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Specialist paediatric palliative care for children and young people with cancer: A mixed-methods systematic review

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Abstract

Background: Specialist paediatric palliative care services are promoted as an important component of palliative care provision, but there is uncertainty about their role for children with cancer.

Aim: To examine the impact of specialist paediatric palliative care for children and young people with cancer and explore factors affecting access.

Design: A mixed-methods systematic review and narrative synthesis (PROSPERO Registration No. CRD42017064874).

Data sources: Database (CINAHL, Cochrane Database of Systematic Reviews, Embase, MEDLINE, PsycINFO) searches (2000–2019) identified primary studies of any design exploring the impact of and/or factors affecting access to specialist paediatric palliative care. Study quality was assessed using The Mixed Methods Appraisal Tool.

Results: An evidence base of mainly low- and moderate-quality studies ($n = 42$) shows that accessing specialist paediatric palliative care is associated with less intensive care at the end of life, more advance care planning and fewer in-hospital deaths. Current evidence cannot tell us whether these services improve children's symptom burden or quality of life. Nine studies reporting provider or family views identified uncertainties about what specialist paediatric palliative care offers, concerns about involving a new team, association of palliative care with end of life and indecision about when to introduce palliative care as important barriers to access. There was evidence that children with haematological malignancies are less likely to access these services.

Conclusion: Current evidence suggests that children and young people with cancer receiving specialist palliative care are cared for differently. However, little is understood about children's views, and research is needed to determine whether specialist input improves quality of life.

Keywords

Palliative care, terminal care, hospice care, child, adolescent, neoplasms, systematic review

What is already known about the topic?

- Specialist paediatric palliative care is promoted as an important component of children and young adult cancer services, but there is uncertainty about the factors that affect access and the benefits for children who receive this specialist input.
- Three reviews, which have aggregated evidence for children with all life-limiting conditions, suggest that the benefits of specialist palliative care include less time in hospital and improvements in quality of life and symptom management.
- The growing number of studies investigating the role of specialist palliative care for children with cancer report mixed results and varying provision, and there is a need to aggregate this evidence to inform future policy and practice.

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What this paper adds?

- Accessing specialist paediatric palliative care is associated with less intensive care at the end of life, more advance care planning and fewer in-hospital deaths for children and young people with cancer, but there is no robust evidence to tell us whether these services lead to improvements in quality of life or symptom management.
- Children with haematological malignancies are less likely to receive specialist palliative care than children with other cancers.
- Uncertainty about when to introduce palliative care services to families, what it comprises and the added value of specialist input was identified as a key barrier to access, as were perceptions that paediatric oncology teams already meet the palliative care needs of their patients.

Implications for practice, theory or policy

- Evidence is still needed to determine whether specialist paediatric palliative care improves the quality of life and symptom management for children and young people with cancer.
- Exploration of why children with certain cancers are less likely to receive specialist palliative care at the end of life may help address this inequality in access.
- A core outcome set study including the views of children and families would help improve future aggregation of evidence in this area.

Background

Approximately 20% of children and young people diagnosed with cancer do not survive despite significant medical advances in recent decades.[1,2] The majority of deaths are due to the malignancy, with some attributable to anticancer treatments.[3] Distress from symptoms and suffering during the end-of-life phase can be significant,[4–6] impacting on the child and their family's quality of life.[7,8] In addition, many children and young people who die from cancer continue to have high-intensity treatments towards the end of life,[9,10] with nearly half dying in the acute care setting,[11] despite preferences from the majority of children and their parents for being at home during the end-of-life phase.[12,13]

Palliative care, defined by the World Health Organization (WHO) as 'the active total care of the child's body, mind and spirit . . . [that] begins when illness is diagnosed, and continues regardless of whether or not a child receives treatment directed at the disease',[14] is recognised as an important component of children and young people's cancer services.[15,16] In addition, and over the last 30 years, specialist paediatric palliative care services for children and young people have been developing in many countries including the United States, Canada, United Kingdom and across Europe.[17] The English National Health Service (NHS) defines specialist paediatric palliative care as 'a consultant-led multi-professional specialist palliative care team . . . led by a medical consultant working at Paediatric Palliative Care Competency Level 4'.[16] In practice, however, the models of providing specialist palliative care vary within and between countries, including, for example, hospital- and community-based teams which support all children with life-limiting conditions,[18] teams embedded within paediatric oncology departments, joint working with, or hospice-led provision, and specialist nurse-led teams as well as services led by a paediatric palliative care consultant.[17,19,20]

Even within the developed world, the availability, referral and uptake of specialist palliative care among children and young people with cancer remains low and variable between and within countries and settings, and it is not clear to what extent these services are addressing all aspects of palliative care as defined by WHO.[17,21–25] For children with cancer, referral to palliative care also often occurs late in the trajectory of illness, sometimes only days before death.[26,27] Recent systematic reviews suggest that access to specialist palliative care services is associated with improvements in quality of life, symptom control, perceived support, reduced time in hospital, less invasive treatment and greater advance care planning.[19,28,29] However, these reviews have aggregated the results for children and young people across conditions, and the evidence for those with cancer remains unclear because of conflicting results between individual studies[28] and the lack of work exploring condition-specific factors that may influence access to and benefit from specialist palliative care services.[30,31]

A rigorous review of the evidence on the impacts of specialist paediatric palliative care for children and young people with cancer and their families is both crucial to informing debates within paediatric oncology regarding the positioning and role of these specialist services and for future service development. This mixed-methods systematic review synthesises the existing evidence on the benefits, drawbacks, facilitators and barriers associated with referral to and uptake of specialist paediatric palliative care for children and young people with cancer and their families.

