4, 2016, and we identified 2150 studies. Figure 1 includes the PRISMA flowchart.

After duplicates were removed, 2111 studies were screened on the basis of titles and abstracts, and 44 studies were screened on the basis of the full text. With this procedure, we identified 19 studies that met all the inclusion criteria.

Study Characteristics

Table 1 includes an overview of the main characteristics of the included studies.

A total of 19 studies were included in this systematic review, and researchers in 15 used quantitative methods, those in 1 used qualitative methods, and 3 were based on mixed methods (qualitative and quantitative). Regarding the design of the studies, 9 were retrospective, 6 were prospective, and 4 consisted of randomized controlled trials. Among all the included studies ($n = 19$), researchers in 14 used instruments ($n = 23$ different instruments). Three studies were based exclusively on a chart review, and researchers in 2 studies used interviews or focus groups to measure outcomes. Researchers in the included studies covered the observational period from 1990 to 2014, a time frame of 24 years, excluding 2 studies in

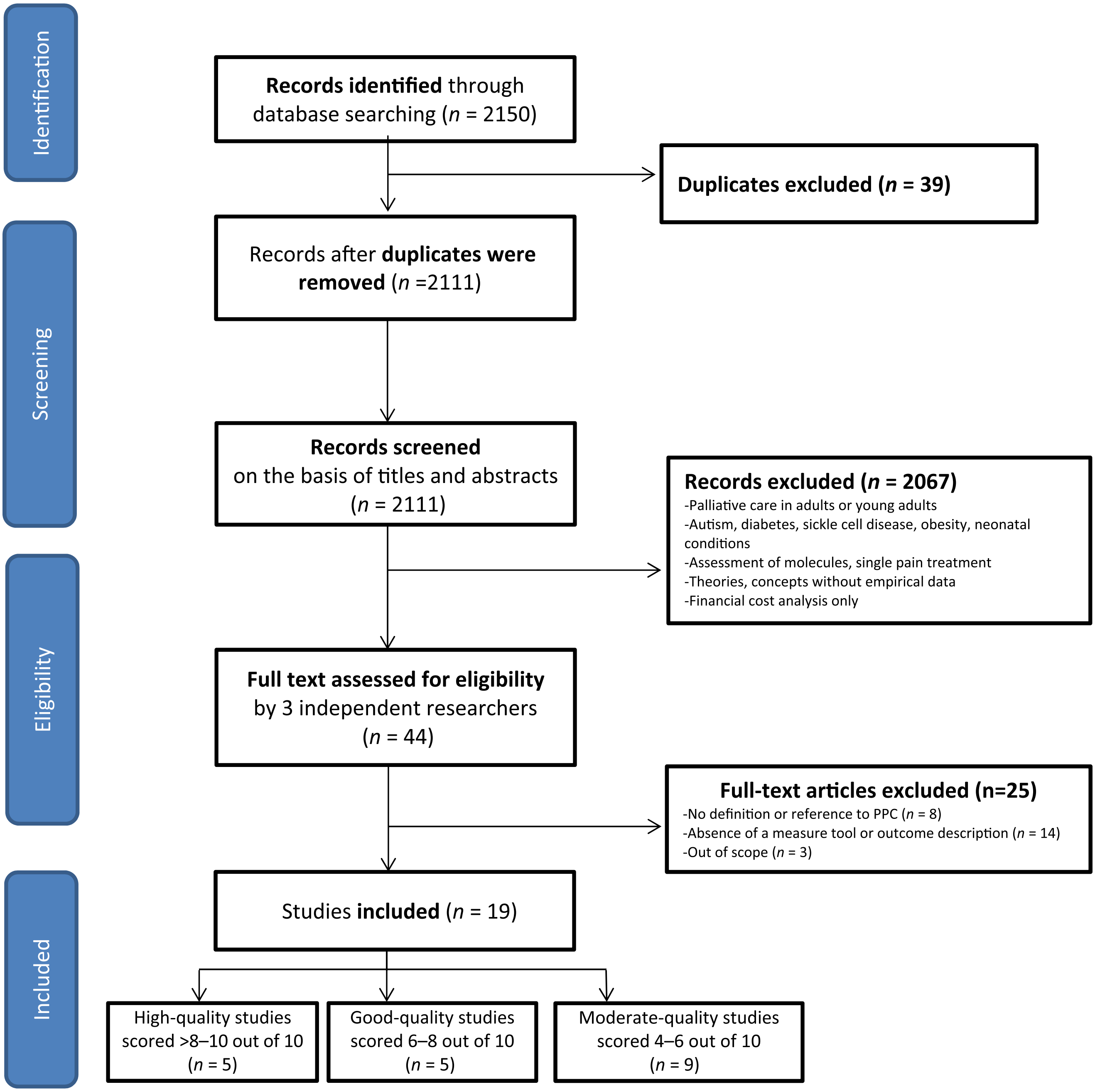

FIGURE 1
PRISMA flow diagram of the included studies in which researchers assess outcomes in PPC.

which researchers did not specify the time period. The patient population included patients with cancer (*n* = 10), children facing various life-limiting or life-threatening conditions (*n* = 8), or children with a "serious illness" (*n* = 1). Of the studies, 8 were conducted in the United States, 7 were conducted in Europe, 2 were conducted in Australia, 1 was conducted in Canada, and 1 was conducted in Lebanon. Researchers in half of the studies collected data exclusively among parents (*n* = 9). Children and parents were both interviewed in 5 studies. Researchers in 1 study collected data among parents and health professionals, and those in another study combined the children's, parents', and health care professionals' perspectives. Sample sizes varied from 11 families to 134 families.

Quality Appraisal of Articles

Classification of the quality of the studies revealed that 5 studies were of high quality (>8 out of 10), 5 were of good quality (6–8 out of 10), and 9 were of moderate quality

TABLE 1 Overview of the Characteristics of the Included Studies in Which Researchers Assess Outcomes in PPC

Reference	Design	Methods	Study Period	Country	Age	Pathology	Sample Size	Instruments	Quality Appraisal (1 = Good; 3 = Moderate)
Akard et al[23]	RCT	Mixed	NR	United States	7–17 y	Cancer	Intervention group *n* = 15; control group *n* = 13	PedsQL 4.0	2
Al-Gharib et al[24]	Cross-sectional	Quantitative	2010–2011	Lebanon	7–18 y	Cancer	*n* = 85	Needs at the End of Life Screening Tool	3
Arland et al[25]	Retrospective cohort	Quantitative	1990–1995; 1996–2005	United States	1 mo–19 y	Cancer (brain tumor)	*n* = 22; *n* = 92	Chart review	3
Bradford et al[26]	Prospective cohort	Quantitative	2010–2011	Australia	0–18 y	LLC	Intervention group *n* = 6; control group *n* = 6	QOLLTI-F questionnaire	3
Conte et al[27]	Match-paired cohort	Quantitative	2008–2012	Canada	NR	LTC	11 pairs	Chart review	3
Eaton[28]	Prospective	Qualitative	NR	United Kingdom	5–16 y	LTC	*n* = 11 families	Interviews	2
Friedrichsdorf et al[29]	Retrospective cohort	Quantitative	2002–2008	United States	0–17 y	Cancer	Intervention group *n* = 30; control group *n* = 30	SCCC questionnaire	2
Gans et al[30]	Prospective	Quantitative	2010–2012	United States	1–20 y	LLC	*n* = 93/50/18	Customized survey	3
Groh et al[31]	Prospective	Quantitative	2011–2012	Germany	1 mo–18 y	Severe illness	*n* = 40	CQ1, CQ2 customized children’s questionnaire, Haüsliche Pflegeskala, HADS, QOLLTI-F	3
Harris et al[32]	Prospective	Mixed	2013–2014	United Kingdom	3 mo–21 y	LLC	*n* = 32 families	Family Empowerment Scale	3
Lyon et al[33]	RCT	Quantitative	2011–2012	United States	14–21 y	Cancer	Intervention group = 17 dyads; control group = 13 dyads	Statement of Treatment Preferences, Decisional Conflict Scale, Quality of Participant-Interviewer Communication, congruence in treatment preferences	1
Lyon et al[34]	RCT	Quantitative	2010–2012	United States	14–21 y	Cancer	Intervention group = 17 dyads Control group = 13 dyads	Satisfaction questionnaire, 5 wishes, Beck Anxiety Inventory, Beck Depression Inventory, PedsQL 4.0, Spiritual Well-Being Scale	1
Remedios et al[35]	Pre-post study	Mixed	2011–2012	Australia	0–18 y	LLC	58 children and/or caregivers	SF-12 version 1, General Health Questionnaire, Chalder Fatigue Scale, Quality of Marriage Index, Multidimensional Scale of Perceived Social Support	2
Schmidt et al[36]	Retrospective cohort study	Quantitative	2000 (cohort 1); 2005 (cohort 2)	Germany	0–18 y	Cancer	46 families (cohort 1); 32 families (cohort 2)	SCCC	3