Methods

The review questions are as follows:

1. What are the reported benefits and drawbacks of referral to specialist paediatric palliative care for children and young people with cancer and their families?
2. What are the factors (e.g. barriers, facilitators) affecting referral to and uptake of specialist paediatric palliative care for children and young people with cancer?

The protocol registration number is CRD42017064874.[32]

Eligibility criteria

Primary studies of any design (e.g. experimental, observational, surveys, consensus and qualitative studies) examining either the impact of or factors affecting specialist paediatric palliative care access for children and young people (age 0–24 years) with cancer were included. Studies of a mixed population were included if (1) the majority of the participants were children and young people with cancer and/or (2) data were reported separately.

We defined specialist paediatric palliative care as care provided by multidisciplinary teams or palliative care services which included clinicians (e.g. oncologists, paediatricians, nurses) with paediatric palliative care training, or services who self-identified as providing specialist paediatric palliative care. We included services delivered in different settings (e.g. inpatient, community or home settings) and both liaison services (e.g. supporting the child's usual care team) and services directly supporting children and their families. Although broad, this reflects the varying provision of specialist palliative care for children within and between different countries and so enabled us to synthesise the evidence about these specialist services.

To understand the different perspectives on referral to specialist palliative care services, studies that included the following participant groups were eligible: children and young people; parents (including bereaved); other family members and health and social care staff. No comparator was required.

We excluded case studies, review articles, descriptive, theoretical or clinical opinion articles, conference abstracts and articles not published in the English language. We also restricted the eligibility to studies conducted in high-income countries (defined as OECD (Organisation for Economic Co-operation and Development)[33] member countries) because of the very different healthcare infrastructure and status of specialist paediatric palliative care in developing countries.[17]

Search strategy

Electronic databases (CINAHL, Cochrane Database of Systematic Reviews (CDSR), Embase, MEDLINE, PsycINFO) were searched on 27 June 2017 (from 2000, in line with increasing availability of specialist paediatric palliative care services internationally[19]). The search strategy consisted of terms and synonyms for [malignancies] AND [children] AND [specialist paediatric palliative care] (see Supplementary Information Appendix A.) Reference lists of included studies and relevant literature reviews were checked, and backward and forward citation searching of included studies and PubMed-related articles link searches were undertaken. An update search in MEDLINE (which identified all the eligible studies in the original search) was performed on 13 September 2019.

Study selection

Titles and abstracts were screened in Covidence,[34] and relevant full-text articles were retrieved and independently assessed for eligibility by two reviewers with disagreements resolved via a third reviewer.[35] Reasons for exclusion at full text were recorded.

Data extraction

Data on study characteristics, methods, study focus (e.g. impact and/or factors affecting access) and quantitative outcome data were extracted into Microsoft Excel using a pre-piloted data extraction template. Qualitative data, including author-reported results, direct quotations and results tables, were imported into NVivo version 11[36] for analysis.

Data extraction was carried out by one reviewer and checked by a second.

Critical appraisal

The Mixed Methods Appraisal Tool (MMAT)[37,38] was used to appraise the methodological quality of all included studies. MMAT comprises two generic screening questions and an additional four criteria for use with specific study designs. Criterion assessments (e.g. Is the sample representative of the population under study?) are categorised 'Yes', 'No' or 'Can't tell'. An overall quality score (of 0%, 25%, 50% or 100%) based on how many study design specific criterion were met (those categorised as 'Yes') was calculated for each study.

The quality assessment was undertaken independently by two reviewers and informed the synthesis methods and reporting of the review results, along with identifying needs for future research. We did not exclude studies based on quality assessment.