TABLE 1 Continued

Reference	Design	Methods	Study Period	Country	Age	Pathology	Sample Size	Instruments	Quality Appraisal (1 = Good; 3 = Moderate)
Swallow et al[37]	Prospective	Qualitative	NR	United Kingdom	14–18 y	LLC	15 families (5 teenagers, 15 mothers, 5 fathers)	Interviews and focus groups	1
van der Geest et al[38]	Retrospective cross-sectional study	Quantitative	2000–2004	Netherlands	0–18 y	Cancer	89 parents of 57 deceased children	Inventory of Traumatic Grief, questionnaire on parents' experience, parent's rating of severity and control of symptoms during palliative phase	1
Vern-Gross et al[39]	Retrospective (pre-post) cohort study	Quantitative	2001–2005; 2007–2012	United States	0–21 y	Cancer	57 children (cohort 1); 134 children (cohort 2)	Chart review	2
Vollenbroich et al[40]	Retrospective study	Quantitative	2004–2007	Germany	0–34 y	LLC	43 parents (of 38 deceased children)	Parental questionnaire 1, HADS, Prolonged Grief Disorder–13 Tool, Questionnaire for Health Care Professionals	3
Wolfe et al[41]	Multicenter pilot RCT	Quantitative	2004–2009	United States	>2 y	Cancer	Intervention group: 51; control group: 53	PediQuest (MSAS and PedsQL)	1

CQ1, child questionnaire 1; CQ2, child questionnaire 2; LLC, life-limiting condition; LTC, life-threatening condition; MSAS, Memorial Symptom Assessment Scale; NR, not reported; RCT, randomized controlled trial; SF-12, 12-Item Short-Form Health Survey.

(4–6 out of 10). None of the studies were labeled as being of poor quality.

Types of Interventions

Interventions or programs presented in the studies varied from home care ($n = 6$) to hospital care ($n = 5$), hospice care ($n = 2$), and respite care ($n = 3$) or a combination of home, community-based, and hospital care ($n = 3$).

Flexibility was found in the individualized approach in PPC interventions, in which the focus is placed on the personal desires and priorities expressed by the children and their families. Table 2 includes the types of interventions offered, the outcomes expected versus achieved, and the definition of PPC.

Types of Outcomes

The outcomes addressed in the studies, when focused on children, were multidimensional and included physical (pain, fatigue, dyspnea, and appetite), psychological (anxiety and depression), social (relationships), and spiritual dimensions. QoL, when explicitly explored, was defined as "having fun or experiencing events that added meaning to life."[23,26,27] Other outcomes were related to satisfaction with care and communication with health care professionals, often in parallel with the opportunity given to the children or adolescents to express their wishes, treatment preferences, or place of care. Finally, the length of hospitalization or the place of death were outcomes searched for through chart review and interpreted by authors as quality indicators of services.

Psychometric Properties of the Instruments Used to Measure Outcomes

Table 3 includes the psychometric properties of the instruments included in the review, and Table 4

TABLE 2 Overview of the Type of Interventions and Outcomes of PPC Assessed in the Included Studies

Reference	Care Setting	Type of Intervention	Expected Outcomes	Observed Outcomes	Definition of PPC
Akard et al[23]	Home care	Legacy-making intervention and/or digital storytelling	Evaluate if a legacy-making intervention is a possible strategy to improve QoL	The intervention group that benefitted from a legacy-making intervention showed slightly better emotional and school functioning than controls. Parents reported that their children's digital story provided emotional comfort to them ($n = 11$; 46%), facilitated communication between parents and children ($n = 9$; 38%), and was a coping strategy for them ($n = 4$; 17%). Parents reported that the intervention helped children express their feelings ($n = 19$; 79%), cope ($n = 6$; 27%), and feel better emotionally ($n = 5$; 23%).	ND
Al-Gharib et al[24]	Hospital care	Tertiary pediatric cancer center	Evaluate the quality of medical care received, effect of cancer on patients' relationships, degree of patients' spirituality and/or religiousness, and degree of parents' financial hardships	Participation in decision-making was significantly higher in adolescents than in children ($P = .004$). Compared by age, adolescents showed more readiness to learn from illness than children ($P = .016$). All respondents reported a high quality of medical care.	PPC is an approach that improves the QoL of patients and families facing a life-limiting disease, which may be achieved through effective symptom management as well as spiritual and psychological support from diagnosis until EOL and bereavement. PC is defined as "the active, total care of the child's body, mind, and spirit, and also involves giving support to the family. It begins when illness is diagnosed and continues regardless of whether or not a child receives treatment directed at the disease" (WHO).
Arland et al[25]	Hospital and home care	Comprehensive EOL discussions, medications for symptom control, primary family liaison, home visits	Examine if a relationship existed between specific patient outcomes (ie, symptoms, hospitalizations, and location of death) before and after implementation of a standardized EOL program among pediatric patients with brain tumors: patients who received the EOL program were hospitalized less often ($n = 114$; $\chi^2 = 5.001$ with df = 1; $P < .05$) than patients who did not receive the program	Patients who received the EOL program were hospitalized less often ($n = 114$; $\chi^2 = 5.001$ with df = 1; $P < .05$) than patients who did not receive the program. The authors suggest that these patients may have had a reduced number of hospital admission days because the EOL program included comprehensive EOL care discussions, a continuous infusion of midazolam, an assigned clinic nurse or PNP liaison for communication with the family and the hospice nurse, and health care provider home visits.	Lack of clarity in recognized definitions for both PC and EOL care. It is recognized that PC is focused on reducing and preventing suffering and improving QoL regardless of the patient's diagnosis. In contrast, EOL is recognized as part of the continuum of PC with the major focus on symptom management and prevention of suffering for patients with a life-threatening diagnosis that is expected to lead to death. The common goal of PC and EOL care is achievement of the best QOL for all affected patients.

TABLE 2 Continued

Reference	Care Setting	Type of Intervention	Expected Outcomes	Observed Outcomes	Definition of PPC
Bradford et al[26]	Home care	Home telehealth program: symptom management, emotional support to caregivers	Improve the QoL of primary caregivers of children receiving PC via the HTP	The descriptive analysis of the QoL data that was collected revealed no differences in QOLLTI-F scores between caregivers in the control and intervention groups. Finances and the caregiver's own physical and emotional state were stated as poor. The patient condition, as expected in PC, deteriorated over time. Domains that were scored highly in both groups included quality of care, satisfaction with care, and the environment of care. No relationship was found between QOLTTI-F scores and HTP intervention.	ND
Conte et al[27]	Hospice care	Not described	Compare the No. admissions and length of stay between PPC patient users versus those children who received usual care	PPC patient users were more likely to have advanced directives (100% vs 27%). After controlling for disease and age, we found no significant difference in the number of admissions; however, PPC patient users had an increase in admissions postreferral compared with prereferral (median 3.08 admissions), driven by the need for critical care. We did not find a significant difference in length of stay but observed longer admissions among PPC patient users pre- (1.91 d per mo) and postreferral (3.66 d per mo) than usual care. More than 60% of inpatient use shifted to hospice postreferral.	ND
Eaton[28]	Respite care	Hospice-based and home-based respite care services	Descriptive study: satisfaction with service, parent's contribution to care, benefits of service	The areas of concern identified as significant to all the families were referral to the respite service, service organization, communication, relinquishing control to respite carers, and satisfaction with service.	Definitions of hospice and home-based respite care
Friedrichsdorf et al[29]	Home care	Not described	Reduce symptoms of distress and improve the QoL experience for children	Prevalence of constipation and high distress from fatigue were more common in the PPC and/or oncology group; other distressing symptoms were similar between groups, showing room for improvement. Children who received PPC and/or oncological care were significantly more likely to have fun (70% vs 45%), experience events that added meaning to life (89% vs 63%), and die at home (93% vs 20%).	Those in PPC aim to provide excellent pain and symptom management as well as psychosocial and/or spiritual care regardless of the expected disease trajectory. Integration of PPC services appears to improve outcomes, such as improved pain and symptom management, improved QoL, fewer emergency room visits, and a reduced PICU length of stay.

TABLE 2 Continued

Reference	Care Setting	Type of Intervention	Expected Outcomes	Observed Outcomes	Definition of PPC
Gans et al[30]	Home care, community-based service	Not described	Impact of service to decrease caregiver level of stress and worry	Worry and stress improved in the overall study population between baseline and follow-up surveys. The reductions over time were significant when controlling for the effects of family support and the child's age and disease severity, resulting in a 0.26-point reduction per survey wave for both worry and stress. Family support was also predictive of reductions in both stress and worry. Disease severity was also significantly predictive of stress. Age was not independently associated with changes in caregiver stress and worry. Caregivers' perceptions of the ability of PPC to reduce or control the children's pain and other symptoms received average ratings of 8.6 and 9.3, respectively, on a 10-point scale.	From the time of diagnosis with an LTD and as a supplement to clinical treatment, PPC provides children with holistic care that considers every aspect of the effect of the diagnosis on the child's and family's physical, psychological, and spiritual health. For the child, PC may include pain and symptom management, child life counselling, and expressive therapies that provide the child with developmentally appropriate coping mechanisms and tools for self-expression. For the family, PC may include training on health care system navigation, treatment decisions, optimal care for the child, and bereavement counselling before and after the death of a child. Pediatric conditions are different from more prevalent terminal illnesses in adults and require different modalities and care plans (ICPCN).
Groh et al[31]	Home care	Palliative medical and nursing care, 24/7 on-call service, psychosocial support, coordination of professional assistance	Address the needs of patients and their families, increase acceptance and effectiveness of PPC, improve QoL in children and their families	After the involvement of specialized PPHC, caregivers' satisfaction with care and quality of care significantly improved, as could be documented for 12 of 14 issues (NRS). The involvement of the PPHC team led to a significant improvement of children's symptoms and QoL ($P < .001$) as perceived by the parents; and the parents' own QoL and burden relief significantly increased (QOLLTI-F: $P < .001$; 7-point change on a 10-point scale), whereas their psychological distress and burden significantly decreased (HADS: $P < .001$; HPS: $P < .001$).	PPC is focused on achieving the best possible QoL for patients and their families and requires a multidisciplinary approach encompassing physical, emotional, social, and spiritual domains (WHO).
Harris et al[32]	Hospice care	MyQuality	Acceptability of the tools and patterns of usage, increase in family empowerment	The mean duration of tool use was 106 d (range 2–301). The 32 families identified 114 priorities in total, with most identifying 2 or 3 (range 1–15). The most common priorities identified were seizures (24 of 32), constipation (9 of 32), pain (6 of 32), and sleep problems (6 of 32). Priorities were modified from the suggested options or entered as free text by 20 parents.	PC for children encompasses symptom control for the child and emotional and psychological support for the child and family and addresses practical, financial, and spiritual needs. The aim of those in PC is to maximize QoL rather than its duration. PC may be required for children with a wide variety of LLCs, including cancer; organ failure; metabolic, genetic, or degenerative illnesses; or static conditions, such as severe cerebral palsy or epilepsy, for which LTC means survival into adulthood is unlikely.

TABLE 2 Continued

Reference	Care Setting	Type of Intervention	Expected Outcomes	Observed Outcomes	Definition of PPC
Lyon et al[33]	Hospital care	Three sessions of family-centered ACP: interviews	Examine the efficacy of family-centered ACP: document specific treatment preferences of patients and a surrogate understanding of what the patients would want, measure the degree of uncertainty about the course of action to be taken, improve quality of communication and congruence in treatment preferences, decrease decisional conflict	Significantly increased congruence was observed for intervention dyads compared with controls for 4 of the 6 disease-specific scenarios; for example, for situation 2 ("treatment would extend my life by not more than 2–3 mo"), intervention dyads demonstrated higher congruence (= 0.660; *P* = .001) versus control dyads (= 0.0636; *P* = .70). Intervention adolescents (100%) wanted their families to do what was best at the time, whereas fewer control adolescents (62%) gave families this leeway. Intervention adolescents were significantly better informed about EOL decisions (t = 2.93; effect size 0.961; 95% CI 0.742 to 1.180; *P* = .007). Intervention families were more likely to concur on limiting treatments than controls. An ethnic difference was found in only 1 situation.	Quality of PC is a health care priority.
Lyon et al[34]	Hospital care	Pediatric advanced care planning intervention (FACE-TC) during cure-directed treatments	Test feasibility, acceptability, and safety of a pediatric ACP intervention called FACE-TC	Acceptability was demonstrated with enrollment of 72% of eligible families, 100% attendance at all 3 sessions, 93% retention at 3 mo postintervention, and 100% data completion. Intervention families rated FACE-TC as worthwhile (100%), whereas adolescents' ratings increased over time (65%–82%). Adolescents' anxiety decreased significantly from baseline to 3 mo postintervention in both groups. Low depressive symptom scores and high QoL scores were maintained by adolescents in both groups. Advance directives were located easily in the medical records (100% of FACE-TC adolescents versus none in controls). Oncologists received electronic copies. Total spirituality scores were significantly higher among FACE-TC adolescents versus controls.	PC currently exists in the context of a changing health care delivery system, in which death is medicalized. Four of 5 children with complex chronic conditions in the United States die in the hospital. Among children with cancer, 72% of those without PC died in the ICU versus 21% of those with PC. ACP is 1 dimension of PC that provides support in decision-making about limiting burdensome medical interventions and advance directives.

TABLE 2 Continued

Reference	Care Setting	Type of Intervention	Expected Outcomes	Observed Outcomes	Definition of PPC
Remedios et al[35]	Respite care	Out-of-home respite care service	Impact of respite care service on levels of fatigue, psychological adjustment, QoL, relationship satisfaction among caregivers	Caregivers had below-standard levels of QoL compared with normative populations. Paired t tests revealed that caregivers' average psychological adjustment scores significantly improved from prerespite (mean = 13.9; SE = 0.71) to postrespite care (mean = 10.7; SE = 1; $P < .001$; 95% CI 1.25 to 5.11). Furthermore, caregivers' average fatigue scores significantly improved from prerespite (mean = 14.3; SE = 0.85) to postrespite care (mean = 10.9; SE = 1.01; $P < .001$; 95% CI 1.69 to 7.94), and caregivers' average mental health QoL scores significantly improved from prerespite (mean = 44.2; SE = 1.8) to postrespite care (mean = 49.1; SE = 1.6; $P < .01$; 95% CI −9.56 to 0.36). Qualitative data revealed that caregivers sought respite for relief from intensive care provision and believed this was essential to their well-being.	Respite has been recognized as an important supportive service in PPC. Broadly, pediatric respite involves individuals other than family members caring for a child with a medical condition for a short period of time. It is generally accepted that caregivers are the intended beneficiary of respite, with the aim being to provide a break from intensive caring and thereby promote a sense of well-being.
Schmidt et al[36]	Home care	Not described	PPC services reduce the symptoms and suffering of children, increase palliative home care, change the place of death	The children of both parental cohorts were similar in terms of disease characteristics and sociodemographic variables. Children suffered in a similar manner from core symptoms, such as pain and dyspnea. However, symptom treatment increased for all symptoms. In the case of the treatment of anxiety, the increase was statistically significant ($P = .035$). The location of care changed, with approximately three-quarters of the 2005 cohort receiving palliative home care, which was significantly more than in the 2000 cohort ($P = .007$). Additionally, fewer children in the 2005 cohort died in the ICU.	ND
Swallow et al[37]	Respite care	Offer a break, nursing and medical care, physiotherapy, social work, play and occupational therapy, clergy support providing residential care 24/7	Adolescent and parent satisfaction with the service provided, skill of staff, partnership, access to leisure activities, opportunities for independence and peer support	Three themes emerged: accessibility and communication, needs and boundaries, and shaping the service. Teenagers enjoyed regularly planned residential breaks, access to skilled staff and facilities to support their needs, opportunities to meet others with life-limiting conditions, and fun time away from home, thereby giving parents peace of mind, a regularly planned break from caregiving, and opportunities to meet other parents and spend exclusive time with their other children.	Definition of respite care

TABLE 2 Continued

Reference	Care Setting	Type of Intervention	Expected Outcomes	Observed Outcomes	Definition of PPC
van der Geest et al[38]	Hospital care	Pediatric oncology center	Parents' perception and satisfaction with the interaction with HCPs (communication, continuity of care, parental involvement), symptom management, reduce long-term parental grief	Parents highly rated communication (4.6 ± 0.6), continuity of care (4.3 ± 0.6), and parental involvement (4.6 ± 0.7) during the palliative phase. Parents' most often reported physical and psychological symptoms of their children during the palliative phase were fatigue (75%), pain (74%), anxiety being alone (52%), and anger (48%). Higher ratings of parents on communication (b = −9.08; $P = .03$) and continuity of care (b = −11.74; $P = .01$) were associated with lower levels of long-term parental grief. The severity of the child's dyspnea (b = 2.96; $P = .05$), anxiety being alone (b = 4.52; $P < .01$), anxiety about the future (b = 5.02; $P < .01$), anger (b = 4.90; $P < .01$), and uncontrolled pain (b = 6.60; $P < .01$) were associated with higher levels of long-term parental grief. Multivariate models used to combine the interaction with HCPs and symptom management revealed a significant influence of both aspects on long-term parental grief.	PC is defined by the WHO (1998) as ''the active total care of the child's body, mind, and spirit,'' and also comprises care for the family during and after the child's death.[16] According to the WHO, ''PC begins when illness is diagnosed and continues regardless of whether or not a child receives treatment directed at the disease.''
Vern-Gross et al[39]	Hospital and home care	Inpatient and/or outpatient follow-up, team meetings, home visits, interdisciplinary team, symptom management, psychosocial and spiritual support, coordinated care, facilitate EOL discussion, decision-making, and bereavement support	Reduce symptom burden, preferred location of death respected, EOL discussion, bereavement support	The median time to the first QoL and/or PC consultation was 17.2 mo (range 9–33). At consultation, 60% of children were not receiving or discontinued cancer-directed therapy. Within the QoL and/or PC cohort, 54 patients had documented symptoms, 94% required intervention for >3 symptoms, and 76% received intervention for >5 symptoms. Eighty-three percent achieved their preferred place of death. Compared with the historical cohort, the QoL and/or PC cohort had more EOL discussions per patient (median 12 vs 3; $P < .001$), earlier EOL discussions with longer times before do-not-resuscitate orders (median 195 vs 2 d; $P < .001$), and greater hospice enrollment (71% vs 46%; $P = .002$).	Integration of PC for these children improves EOL care, enhances ACP, and substantially decreases suffering by improving communication, providing optimal symptom control, and emphasizing QoL.