Data synthesis

An integrative narrative synthesis was planned drawing on interpretative review methodology[39] with thematic analysis as the principal method of synthesis.[40] The synthesis plan outlined in the protocol[32] was modified following an assessment of the potential for aggregation, configuration and integration of study findings. The final method involved separate syntheses of data reporting the impact of specialist paediatric palliative care and factors affecting access (both included quantitative and qualitative data). Each comprised the following steps:

Data reduction. This involved reviewing and summarising extracted data and identifying recurring categories from across the studies, and distinct service types. Each synthesis included quantitative and qualitative data and all numerical and statistical findings were converted to descriptive summaries. For qualitative data, 'meaning units' (comprehensible segments of text which contain one idea or piece of information) were identified and named (or 'coded') drawing both on author-reported results and participant quotes to capture the full range of concepts or themes across the studies.[41]

Data display. Using the method of constant comparison,[39] descriptive summaries of quantitative data and coded qualitative data were compared to ensure that similar data were grouped together to develop a thematic coding matrix consisting of descriptive themes and overarching categories which grouped similar themes together. This was performed by one reviewer with regular input from the wider review team for sense checking and validation. Data from each study were then synthesised into the coding matrix, retaining reference to the service type and critical appraisal score to facilitate greater interpretation. Data display techniques were used to illustrate the spread of themes across studies and specialist paediatric palliative care models, and narrative weaving describes the results of each synthesis.[42]

Results

A total of 8549 unique records were screened by title and abstract, 626 full-text articles were retrieved and reviewed, and 49 articles describing 42 studies[23,43–90] were included in the review (see Figure 1).

Study characteristics

Of the 42 included studies, 11 examined the impact of specialist paediatric palliative care,[44,48,57,58,62,64,66,70,73,74,88] 14 explored factors affecting access[23,43,45,56,65,68,71,75,76,78–83,89] and 17 studies investigated both[46,47,49–55,59–61,63,67,69,72,77,84–87,90] (see Table 1 for study characteristics). Using the MMAT, 25 studies were categorised as quantitative non-randomised, 12 as quantitative descriptive and 5 as qualitative studies. The majority of studies were conducted in the United States ($n = 28$). Others were in the United Kingdom ($n = 3$),[23,47,76] Canada ($n = 3$),[50,51,63,72] Germany ($n = 3$),[57,73,90] France ($n = 1$),[87] Switzerland ($n = 1$),[79] Israel ($n = 1$)[49] and two in multiple countries.[65,71]

Study populations

Thirty-one studies (all quantitative) examined the impact of and/or characteristics of children receiving specialist palliative care; 24 included a comparator group of children not receiving this,[23,43–51,54–64,78,86–88,90] 1 compared children receiving late and early specialist input,[52,53,84,85] and 6 used a single-group study design.[66,67,69,70,73,74] Of the remaining 11 studies (6 quantitative and 5 qualitative), 10 explored the views of healthcare staff[65,71,72,75–77,79–83,89] and 1 the views of parents and young people.[68]

Of the 31 studies examining outcomes and/or characteristics of children, the majority ($n = 21$) drew their sample of children and young people from a single centre. Several studies used the same or potentially overlapping samples as other included studies (see Figure 2).

In total, data for 7933 children and young people, 4289 of whom had received specialist palliative care compared to 3644 who did not, were included. While the majority ($n = 23$) included children and young people with any cancer, eight studies concerned children and young people with particular diagnoses or treatments (see Table 1 for details).[44,56,61,62,66,67,69,78] Overall, these 31 studies address diagnoses from infancy to young adulthood, with only three studies focused on young adults, which included any malignancy.[54,58,86] Five studies included some children and young people with conditions other than cancer.[61,67,70,73,88]

Of the 11 studies exploring stakeholder views, 3 recruited paediatric oncology staff from single hospitals[75,77,82,83] and 1 from multiple hospitals,[79,80] 1 recruited staff involved in providing palliative care to children with cancer from primary, tertiary and community settings,[76] 4 recruited paediatric oncologists via professional organisations[65,71,72,81] and 1 recruited parent and young person dyads from three hospitals.[68] In total, these studies represented the views of 1133 physicians, 986 other healthcare professionals (mainly nurses, social workers and other staff working in paediatric oncology, but also in palliative care and other settings), 129 parents and 129 young people.

Models of delivery

We identified five broad service types from the included studies: hospital-based palliative care teams with referral triggering an initial consultation[46,50–53,58,61–64,67,69,73–75,77,82–85,87,88] ($n = 17$), hospice services[23,45,47,55,56,59,60,65,86] ($n = 8$), home-based services[48,57,70,90] ($n = 4$), integrated oncology