TABLE 2 Continued

Reference	Care Setting	Type of Intervention	Expected Outcomes	Observed Outcomes	Definition of PPC
Vollenbroich et al[40]	PPHCT	Coordination of professional assistance, provision of palliative treatment, interdisciplinary care and HCPs, 24/7 service	To evaluate satisfaction with the service, satisfaction with the course of the dying phase, and reduce anxiety, depression, and prolonged grief disorder	Satisfaction with the PPHCT was scored a median of 10 (numeric rating scale, 0–10). The child's death was predominantly experienced as peaceful (median 9); 71% died at home. According to parents, involvement of the PPHCT led to highly significant ($P < .001$) improvements in the children's symptoms and QoL as well as in aspects of communication and administrative barrier reduction. Anxiety was detected in 25% of parents, depression in 19%, and prolonged grief disorder in 13%. HCPs (return rate 83%) evaluated all investigated care domains (particularly cooperation, communication, and/or family support) as being significantly improved ($P < .001$). Thirty-five percent of HCPs felt uncertain concerning PPC; 79% would welcome specific training opportunities.	The goal of PPC is to achieve the best QoL for patients and their families, consistent with their values, regardless of the location of the patient (WHO).
Wolfe et al[41]	Hospital care	—	To determine if feedback on patient-reported outcomes to providers and families of children with advanced cancer improves symptom distress and health-related QoL	Feedback did not significantly affect the average MSAS, PedsQL 4.0, or sickness score trends. Post hoc subgroup analyses among children age 8 y who survived 20 wk revealed that feedback improved PedsQL 4.0 emotional (8.1; 95% CI 1.8 to 14.4) and sickness (8.2; 95% CI 14.2 to 2.2) scores. PediQuest reports were valued by children, parents, and providers and contributed at least sometimes to physician initiation of a psychosocial consult (56%).	Ensuring the best possible QoL for children with cancer is a high national priority.

ACP, advanced care planning; df, degrees of freedom; EOL, end of life; FACE-TC, family-centered advance care planning for teens with cancer; HCP, health care professional; HPS, Haüsliche Pflegeskala; HTP, home telehealth program; ICPCN, International Children's Palliative Care Network; LLC, life-limiting condition; LTC, life-threatening condition; LTD, life-threatening disease; MSAS, Memorial Symptom Assessment Scale; ND, not described; NRS, Numeric Rating Scale; PC, palliative care; PNP, pediatric nurse with palliative training; PPHCT, pediatric palliative home care team; WHO, World Health Organization; —, not applicable.

includes the most often cited instruments.

Among the studies, 23 different instruments were identified (Pediatric Quality of Life Inventory 4.0 [PedsQL 4.0] in 3 studies, Quality of Life in Life-Threatening Illness–Family Carer version [QOLLTI-F] in 2 studies, Survey About Caring for Children With Cancer [SCCC] in 2 studies, and Hospital Anxiety and Depression Scale [HADS] in 2 studies), all of which were standardized measures. The average of the number of instruments used per study was 2, ranging from 1 to 6 instruments.

Among the psychometric properties, the Cronbach α was the most frequently reported value (indicated in 15 of 23 instruments). The Cronbach α is a measure of internal consistency that reveals how closely related a set of items are as a group. However, no data other than consideration of the face validity of the instruments were found among the included studies.

The SEM could be computed for 12 different instruments presented in 8 different studies. The SEM varied from 0.38 on a scale from 0 to 70 (with 95% CI = ± 0.74) for the QOLLTI-F to 6.27 on a scale from 0 to 100 (with 95% CI = ± 12.29) for the PedsQL 4.0.

DISCUSSION

PPC is a recent subspecialty in medicine that has rapidly developed over the past 2 decades. Attempts to assess the impact of such interventions are progressively published, but the findings of this review reveal that the psychometric properties of the instruments used in this field are not well documented. The aim of those in PCC is to promote QoL, but a precise description of how this construct is operationalized has not been provided in any of the

TABLE 3 Overview of the Instruments Used in the Included Studies in Which Researchers Assess Outcomes in PPC

Reference	Name of Instruments and/or Methods Used for Assessment	References	Child, Parent, or HCP Report	No. Items	Psychometric Properties	SEM and 95% CI
Akard et al[23]	PedsQL 4.0	Varni et al[42,43]	Parent and child	22	PedsQL 4.0 Cronbach α 0.88	SEM PedsQL 4.0 = 6.27; 95% CI = ± 12.29
Al-Gharib et al[24]	NEST	Emmanuel et al[44]	Parent and child	—	NEST Cronbach α 0.64–0.86	SEM NEST = 0.53–0.7; 95% CI = ± 1.18
Arland et al[25]	Chart review	NA	NA	NA	NA	NA
Bradford et al[26]	QOLLTI-F	Cohen et al[45]	Parent	16	QOLLTI-F Cronbach α 0.857	No sufficient data available
Conte et al[27]	Chart review	NA	NA	NA	NA	NA
Eaton[28]	Interview	NA	Parent	NA	NA	NA
Friedrichsdorf et al[29]	SCCC	Wolfe et al,[46] Hechler et al[47]	Parent	211	ND	No sufficient data available
	—	Mack et al[48,49]	Parent	—	—	No sufficient data available
	—	Edwards et al[50]	Parent	—	—	No sufficient data available
	—	Dussel et al[51]	Parent	—	—	No sufficient data available
Gans et al[30]	Chart review, own survey	—	Parent	—	NA	NA
Groh et al[31]	Child questionnaire 1	Gräßel,[52] Gräßel et al[53]	Parent	71	—	—
	Child questionnaire 2	Snaith[54]	Parent	61	HADS Cronbach α 0.78–0.90	For HPS and HADS, insufficient data available
	Haüsliche Pflegeskala	Hermann-Lingen et al[55]	Parent	10	Pearson correlation coefficient 0.49–0.63	SEM QOLLTI-F = 0.38
	HADS	Cohen et al[45]	Parent	14	QOLLTI-F Cronbach α 0.857	95% CI = 0.74
	QOLLTI-F	—	Parent	19	QOLLTI-F Cronbach α 0.857	95% CI = 0.74
Harris et al[32]	MyQuality	Harris,[56] Harris et al[57]	Parent	34	FAS	—
	FAS	Koren et al,[58] Nachshen[59] Herbert et al[60]	Parent	—	Congruence coefficients 0.88–0.98	No sufficient data available
	—	—	—	—	Cronbach's α 0.78–0.89	—
	—	—	—	—	Split half 0.93	—
Lyon et al[33]	Statement of Treatment Preferences	Kirchhoff et al[61]	Parent and child	10	DCS test and retest 0.81	For DCS and CTP, no sufficient data available
	DCS	O'Connor[62]	Parent and child	—	Cronbach α 0.78–0.92	SEM QPIC = 0.54
	QPIC	Engelbert et al[63]	Parent and child	—	Effect size 0.961 (95% CI 0.742–1.180; $P = .007$)	95% CI = ± 1.06
	CTP	—	—	—	—	—
Lyon et al[34]	Satisfaction questionnaire	Beck and Steer,[64] Beck et al[65]	Parent and child	21	BAI Cronbach α > 0.70	SEM BAI = 3.18
	Five wishes	Varni et al[66]	Parent and child	22	BDI Cronbach α 0.91	95% CI = ± 6.23
	BAI	Peterman et al[67]	Parent and child	12	PedsQL 4.0 Cronbach α 0.88	SEM BDI = 1.71
	BDI	—		—	FACIT: Cronbach α 0.86	95% CI = ± 3.35
	PedsQL 4.0	—		—	—	SEM PedsQL 4.0 = 6.02
	Spiritual Well-Being Scale (FACIT)	—		—	—	95% CI = ± 11.8
	—	—		—	—	SEM FACIT = 6.14
	—	—		—	—	95% CI = ± 12.03
Remedios et al[35]	SF-12 version 1	Ware et al[68]	Parent	12	SF-12 Cronbach α 0.72–0.89	SEM SF-12 = 4.65
	GHQ	Goldberg et al[69]	Parent	12	SF-12 test and retest 0.73–0.86	95% CI = ± 9.11
	CFS	Chalder et al[70]	Parent	11	GHQ Cronbach α 0.82–0.86	SEM GHQ = 1.76
	QMI	Welch et al[71]	Parent	6	CFS Cronbach α 0.80–0.86	95% CI = ± 3.45
	MSPSS	Zimet et al[72]	Parent	12	QMI Cronbach α > 0.90 test 0.65	SEM CFS = 2.06
	—	—	—	—	MSPSS Cronbach α 0.89	95% CI = ± 4.04

TABLE 3 Continued

Reference	Name of Instruments and/or Methods Used for Assessment	References	Child, Parent, or HCP Report	No. Items	Psychometric Properties	SEM and 95% CI
	—	—	—	—	—	SEM QMI = 1.74
	—	—	—	—	—	95% CI = ± 3.41
Schmidt et al[36]	SCCC	Hechler et al,[47] Wolfe et al[46]	Parent	NR	NR	No sufficient data available
Swallow et al[37]	Interviews and focus group	NA	Parent and child	—	NA	NA
van der Geest et al[38]	Inventory of traumatic grief, questionnaire on parents' experience, parents' rating of severity and control of symptoms during palliative phase	Boelen et al[73]	Parent	29	—	Only subscales, no total score of the SEM could be computed
	—	—	—	17	—	—
	—	—	—	9	—	—
Vern-Gross et al[39]	—	—	Chart review	—	NA	NA
Vollenbroich et al[40]	Parental Questionnaire 1, HADS	Snaith[54]	Parent and HCP	56	HADS Cronbach α 0.78–0.90	No sufficient data available
	Prolonged Grief Disorder–13 Tool, Questionnaire for Health Care Professionals	Prigerson et al[74]	Parent and HCP	14	Pearson correlation coefficient 0.49–0.63	No sufficient data available
	—	—	—	13	—	—
	—	—	—	22	—	—
Wolfe et al[41]	PediQuest composed by Collins et al[75]	Collins et al[75]	Parent, child, and HCP	24	MSAS Cronbach α 0.67	SEM MSAS = 4.94
	Memorial Symptom Assessment Scale	Varni et al[42]	Parent, child, and HCP	22	PedsQL 4.0 Cronbach α 0.88	95% CI = ± 9.68
	PedsQL 4.0	—	—	1	PedsQL 4.0 Cronbach α 0.88	SEM PedsQL 4.0 = 5.85
	Sickness question	—	—	—	—	95% CI = ± 11.47

BAI, Beck Anxiety Inventory; BDI, Beck Depression Inventory; CFS, Chalder Fatigue Scale; CTP, congruence in treatment preferences; DCS, Decisional Conflict Scale; FACIT, Functional Assessment of Chronic Illness Therapy; FAS, Family Empowerment Scale; GHQ, General Health Questionnaire; HCP, health care professional; MSAS, Memorial Symptom Assessment Scale; MSPSS, Multidimensional Scale of Perceived Social Support; NA, not available; ND, not described; NEST, Needs at the End of Life Screening Tool; NR, not reported; QMI, Quality of Marriage Index; QPIC, Quality of Participant-Interviewer Communication Scale; SF-12, 12-Item Short-Form Health Survey; —, not applicable.

included studies. As a matter of fact, QoL was addressed as an outcome directly or indirectly in only 6 of the 19 studies. Measuring the QoL of children in the context of a life-limiting disease requires rigorous conceptual and methodological foundations to capture core domains by using a holistic and family-centered approach. Some methodological, clinical, and conceptual challenges will be further discussed.

Patient-Reported Outcome Measures Versus Proxy-Reported Outcome Measures

Overall, this review revealed that outcome measures are used predominantly to explore parental (proxy) perspectives retrospectively concerning a past event, such as children's symptoms, circumstances of death, or satisfaction with the care received. However, retrospective studies have been found to be less reliable than prospective ones regarding assessing outcomes. Conte et al[76] affirm that prospective studies are needed to better evaluate the overall impact of PPC on the health care system from the perspectives of children and families. Furthermore, outcome measures in PPC seldom involve the perspectives of the children, as found in our review, in which researchers in only 5 studies addressed children's views. Ethical concerns are widely documented, and researchers argue that participating in interviews may be burdensome for children facing life-limiting conditions.[77] In recent decades, however, several authors have recommended the involvement of children in research, showing that if the design is adapted to the children's cognitive and physical conditions and researchers adopt a compassionate and sensitive approach, participation in studies would be valued by children and their parents and would even improve their decision-making

TABLE 4 Overview of the Most Cited Instruments Used to Assess Outcomes in PPC in the Included Studies

Name of Instrument	Dimensions Assessed	Form	No. Items	Proxy or Self-Report	References of Studies With This Instrument
PedsQL 4.0	Children's QoL: physical, emotional, social functioning, and school	Questionnaire and rating scale	23	Self-report (child) and proxy report (parent)	23·34·41
QOLLTI-F	Caregiver's QoL: perceived QoL; patient's environment and condition; caregiver's physical, emotional, and cognitive condition; meaning in life; quality of care; relationships; and financial worries	Questionnaire and rating scale	16	Self-report (parent)	26·31
SCCC	End-of-life care domains: symptoms and their treatment, QoL, and parent sociodemographic characteristics	Questionnaire and rating scale	211	Proxy report (parent)	29·36
Needs at the End of Life Screening Tool	Adult's financial burden, quality of medical care, spirituality and/or religion, and relationships	Questionnaire and rating scale	35	Self-report (child) and proxy report (parent)	24
HADS	Parent's anxiety and depression	Questionnaire and rating scale	14	Self-report (parent)	31·40

capacity.[78] When children are involved in research, they are frequently associated or compared with proxy parental measures. In doing so, the discrepancy between self-report (by children) and proxy reporting (by parents) is found, as reported in other studies,[79–82] because of the unique subjective evaluations made by children, which clearly differs from those made by their parents.

The Need for Measurement Instruments in PPC

It is hypothesized that the use of patient-reported outcome measures (PROMs) would improve the quality of care provided. Nevertheless, this link is rarely assessed. Measurement instruments can be used for several purposes. They can be intended to be used as a primary guide for the caregiving process rather than an assessment of the impact of the interventions or a monitor of patient-related health outcomes over time. Physicians often consider PROMs to be useful for the assessment and screening of patients, whereas nurses perceive those instruments as almost relevant for clinical decision-making.[83]

Difficulty measuring outcomes lies in the fact that all the described interventions could be labeled as complex interventions, which are defined by Craig et al[84] as requiring a large number of health care professionals, organizational levels, and outcomes and a large degree of flexibility. Whether at home or in a hospital, PPC interventions are characterized by interdisciplinary teams composed of nurses, physicians, psychologists, and social workers. Coordination of care is frequently ensured by several levels of care: primary home-care teams collaborating with specialist hospital-based teams. Interdisciplinary teams are a core element of quality care in PPC because the aim is to promote QoL not only for a child but also for the whole family, considering all the dimensions, such as physical health, psychological well-being, and social relationships.

Core Dimensions to Be Measured

In adults, a study revealed that questions relating to pain, symptoms, emotional needs, and family concerns are consistently considered to be the most useful and important components of palliative PROMs.[85] Dimensions that matter the most for children in PPC are currently not known. According to its definition, palliative care of children consists of a family-centered approach to achieve the best QoL of the children embedded among family members. This focus should consequently be addressed through the choice of a measure that is used to evaluate the outcomes of PPC on the children and their families. This idea was pointed out by Dussel and Medin.[86] However, patient involvement in the development of a PROM is paramount and has been documented in adults by Wiering et al[87] but not in children. As already mentioned, health-related outcome measures are either based on formative (causal) or reflective models or a mix of the 2.[17] For this review, we were interested in discovering which model was used for the instruments that we identified. We found that all the instruments identified in this review were based on reflective models, which indicated that the explored dimensions reflected the underlying construct, such as the quantity of pain or the degree of physical impairment.

Generic Versus Specific Disease-Oriented Instruments

Another challenge is related to the choice of specific (disease-specific, such as cancer) versus generic instruments.[88] Generic measures may be useful in comparing outcomes across different populations and interventions, especially for

TABLE 5 Presentation of the APCA c-POS

The APCA c-POS[96] is an instrument derived from the validated palliative outcome scale developed at Cicely Saunders Institute, King's College London.[9]

In the same unique document, the APCA c-POS is used to explore the multidimensional outcomes of PPC and the physical and psychological domains for any child facing life-limiting or life-threatening conditions independently of age or type of disease and the psychological dimensions of the parents.

The instrument is composed of 2 parts: part A is used to address 7 questions in children (self-report), and part B is 5 questions for their parents.

If the child isn't able to respond for himself or herself in part A because of young age (<6 y) or poor cognitive capacities, a proxy report (to be filled in by parents) is planned to address the same dimensions.

Example of questions in part A used to explore child's perceptions:

"Can you tell me how often you have felt happy since yesterday?"

Example of questions in part B used to explore parental perceptions:

"Have you been able to share how you are feeling about your child's illness with others when you have wanted to?"

---

cost-effectiveness studies. Disease-specific measures are used to assess the special states and concerns of diagnostic groups. Specific measures may be more sensitive for the detection and quantification of small changes that are important to clinicians or patients.[89] A systematic review performed by Janssens et al[90] revealed that many generic instruments are available to assess children's health. However, the authors found that only a few PROMs were usable across all age ranges up to 18 years. Most PROMs excluded at least 1 major domain, and all of them conflated elements of functioning and well-being in the scales.[90] Additionally, the authors found at least 5 different psychometric properties for the Child Health and Illness Profile, Healthy Pathways, the Kidscreen, and the Multidimensional Student's Life Satisfaction Scale.[91]

Standardized Versus Individualized Measures

Standardized measures are instruments that contain predefined domains, whereas individualized measures are instruments that encourage patients to describe which domains are important to them. The only individualized measure of QoL is called the Scheduled Evaluation of Individual Quality of Life and was developed by Hickey et al[92] in 1996.