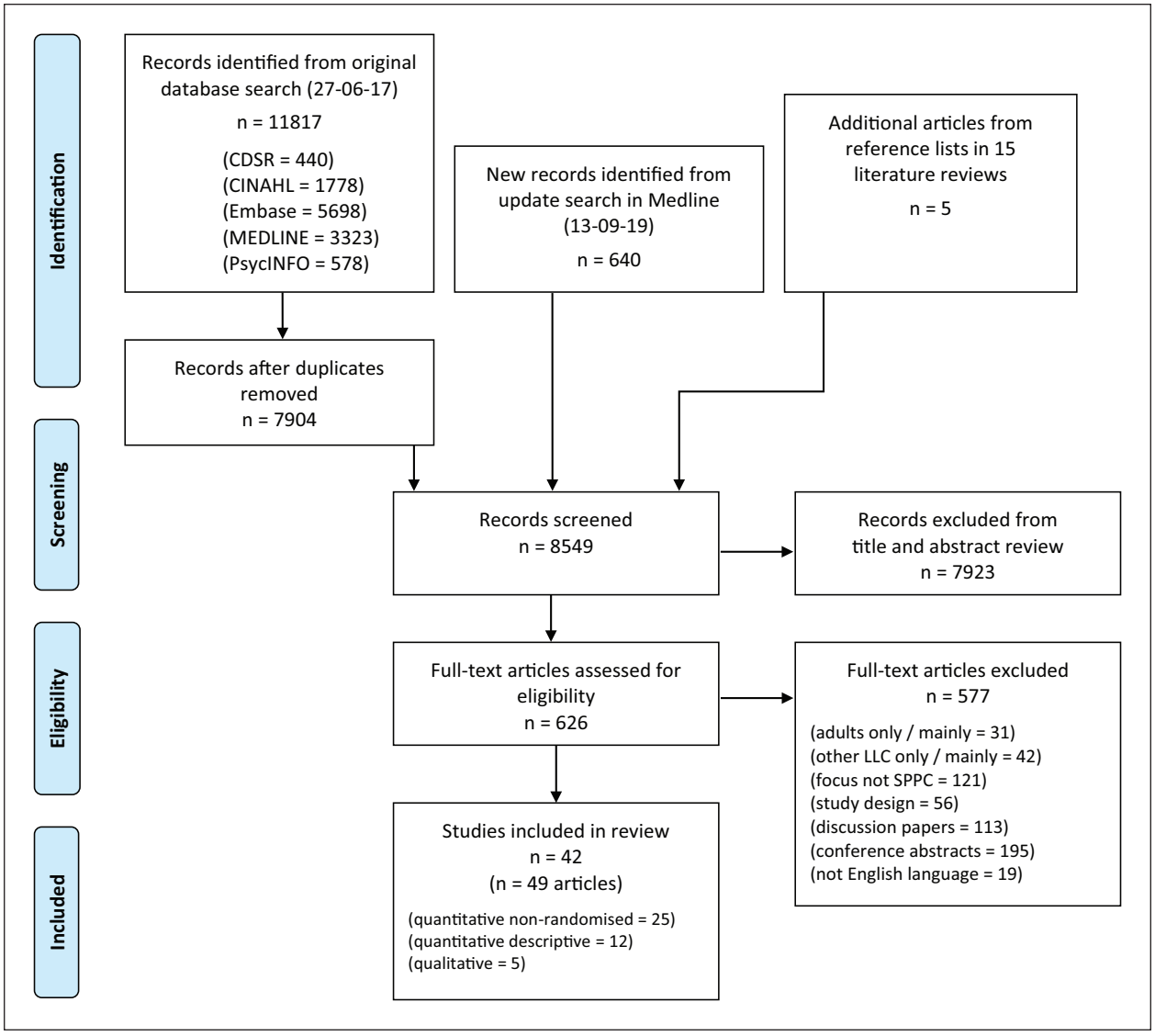


Figure 1. Study flow diagram.

services[44,66] (*n* = 2) and an integrated oncology inpatient unit[49] (*n* = 1). Three studies referred to palliative care consultation only,[43,54,78] and three a palliative care team,[68,79,81] although it is likely that these were all referring to hospital-based teams. Of the remaining four studies, three included a range of models[71,72,89] and one referred to specialist paediatric palliative care explicitly but did not define this.[76]

Very few studies provided specific details about the team or service providing palliative care in terms of skills mix, role and the extent of provision. There were also some anomalies between studies; for example, five studies used hospice discussions or enrolment as an outcome of specialist palliative care,[46,61,62,64,90] whereas in other studies hospices were defined as the source of specialist intervention. Not all studies investigating hospice settings specified the characteristics (e.g. adult vs children's hospice), and it was difficult to determine what 'home-based services' might comprise. Among the studies exploring stakeholder views, three offered only 'hypothetical models' for participants to consider[68,75,76] and three focused primarily on views about early integration of specialist palliative care, which was defined as close to the time of diagnosis.[68,75,81]

Only two studies accounted for the timing of the initial palliative care consult/enrolment when deciding which children to classify as having received specialist intervention; one only included children who had received specialist palliative care for more than 30 days[63] and the second for more than 1 day.[54] A third study, which compared early

Table 1. Characteristics of included studies.