Standardized measures of QoL have been criticized by several authors, who argue that some psychometric properties of those measures, such as the test and retest, are not valid in a PPC context. Rather, the same authors suggest the use of individualized measures of QoL,[13,14] which seem to better reflect the subjective perception of QoL independently of the decline in physical function. Another perspective is presented by Wolpert,[93] who recommended combining at least 1 standardized measure with 1 individualized measure to optimally assess QoL.

New Developments in the Field

Systematic reviews on PROMs have been performed for children suffering from burns[94] or who are visually impaired,[95] but specific PROMs for PPC are clearly lacking.

Recently, through expert meetings and international conferences, a generic promising PROM was developed that is used to address the specific methodological and conceptual challenges emerging from the PPC context.[96,97] This multidimensional instrument is used to explore children's perspectives on psychical, psychological, and social aspects of care while receiving palliative care. Parental perspectives on information, confidence, and worries are equally assessed by using the same tool.

See Table 5, in which we present the African Palliative Care Association Children's Palliative Outcome Scale (APCA c-POS). Because no reports were published yet on the use of this new instrument in relation to an intervention, we did not integrate this development in our overview.

PROMs make up an instrument that is used to measure outcomes of care provided and to provide valuable information on quality of care. Nevertheless, the impact of care should be assessed not only by using outcomes measures. Documentation of the processes and types of interventions are required. Because those in PPC intend to promote QoL, a question could concern how much PROMs contribute to the overall aim to evaluate QoL or the common ground or convergent domains assessed equivalently by using PROMs compared with QoL instruments.[98]

Implementation of PROMs in Clinical Practice

After the validation of an outcomes measure tool in PPC, a next step would be a rigorous implementation process for which guidelines are issued.[99,100] Dissemination and implementation science is a growing research field that is focused on the best strategies to implement evidence practice.[101] A lack of education and training were found to be the most common reasons leading to the poor implementation of PROMs and therefore must be strengthened.[102] Some authors point out the importance of involving health care professionals right from the start during the developmental stages of a measure tool to improve its implementation.[103,104] Patient satisfaction toward PROMs is an important element to consider in implementation research, which was explored by Recinos et al[105] in adults; however, similar research is lacking in children.

Strengths and Limitations

With this systematic review, we offer for the first time (to our knowledge) an in-depth description of the instruments used to assess the impact of PPC interventions.

Our assessment of the psychometric properties related to instruments was restricted to the data that were retrievable from the publications included. We had no access to the developmental process used for the instruments or the underlying concepts underpinning the construction of the scales.

CONCLUSIONS

This systematic review led to 2 observations. First, a broad spectrum of outcomes are targeted in the studies, demonstrating a lack of consensus on the dimensions that should be measured when assessing PPC interventions. Second, the psychometric properties of outcome measurement instruments are poorly documented.

It is recommended to strengthen prospective studies involving children in a sensitive and age-appropriate way. Rigorous methods are needed to develop new instruments that are able to measure QoL by using a multidimensional and family-centered approach. The use of instruments that combine self and proxy reports or even those that are used to address individual QoL requires further investigation. Coconstruction of the tool should be planned with health care professionals to guarantee optimal implementation into clinical care.

ABBREVIATIONS

APCA c-POS: African Palliative Care Association Children's Palliative Outcome Scale
CI: confidence interval
HADS: Hospital Anxiety and Depression Scale
PedsQL 4.0: Pediatric Quality of Life Inventory 4.0
PPC: pediatric palliative care
PRISMA: Preferred Reporting Items for Systematic Reviews and Meta-analyses
PROM: patient-reported outcome measure
QoL: quality of life
QOLLTI-F: Quality of Life in Life-Threatening Illness–Family Carer Version
SCCC: Survey About Caring for Children With Cancer
SEM: standard error of measurement

---

studies, and discussion section and reviewed and revised the final manuscript; Ms Dubois contributed to the design of the extraction tables and extraction of data from the included studies and reviewed and revised the final manuscript; Dr Degryse contributed to the design of the study, screening of all full-text articles, design of the extraction tables, extraction of all data from the included studies, and discussion section and reviewed and revised the manuscript; and all authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

This trial has been registered with the International Prospective Register of Systematic Reviews (https://www.crd.york.ac.uk/prospero/) (identifier CRD42017080541).

DOI: https://doi.org/10.1542/peds.2018-2379

Accepted for publication Oct 1, 2018

Address correspondence to Marie Friedel, PNP, MPH, Institut de Recherche Santé et Société, Université Catholique de Louvain, 30, Clos Chapelle-aux-Champs, Boite 1.30.13, 1200 Brussels, Belgium. E-mail: marie.friedel@uclouvain.be

PEDIATRICS (ISSN Numbers: Print, 0031-4005; Online, 1098-4275).

FINANCIAL DISCLOSURE: The authors have indicated they have no financial relationships relevant to this article to disclose.

FUNDING: Funded by the Fonds de Soutien Marguerite-Marie Delacroix and the Institute Parnasse-Institut Supérieur d'Enseignement Infirmier at Haute Ecole Léonard de Vinci (Brussels, Belgium) as part of a doctoral project (Ms Friedel).

POTENTIAL CONFLICT OF INTEREST: The authors have indicated they have no potential conflicts of interest to disclose.

COMPANION PAPER: A companion to this article can be found online at www.pediatrics.org/cgi/doi/10.1542/peds.2018-3347.

REFERENCES

1. World Health Organization. WHO definition of palliative care. 1998. Available at: www.who.int/cancer/palliative/definition/en/. Accessed February 12, 2018
2. Connor SR, Downing J, Marston J. Estimating the global need for palliative care for children: a cross-sectional analysis. *J Pain Symptom Manage.* 2017;53(2):171–177
3. Chong PH, De Castro Molina JA, Teo K, Tan WS. Paediatric palliative care improves patient outcomes and reduces healthcare costs: evaluation of a home-based program. *BMC Palliat Care.* 2018;17(1):11
4. Goldhagen J, Fafard M, Komatz K, Eason T, Livingood WC. Community-based pediatric palliative care for health related quality of life, hospital utilization and costs lessons learned