Study ID and country	Population and sample size	Setting	SPPC description	Study aim	Overview of relevant methods	Study investigated the following:	
						SPPC outcomes	Factors affecting SPPC referral/uptake
Quantitative non-randomised (n = 25)							
Ananth et al.[43] United States	Paediatric oncology patients who died *n* = 125	One children's cancer and blood disorder centre	Paediatric palliative care consultation	To examine healthcare utilisation and palliative care delivery for children with cancer by enrolment in early phase clinical trial	Retrospective cohort study comparing receipt of SPPC between children enrolled (*n* = 33) and not enrolled (*n* = 92) in an early phase clinical trial	×	Enrolment in early phase clinical trial
Arland et al.[44] United States	Paediatric patients (age 1 month–19 years) who died of a brain tumour *n* = 114	One children's hospital	End-of-life programme including advance care planning, symptom management, family liaison and home care	To examine if a relationship existed between patient outcomes before and after implementation of SPPC	Retrospective chart review comparing children who received SPPC (*n* = 92) with a historical cohort (*n* = 22) prior to SPPC implementation	Hospital admissions (number, duration of stay) Location of death	×
Baker et al.[45] United States	Children and young people who died from cancer (at age < 22) *n* = 345	One children's research hospital	Hospice enrolment	To assess whether race is associated with end-of-life care provision	Retrospective chart review to assess differences in race between children enrolled (*n* = 146) and not enrolled (*n* = 199) in SPPC	×	Race
Brock et al.[46] United States	Haematology, oncology and stem cell transplant patients who died (age 0–35) *n* = 445	The paediatric haematology, oncology and stem cell treatment divisions in one children's hospital	Specialty palliative care consultation and hospice enrolment	To explore the associations between demographic variables and end-of-life care characteristics	Retrospective analysis of a prospectively collected database to compare 410 patients who received (*n* = 69) and did not receive SPPC (*n* = 341) (missing data *n* = 35)	Hospice enrolment Location of death DNR order POLST	Race and ethnicity Language Religion Diagnosis Prognosis DNR order and timing Treatment intensity
Cuviello et al.[78] United States	Paediatric and young adult patients with cancer enrolled in a phase 1 oncology trial *n* = 149	National Cancer Institute (NCI) Paediatric Oncology Branch's phase 1 clinical trials	Palliative care consultation	To investigate factors influencing utilisation of SPPC in paediatric phase 1 trial patients	Retrospective review of medical records to compare the characteristics of children who received (*n* = 23) and did not receive (*n* = 126) SPPC	×	Gender Age Race/ethnicity Diagnosis
Fraser et al.[23] United Kingdom	Children (age 0–18) who died from cancer *n* = 476	Yorkshire Specialist Register of Cancer in Children and Young People (YSRCCYP)	Children's hospice providing in-hospice care and community palliative care services in the Yorkshire region	To determine which children and young people with cancer use SPPC services	Retrospective analysis linking YSRCCYP and hospice records to assess characteristics of children referred to SPPC (*n* = 179) compared to the whole cohort	×	Gender Age group Diagnosis Deprivation Time period
Fraser et al.[47] United Kingdom	Children (age 0–19) who died from cancer *n* = 657	YSRCCYP	Children's hospice providing in-hospice care and community palliative care services in the Yorkshire region	To assess the impact of SPPC services on planned and emergency hospital admissions before death	Retrospective analysis linking YSRCCYP, hospice and hospital records to compare admissions in children who received (*n* = 182) and did not receive (*n* = 475) hospice care	Hospital admissions (total, planned and emergency)	Gender Age group Diagnosis Deprivation Ethnicity Time period

(Continued)

Table 1. (Continued)

Study ID and country	Population and sample size	Setting	SPPC description	Study aim	Overview of relevant methods	Study investigated the following:	
						SPPC outcomes	Factors affecting SPPC referral/uptake
Friedrichsdorf et al.[48] United States	Parents of children (age 0–17 at diagnosis) who died of cancer *n* = 60	Children's Hospitals and Clinics of Minnesota	Palliative/hospice home-based programmes	To compare symptom distress and quality of life in children receiving and not receiving SPPC	Survey of parents using SCCC to compare outcomes for children receiving (*n* = 30) and not receiving (*n* = 30) SPPC	Symptom distress and management Participation in end-of-life planning Quality of life Location of death	×
Golan et al.[49] Israel	Hospitalised paediatric oncology and haematology patients *n* = 568	The Paediatric Haematology Oncology Department (PHOD) in one children's hospital	Hospital-based paediatric palliative and terminal care unit (PCU) integrated with the PHOD	To explore the impact of SPPC on hospitalisation and exposure to palliative care	Retrospective analysis of hospital records to compare children who were hospitalised in the PCU (*n* = 337) with those who were not (*n* = 231)	Location of death	Gender Age Ethnicity Diagnosis Prognosis
Kassam et al.[50,51] Canada (two papers reporting different outcomes from the same data set)	Children who died of cancer and their parent/s *n* = 75 Paediatric oncologists, oncology nurses or social workers *n* = 48	One children's hospital	Tertiary care palliative care team	To examine end-of-life care location preferences and experiences and impact of SPPC involvement on end-of-life experiences	Parent and provider questionnaires (using SCCC) and review of child's medical records to explore preferences and experiences and compare outcomes for those receiving (*n* = 42) and not receiving (*n* = 33) SPPC	Congruence between preferred and actual location of death End-of-life communication	Diagnosis Stem cell transplant Disease duration Age at death Parent demographics
Kaye et al.[52,53,84,85] United States (four papers reporting different outcomes from the same data set)	Patients with a primary cancer diagnosis who were enrolled in SPPC at the time of death *n* = 321	One large academic paediatric cancer centre	Formal palliative care consultation and follow-up provided by subspecialty team of expert palliative care clinicians	To explore demographic, end-of-life and illness characteristics of children who receive SPPC and how these impact on timing of SPPC	Retrospective cohort study comparing outcomes and characteristics in those receiving early (*n* = 236) and late (<30 days before death) SPPC (*n* = 85)	Location of death (early/late SPPC and hospice involvement)	Age Gender Race and ethnicity Diagnosis Cancer treatment ICU stays CPR used Advance directive Hospice involvement (early/late SPPC)
Keim-Malpass et al.[54] United States	Young adults with cancer who died (age 18–39) in hospital *n* = 61	One tertiary academic hospital	Palliative care consultation	To examine death characteristics and end-of-life care trajectories for young adults with cancer	Retrospective chart review to compare differences between young adults receiving (*n* = 19) and not receiving (*n* = 42) SPPC[a] and outcomes associated with SPPC	Location of death Documented family meeting DNR order and timing ICU length of stay CPR used Hospital cost	Diagnosis
Klopfenstein et al.[55] United States	Children and adolescents who died of cancer *n* = 95	One children's hospital's tumour registry database	Children's hospice	To describe the variables influencing end-of-life care including the availability of children's hospice	Retrospective chart review examining end-of-life care patterns and impact of receiving (*n* = 31) or not receiving (*n* = 64) SPPC	Location of death DNR order and timing Length of the last hospital stay	Age Diagnosis Disease status Location of family (rural/urban)
Levine et al.[56] United States	Patients who died (at age < 22) from a brain or solid tumour *n* = 277	One children's research hospital	Hospice enrolment	To determine whether enrolment in a phase 1 trial affects end-of-life care for children with cancer and their families	Retrospective chart review comparing receipt of SPPC between children enrolled (*n* = 120) and not enrolled (*n* = 157) in a phase 1 trial	×	Enrolment in early phase clinical trial

(Continued)

Table 1. (Continued)

Study ID and country	Population and sample size	Setting	SPPC description	Study aim	Overview of relevant methods	Study investigated the following:	
						SPPC outcomes	Factors affecting SPPC referral/uptake
Mark et al.[86] United States	Young adults who died (at age 18 or over) from of relapsed/refractory cancer *n* = 71	One large quaternary care paediatric hospital	Hospice involvement (nearly all participants had hospital SPPC involvement)	To understand the effect of treatments on end-of-life experiences for young adults with cancer	Retrospective cohort study to determine the factors associated with inpatient death in young adults who died from cancer including SPPC	Location of death	Diagnosis
Revon-Rivière et al.[87] France	Patients aged 0–25 at the time of death who died of cancer *n* = 1899	French national hospital database (national data set)	Hospital palliative care units, identified palliative care hospital beds, hospital mobile end-of-life teams and inpatient services	To determine the patient- and hospital-related predictors of high-intensity end-of-life care in children and young people with cancer	Retrospective cohort study involving multivariable regression to determine the predictors of high-intensity end-of-life care including SPPC	High-intensity end-of-life care[b] Most invasive end-of-life care[c] Chemotherapy ICU admission Emergency admission Acute care admission	Social disadvantage
Rossfeld et al.[88] United States	Patients aged 1–21 with a palliative care diagnosis who stayed in hospital for more than one night *n* = 777 (153 cancer cases)	One academic children's hospital	Palliative care consultation provided by a palliative care team	To estimate the impact of SPPC on ICU stays for children with cancer and non-cancer diagnoses during hospital admissions	Retrospective analysis involving multivariable regressions to determine the predictors for being in intensive care including SPPC	ICU stay	×
Schmidt et al.[57] Germany	Parents of children who died of cancer *n* = 96	16 specialised departments for paediatric oncology in North Rhine Westphalia	Specialised paediatric palliative home care services	To assess whether increased national availability of SPPC improves end-of-life outcomes	Interviews with parents (face-to-face or phone) to compare a historical cohort of children (*n* = 48) with children exposed to SPPC (*n* = 48) to explore patterns of end-of-life care	Symptom prevalence Symptom distress Treatment success Receipt of palliative home care Location of death Congruence between preferred and actual location of death	×
Snaman et al.[58] United States	Young adults (age 15–26) who died in study hospital from cancer *n* = 69	One children's research hospital	Involvement of palliative care team in the form of a palliative care consultation	To characterise and compare illness and end-of-life experiences and compare end-of-life experiences by SPPC involvement	Exploratory retrospective analysis of data extracted from medical records to compare young adults who received (*n* = 50) and did not receive (*n* = 19) SPPC	Treatment intensity Location of death Timing of DNR/POST order Symptom prevalence	×
Thienprayoon et al.[59,60] United States (two papers reporting different outcomes from the same data set)	Children with cancer or bone marrow transplant (at age 0–18) who died *n* = 114	The Center for Cancer and Blood Disorders (CCBD) in North Texas	Two hospice organisations serving patients of CCBD	To determine if ethnicity is associated with hospice enrolment in children with cancer and to determine place of death	Retrospective analysis of CCBD records and hospice records to explore differences in children enrolled (*n* = 95) and not enrolled (*n* = 19) in SPPC	Location of death	Gender Race/ethnicity Religion Payor status Diagnosis Language DNR status