from a pilot study. *BMC Palliat Care*. 2016;15:73

5. Downing J, Knapp C, Muckaden MA, Fowler-Kerry S, Marston J; ICPCN Scientific Committee. Priorities for global research into children's palliative care: results of an International Delphi Study. *BMC Palliat Care*. 2015;14:36
6. Harding R, Wolfe J, Baker JN. Outcome measurement for children and young people. *J Palliat Med*. 2017; 20(4):313
7. Voyles E. The development and outcomes of a pediatric palliative care program: a quality improvement process. *J Pediatr Nurs*. 2013;28(2):196–199
8. Baker JN, Levine DR, Hinds PS, et al. Research priorities in pediatric palliative care. *J Pediatr*. 2015;167(2):467–470.e3
9. Hearn J, Higginson IJ. Outcome measures in palliative care for advanced cancer patients: a review. *J Public Health Med*. 1997;19(2): 193–199
10. Bausewein C, Daveson BA, Currow DC, et al. EAPC White Paper on outcome measurement in palliative care: improving practice, attaining outcomes and delivering quality services - recommendations from the European Association for Palliative Care (EAPC) task force on outcome measurement. *Palliat Med*. 2016;30(1):6–22
11. Araújo J, Dourado M, Ferreira PL. Measuring instruments of the quality of life pediatric palliative care [in Portuguese]. *Acta Med Port*. 2015;28(4):501–512
12. Knapp C, Madden V. Conducting outcomes research in pediatric palliative care. *Am J Hosp Palliat Care*. 2010;27(4):277–281
13. Coombes LH, Wiseman T, Lucas G, Sangha A, Murtagh FE. Health-related quality-of-life outcome measures in paediatric palliative care: a systematic review of psychometric properties and feasibility of use. *Palliat Med*. 2016;30(10):935–949
14. Huang IC, Shenkman EA, Madden VL, Vadaparampil S, Quinn G, Knapp CA. Measuring quality of life in pediatric palliative care: challenges and potential solutions. *Palliat Med*. 2010;24(2):175–182
15. Fayers P, Machin D. *Quality of Life: The Assessment, Analysis and Interpretation of Patient-Reported Outcomes*, 2nd ed. Hoboken, NJ: Wiley-Blackwell; 2008
16. Walters SJ. *Quality of Life Outcomes in Clinical Trials and Health-Care Evaluation: A Practical Guide to Analysis and Interpretation*, 1st ed. Chichester, United Kingdom: John Wiley & Sons; 2009
17. de Vet HCW, Terwee CB, Mokkink LB, Knol DL. *Measurement in Medicine: A Practical Guide*, 1st ed. Cambridge, United Kingdom: Cambridge University Press; 2011
18. Moher D, Liberati A, Tetzlaff J, Altman DG; PRISMA Group. Preferred Reporting Items for Systematic Reviews and Meta-analyses: the PRISMA statement. *PLoS Med*. 2009;6(7):e1000097
19. Craig F, Abu-Saad Huijer H, Benini F, et al. IMPaCCT: standards for paediatric palliative care in Europe. *Eur J Palliat Care*. 2007;14(3):109–114
20. Hain R, Devins M, Hastings R, Noyes J. Paediatric palliative care: development and pilot study of a 'directory' of life-limiting conditions. *BMC Palliat Care*. 2013;12(1):43
21. Kmet LM, Lee RC, Cook LS. *Standard Quality Assessment Criteria for Evaluating Primary Research Papers From a Variety of Fields*. Edmonton, Canada: Alberta Heritage Foundation for Medical Research; 2004. Available at: https://www.ihe.ca/advanced-search/standard-quality-assessment-criteria-for-evaluating-primary-research-papers-from-a-variety-of-fields. Accessed May 25, 2018
22. Norman G, Streiner D. *Health Measurement Scales: A Practical Guide to Their Development and Use*, 2nd ed. Oxford, United Kingdom: Oxford University Press; 2008
23. Akard TF, Dietrich MS, Friedman DL, et al. Digital storytelling: an innovative legacy-making intervention for children with cancer. *Pediatr Blood Cancer*. 2015;62(4):658–665
24. Al-Gharib RM, Abu-Saad Huijer H, Darwish H. Quality of care and relationships as reported by children with cancer and their parents. *Ann Palliat Med*. 2015;4(1):22–31
25. Arland LC, Hendricks-Ferguson VL, Pearson J, Foreman NK, Madden JR. Development of an in-home standardized end-of-life treatment program for pediatric patients dying of brain tumors. *J Spec Pediatr Nurs*. 2013;18(2):144–157
26. Bradford N, Young J, Armfield NR, et al. A pilot study of the effectiveness of home teleconsultations in paediatric palliative care. *J Telemed Telecare*. 2012;18(8):438–442
27. Conte T, Mitton C, Erdelyi S, Chavoshi N, Siden H. Pediatric palliative care program versus usual care and healthcare resource utilization in British Columbia: a matched-pairs cohort study. *J Palliat Med*. 2016;19(11):1218–1223
28. Eaton N. 'I don't know how we coped before': a study of respite care for children in the home and hospice. *J Clin Nurs*. 2008;17(23):3196–3204
29. Friedrichsdorf SJ, Postier A, Dreyfus J, Osenga K, Sencer S, Wolfe J. Improved quality of life at end of life related to home-based palliative care in children with cancer. *J Palliat Med*. 2015;18(2):143–150
30. Gans D, Hadler MW, Chen X, et al. Impact of a pediatric palliative care program on the caregiver experience. *J Hosp Palliat Nurs*. 2015;17(6):559–565
31. Groh G, Borasio GD, Nickolay C, Bender HU, von Lüttichau I, Führer M. Specialized pediatric palliative home care: a prospective evaluation. *J Palliat Med*. 2013;16(12):1588–1594
32. Harris N, Beringer A, Fletcher M. Families' priorities in life-limiting illness: improving quality with online empowerment. *Arch Dis Child*. 2016;101(3):247–252
33. Lyon ME, Jacobs S, Briggs L, Cheng YI, Wang J. Family-centered advance care planning for teens with cancer. *JAMA Pediatr*. 2013;167(5):460–467
34. Lyon ME, Jacobs S, Briggs L, Cheng YI, Wang J. A longitudinal, randomized, controlled trial of advance care planning for teens with cancer: anxiety, depression, quality of life, advance directives, spirituality. *J Adolesc Health*. 2014;54(6):710–717

35. Remedios C, Willenberg L, Zordan R, Murphy A, Hessel G, Philip J. A pre-test and post-test study of the physical and psychological effects of out-of-home respite care on caregivers of children with life-threatening conditions. *Palliat Med*. 2015;29(3):223–230

36. Schmidt P, Otto M, Hechler T, Metzing S, Wolfe J, Zernikow B. Did increased availability of pediatric palliative care lead to improved palliative care outcomes in children with cancer? *J Palliat Med*. 2013;16(9):1034–1039

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

38. van der Geest IM, Darlington AS, Streng IC, Michiels EM, Pieters R, van den Heuvel-Eibrink MM. Parents' experiences of pediatric palliative care and the impact on long-term parental grief. *J Pain Symptom Manage*. 2014;47(6):1043–1053

39. Vern-Gross TZ, Lam CG, Graff Z, et al. Patterns of end-of-life care in children with advanced solid tumor malignancies enrolled on a palliative care service. *J Pain Symptom Manage*. 2015;50(3):305–312

40. Vollenbroich R, Duroux A, Grasser M, Brandstätter M, Borasio GD, Führer M. Effectiveness of a pediatric palliative home care team as experienced by parents and health care professionals. *J Palliat Med*. 2012;15(3):294–300

41. Wolfe J, Orellana L, Cook EF, et al. Improving the care of children with advanced cancer by using an electronic patient-reported feedback intervention: results from the PediQUEST randomized controlled trial. *J Clin Oncol*. 2014;32(11):1119–1226

42. Varni JW, Seid M, Kurtin PS. PedsQL 4.0: reliability and validity of the Pediatric Quality of Life Inventory version 4.0 generic core scales in healthy and patient populations. *Med Care*. 2001;39(8):800–812

43. Varni JW, Burwinkle TM, Seid M. The PedsQL as a pediatric patient-reported outcome: reliability and validity of the PedsQL Measurement Model in 25,000 children. *Expert Rev Pharmacoecon Outcomes Res*. 2005;5(6):705–719

44. Emanuel LL, Alpert HR, Emanuel EE. Concise screening questions for clinical assessments of terminal care: the needs near the end-of-life care screening tool. *J Palliat Med*. 2001;4(4):465–474

45. Cohen R, Leis AM, Kuhl D, Charbonneau C, Ritvo P, Ashbury FD. QOLLTI-F: measuring family carer quality of life. *Palliat Med*. 2006;20(8):755–767

46. Wolfe J, Grier HE, Klar N, et al. Symptoms and suffering at the end of life in children with cancer. *N Engl J Med*. 2000;342(5):326–333

47. Hechler T, Blankenburg M, Friedrichsdorf SJ, et al. Parents' perspective on symptoms, quality of life, characteristics of death and end-of-life decisions for children dying from cancer. *Klin Padiatr*. 2008;220(3):166–174

48. Mack JW, Hilden JM, Watterson J, et al. Parent and physician perspectives on quality of care at the end of life in children with cancer. *J Clin Oncol*. 2005;23(36):9155–9161

49. Mack JW, Joffe S, Hilden JM, et al. Parents' views of cancer-directed therapy for children with no realistic chance for cure. *J Clin Oncol*. 2008;26(29):4759–4764

50. Edwards KE, Neville BA, Cook EF Jr, Aldridge SH, Dussel V, Wolfe J. Understanding of prognosis and goals of care among couples whose child died of cancer. *J Clin Oncol*. 2008;26(8):1310–1315

51. Dussel V, Kreicbergs U, Hilden JM, et al. Looking beyond where children die: determinants and effects of planning a child's location of death. *J Pain Symptom Manage*. 2009;37(1): 33–43

52. Gräßel E. *Home Care Scale for the Assessment of Subjective Burden of Mentoring or Nurturing Persons*. Ebersberg, Germany: Vless Verlag; 2001

53. Gräßel E, Chiu T, Oliver R. *Development and Validation of the Burden Scale for Family Caregivers (BSFC)*. Toronto, Canada: COTA: Comprehensive Rehabilitation and Mental Health Services; 2003

54. Snaith RP. The Hospital Anxiety and Depression Scale. *Br J Gen Pract*. 1990;40(336):305

55. Herrmann-Lingen C, Buss U, Snaith RP. *Hospital Anxiety and Depression Scale – German Version*. Berne, Switzerland: Verlag Hans Huber; 2011

56. Harris N. MyQuality. 2015. Available at: http://my-quality.net. Accessed May 24, 2018

57. Harris N, Beringer A, Fletcher M. Promoting patients' priorities through user-generated quality of life measures. 2014. Available at: https://www.health.org.uk/programmes/shine-2012/projects/promoting-patients'-priorities-through-user-generated-quality-life. Accessed October 21, 2017

58. Koren PE, DeChillo N, Friesen BJ. Measuring empowerment in families whose children have emotional disabilities: a brief questionnaire. *Rehabil Psychol*. 1992;37(4):305–321

59. Nachshen JS. Empowerment and families: building bridges between parents and professionals, theory and research. *J Dev Disabil*. 2005;11(1):67–76

60. Herbert RJ, Gagnon AJ, Rennick JE, O'Loughlin JL. A systematic review of questionnaires measuring health-related empowerment. *Res Theory Nurs Pract*. 2009;23(2):107–132

61. Kirchhoff KT, Hammes BJ, Kehl KA, Briggs LA, Brown RL. Effect of a disease-specific planning intervention on surrogate understanding of patient goals for future medical treatment. *J Am Geriatr Soc*. 2010;58(7):1233–1240

62. O'Connor AM. Validation of a decisional conflict scale. *Med Decis Making*. 1995;15(1):25–30

63. Engelberg R, Downey L, Curtis JR. Psychometric characteristics of a quality of communication questionnaire assessing communication about end-of-life care. *J Palliat Med*. 2006;9(5):1086–1098