(Continued)

Table 1. (Continued)

Study ID and country	Population and sample size	Setting	SPPC description	Study aim	Overview of relevant methods	Study investigated the following:	
						SPPC outcomes	Factors affecting SPPC referral/uptake
Ullrich et al.[61] United States	Children who underwent stem cell transplantation (SCT) and did not survive *n* = 147 (118 cancer)	One children's hospital and cancer institute	Paediatric palliative care consultation	To evaluate whether SPPC is associated with differences in end-of-life care	Retrospective chart review comparing children who received SPPC (*n* = 37) with those who did not (*n* = 110)	End-of-life discussions DNR order End-of-life treatment (CPR, intubation) Hospice enrolment Location of death	Age (diagnosis, SCT) Gender Ethnicity Diagnosis Transplant type Treatment toxicity
Vern-Gross et al.[62] United States	Children (age 0–20) who died from solid tumours *n* = 191	One children's research hospital	An integrated quality-of-life/palliative care service delivered by an interdisciplinary hospital-based team	To evaluate end-of-life care patterns and outcomes following SPPC implementation	Retrospective chart review comparing children who received SPPC (*n* = 57) with a historical cohort (*n* = 134) prior to SPPC implementation	End-of-life discussions DNR order and timing Hospice enrolment Location of death Bereavement support	×
Widger et al.[63] Canada	Children diagnosed with a primary cancer (age < 15) who died (age < 19) *n* = 572	Paediatric institutions in Ontario with an SPPC team (*n* = 3)	Hospital-based specialist team with expertise in both paediatrics and palliative care	To determine which children with cancer access SPPC and examine the impact of SPPC on high-intensity end-of-life care	Retrospective cohort study comparing children who received SPPC[d] (*n* = 166), general palliative care (*n* = 100) and no palliative care (*n* = 306)	ICU admission Mechanical ventilation In-hospital death Composite measure of high-intensity end-of-life care[e]	Age at death Sex Diagnosis Urban/rural status Deprivation Distance from treatment centre Time period
Wolfe et al.[64] United States	Children who died of cancer and their parent(s) *n* = 221	One children's hospital and cancer institute	Paediatric Advanced Care Team (PACT) consultation	To determine whether introduction of SPPC has led to changes in end-of-life care and outcomes	Parent survey (using SCCC) and review of child's medical records to compare end-of-life care and outcomes for children before (*n* = 102) and after (*n* = 119) SPPC implementation	End-of-life discussions Hospice discussions End-of-life treatments DNR orders Location of death Symptom distress Parent preparedness for end of life	×
Zernikow et al.[90] Germany	Parents of children who died of cancer 5 years earlier *n* = 124	All paediatric oncology departments in one German federal state	Specialised paediatric palliative home care services	To assess whether changes in SPPC provision and SPPC receipt were associated with quality and location of care and death	Repeated cross-sectional cohort study using interviews with parents (using SCCC) and questionnaire of paediatric oncology departments to compare cohorts of children from 2000 (*n* = 48), 2005 (*n* = 48) and 2010 (*n* = 28)	Symptom prevalence Symptom distress Treatment success Parent satisfaction Location of death Congruence between preferred and actual location of death Hospital admissions Hospice utilisation Outpatient treatment	Time period

(Continued)

Table 1. (Continued)