64. Beck AT, Steer RA. *Beck Anxiety Inventory Manual*. San Antonio, TX: Psychological Corporation, Harcourt Brace & Company; 1993

65. Beck AT, Steer RA, Brown GK. *Beck Depression Inventory Manual*. 2nd ed. San Antonio, TX: Psychological Corporation, Harcourt Brace & Company; 1996

66. Varni JW, Burwinkle TM, Seid M, Skarr D. The PedsQL 4.0 as a pediatric

population health measure: feasibility, reliability, and validity. *Ambul Pediatr*. 2003;3(6):329–341

67. Peterman AH, Fitchett G, Brady MJ, Hernandez L, Cella D. Measuring spiritual well-being in people with cancer: the Functional Assessment of Chronic Illness Therapy–Spiritual Well-Being Scale (FACIT-Sp). *Ann Behav Med*. 2002;24(1):49–58
68. Ware JE, Kosinski M, Keller SD. *SF-36 Physical and Mental Health Summary Scales: A User's Manual*. Boston, MA: The Health Institute, New England Medical Center; 1994
69. Goldberg DP, Gater R, Sartorius N, et al. The validity of two versions of the GHQ in the WHO study of mental illness in general health care. *Psychol Med*. 1997;27(1):191–197
70. Chalder T, Berelowitz G, Pawlikowska T, et al. Development of a fatigue scale. *J Psychosom Res*. 1993;37(2):147–153
71. Welch V, Hatton C, Emerson E, et al. Do short break and respite services for families with a disabled child in England make a difference to siblings? A qualitative analysis of sibling and parent responses. *Child Youth Serv Rev*. 2012;34(2):451–459
72. Zimet GD, Powell SS, Farley GK, Werkman S, Berkoff KA. Psychometric characteristics of the multidimensional scale of perceived social support. *J Pers Assess*. 1990;55(3–4):610–617
73. Boelen PA, Van Den Bout J, De Keijser J, Hoijtink H. Reliability and validity of the Dutch version of the inventory of traumatic grief (ITG). *Death Stud*. 2003;27(3):227–247
74. Prigerson HG, Vanderwerker LC, Maciejewski PK. A case for the inclusion of prolonged grief disorder in DSM-V. In: Stroebe M, Hansson R, Schut H, eds. *Handbook of Bereavement Research and Practice: 21st Century Perspectives*. Washington, DC: American Psychological Association Press; 2008:165–186
75. Collins JJ, Devine TD, Dick GS, et al. The measurement of symptoms in young children with cancer: the validation of the Memorial Symptom Assessment Scale in children aged 7-12. *J Pain Symptom Manage*. 2002;23(1):10–16
76. Conte T, Mitton C, Trenaman LM, Chavoshi N, Siden H. Effect of pediatric palliative care programs on health care resource utilization and costs among children with life-threatening conditions: a systematic review of comparative studies. *CMAJ Open*. 2015;3(1):E68–E75
77. Wolfe J, Siden H. Research in paediatric palliative care. In: Goldman A, Hain R, Liben S, eds. *Oxford Textbook of Palliative Care for Children*. Oxford, United Kingdom: Oxford University Press; 2012
78. Rahimzadeh V, Bartlett G, Longo C, et al. Promoting an ethic of engagement in pediatric palliative care research. *BMC Palliat Care*. 2015;14:50
79. Silva N, Crespo C, Carona C, Bullinger M, Canavarro MC. Why the (dis) agreement? Family context and child-parent perspectives on health-related quality of life and psychological problems in paediatric asthma. *Child Care Health Dev*. 2015;41(1):112–121
80. Lim Y, Velozo C, Bendixen RM. The level of agreement between child self-reports and parent proxy-reports of health-related quality of life in boys with Duchenne muscular dystrophy. *Qual Life Res*. 2014;23(7):1945–1952
81. Sattoe JN, van Staa A, Moll HA; On Your Own Feet Research Group. The proxy problem anatomized: child-parent disagreement in health related quality of life reports of chronically ill adolescents. *Health Qual Life Outcomes*. 2012;10:10
82. Vetter TR, Bridgewater CL, McGwin G Jr. An observational study of patient versus parental perceptions of health-related quality of life in children and adolescents with a chronic pain condition: who should the clinician believe? *Health Qual Life Outcomes*. 2012;10:85
83. Daveson BA, Simon ST, Benalia H, et al; PRISMA. Are we heading in the same direction? European and African doctors' and nurses' views and experiences regarding outcome measurement in palliative care. *Palliat Med*. 2012;26(3):242–249
84. Craig P, Dieppe P, Macintyre S, Michie S, Nazareth I, Petticrew M; Medical Research Council Guidance. Developing and evaluating complex interventions: the new Medical Research Council guidance. *BMJ*. 2008;337:a1655
85. Higginson IJ, Simon ST, Benalia H, et al; PRISMA. Which questions of two commonly used multidimensional palliative care patient reported outcome measures are most useful? Results from the European and African PRISMA survey. *BMJ Support Palliat Care*. 2012;2(1):36–42
86. Dussel V, Medin G. Pediatric issues. In: Bruera E, De Lima L, Wenk R, Farr W, eds. *Palliative Care in the Developing World: Principles and Practice*. Houston, TX: IAHPC Press; 2004
87. Wiering B, de Boer D, Delnoij D. Patient involvement in the development of patient-reported outcome measures: the developers' perspective. *BMC Health Serv Res*. 2017;17(1):635
88. Kane JR, Baker JN. Quality improvement. In: Goldman A, Hain R, Liben S, eds. *Oxford Textbook of Palliative Care for Children*. Oxford, United Kingdom: Oxford University Press; 2012:430–440
89. Patrick DL, Deyo RA. Generic and disease-specific measures in assessing health status and quality of life. *Med Care*. 1989;27(suppl 3):S217–S232
90. Janssens A, Thompson Coon J, Rogers M, et al. A systematic review of generic multidimensional patient-reported outcome measures for children, part I: descriptive characteristics. *Value Health*. 2015;18(2):315–333
91. Janssens A, Rogers M, Thompson Coon J, et al. A systematic review of generic multidimensional patient-reported outcome measures for children, part II: evaluation of psychometric performance of English-language versions in a general population. *Value Health*. 2015;18(2):334–345
92. Hickey AM, Bury G, O'Boyle CA, Bradley F, O'Kelly FD, Shannon W. A new short form individual quality of life measure (SEIQoL-DW): application in a cohort of individuals with HIV/AIDS. *BMJ*. 1996;313(7048):29–33
93. Wolpert M. Do patient reported outcome measures do more harm than good? *BMJ*. 2013;346:f2669

94. Griffiths C, Armstrong-James L, White P, Rumsey N, Pleat J, Harcourt D. A systematic review of patient reported outcome measures (PROMs) used in child and adolescent burn research. *Burns*. 2015;41(2):212–224

95. Tadić V, Rahi JS. One size doesn't fit all: time to revisit patient-reported outcome measures (PROMs) in paediatric ophthalmology? *Eye (Lond)*. 2017;31(4):511–518

96. Downing J, Ojing M, Powell RA, et al. A palliative care outcome measure for children in sub-Saharan Africa: early development findings. *Eur J Palliat Care*. 2012;19(6): 292–294

97. Downing J. Global challenges? Research in low-income settings – palliative care experiences. 2017. Available at: https://dtu-panopto.lancs.ac.uk/Panopto/Pages/Viewer.aspx?id=505d0b14-129f-4b64-9d06-408abc60bf2c. Accessed May 20, 2018

98. van Leeuwen M, Husson O, Alberti P, et al; EORTC QLG. Understanding the quality of life (QOL) issues in survivors of cancer: towards the development of an EORTC QOL cancer survivorship questionnaire. *Health Qual Life Outcomes*. 2018;16(1):114

99. Evans CJ, Benalia H, Preston NJ, et al; MORECare. The selection and use of outcome measures in palliative and end-of-life care research: the MORECare International Consensus Workshop. *J Pain Symptom Manage*. 2013;46(6):925–937

100. Antunes B, Harding R, Higginson IJ; EUROIMPACT. Implementing patient-reported outcome measures in palliative care clinical practice: a systematic review of facilitators and barriers. *Palliat Med*. 2014;28(2):158–175

101. Holtrop JS, Rabin BA, Glasgow RE. Dissemination and implementation science in primary care research and practice: contributions and opportunities. *J Am Board Fam Med*. 2018;31(3):466–478

102. Bausewein C, Simon ST, Benalia H, et al; PRISMA. Implementing patient reported outcome measures (PROMs) in palliative care–users' cry for help. *Health Qual Life Outcomes*. 2011;9(9):27

103. Wells S, Tamir O, Gray J, Naidoo D, Bekhit M, Goldmann D. Are quality improvement collaboratives effective? A systematic review. *BMJ Qual Saf*. 2018;27(3):226–240

104. Drahota A, Meza RD, Brikho B, et al. Community-academic partnerships: a systematic review of the state of the literature and recommendations for future research. *Milbank Q*. 2016;94(1):163–214

105. Recinos PF, Dunphy CJ, Thompson N, Schuschu J, Urchek JL III, Katzan IL. Patient satisfaction with collection of patient-reported outcome measures in routine care. *Adv Ther*. 2017;34(2):452–465