Study ID and country	Population and sample size	Setting	SPPC description	Study aim	Overview of relevant methods	Study investigated the following:	
						SPPC outcomes	Factors affecting SPPC referral/uptake
Quantitative descriptive (n = 12)							
Dalberg et al.[81] United States	Paediatric oncology providers (physicians, nurses and social workers) *n* = 1005	US members of the Children's Oncology Group and APOSW	Specialist paediatric palliative care team early integration model	To assess paediatric oncology providers' perceptions of the barriers and facilitators to early integration of SPPC	Online survey to assess agreement among participants about factors affecting SPPC early integration identified in a previous study[72]	×	Perceived barriers Perceived facilitators
Fowler et al.[65] Multiple	Paediatric oncologists *n* = 623	Children's Oncology Group members (providing active care)	Referral to hospice services/care	To examine hospice referral patterns and identify barriers to referral	Online survey exploring comfort level in dealing with end-of-life care, hospice referrals, barriers and facilitators to referral	×	Hospice availability Decision-making in referral Perceived barriers Perceived facilitators
Kline et al.[66] United States	Parents of high-risk[f] paediatric haematology–oncology patients supported by SPPC *n* = 20	One children's hospital	Embedded paediatric haematology–oncology palliative care programme and decision-making tool	To evaluate family satisfaction with SPPC and its decision-making tool	Quantitative survey of all parents to assess SPPC effectiveness and follow-up interviews (open-ended questions) with nine (of the 20) parents, analysed for patterns and trends	Child's quality of life Communication Treatment options Parental understanding Information and advice	×
Lafond et al.[67] United States	Children and adolescents admitted for haematopoietic stem cell transplantation (HSCT) *n* = 12 (seven cancer cases)	One urban, tertiary centre, freestanding children's hospital	Early nurse-led palliative care consultation and service provided by a palliative care team	To establish the feasibility of integrating palliative care early in the trajectory of HSCT and to measure care outcomes	A prospective study collecting descriptive data on uptake and delivery of SPPC and parent and clinician views (by survey) on acceptability	Family satisfaction Provider satisfaction Transcendent comfort[g]	Acceptability of SPPC
Levine et al.[68] United States	Children with oncologic diagnosis (at age 10–17) and their parent *n* = 129 dyads	Three hospital-based paediatric oncology ambulatory clinics and inpatient units	Palliative care team (described as a group of experts that specialise in treating symptoms and improving quality of life)	To examine attitudes towards early integration of palliative care among young people and parents	Separate surveys of young people and parents (face to face) to compare views about SPPC and explore differences by symptom profile	×	Early SPPC Symptom management Quality of life Awareness of palliative care Perceived effect
Mahmood et al.[69] United States	Children and adolescents (age < 22) with high-risk[h] malignancies and their parents *n* = 20	One university medical centre	Early palliative care consultation	To assess the feasibility and acceptability of early palliative care consultation	Prospective study collecting descriptive data on uptake and delivery of SPPC and parent views (survey) on acceptability	Parent satisfaction	Acceptability of SPPC
Postier et al.[70] United States	Children (age 1–21) with life-threatening conditions who received SPPC *n* = 425 (200 cancer cases)	Children's Hospitals and Clinics of Minnesota's SPPC programmes	Palliative/hospice home-based programmes	To compare hospital resource utilisation before and after enrolment in SPPC	Retrospective analysis of electronic medical record data for 12 months before and after a child is enrolled for SPPC	Hospital admissions (number, length of stay, cost)	×

(Continued)

Table 1. (Continued)

Study ID and country	Population and sample size	Setting	SPPC description	Study aim	Overview of relevant methods	Study investigated the following:	
						SPPC outcomes	Factors affecting SPPC referral/uptake
Spruit et al.[89] United States	Paediatric oncology healthcare providers (physicians, nurses, advance practice professionals) *n* = 156	Children's haematology alliance in one US state	All paediatric palliative care services including inpatient, outpatient and consultation	To evaluate the knowledge and beliefs of paediatric oncology providers regarding the involvement of and barriers to SPPC	Cross-sectional descriptive online survey including 30 questions to explore attitudes and beliefs about SPPC including barriers to access	×	Availability and use of SPPC Beliefs about SPPC Barriers to SPPC involvement
Weaver et al.[71] Multiple countries[i]	Representatives of medical settings providing clinical care to children with cancer *n* = 142	Members of AAP SOHPM, ASPHO WG, AAHPM SIG and SIOP SIG	Palliative care subspecialty team, palliative care consultations, specialist inpatient units, paediatric hospice services	To assess the current status of SPPC provision and practice for children with cancer	Cross-sectional online survey to examine the structure, processes and range of services offered by palliative care teams, triggers to consultation and barriers to provision	×	Perceived barriers to SPPC referral Triggers[j] to SPPC referral
Wentlandt et al.[72] Canada	Paediatric oncologists (*n* = 48) Adult oncologists (*n* = 595)	Canadian members of ASPHO, CAMO, CARO and CSSO	Specialised palliative care (SPC) (e.g. hospice, consultation, unit, clinic)	To describe attitudes and referral practices to SPC	Anonymous online and postal survey of SPC availability and referral practices and attitudes, comparing adult and paediatric oncologists	Provider satisfaction	Attitudes about when, why and who to refer Satisfaction with SPC Perceptions about palliative care Barriers to referral
Wolff et al.[73] Germany	Children treated by SPPC service prior to death *n* = 51 (29 cancer cases) Bereaved parents (*n* = 35 of the 51 children)	One paediatric teaching hospital	Palliative care programme with advanced care planning and home-based service option	To evaluate the impact of SPPC programme on end-of-life experiences	Parent survey (10 items) to assess satisfaction with SPPC and review of children's medical records to determine end-of-life care received	Parent satisfaction Location of death	×
Zhukovsky et al.[74] United States	Children with cancer referred for palliative care consultation *n* = 15	One NCI-designated comprehensive cancer centre	Paediatric palliative care programme in the form of a palliative care consultation	To examine changes in treatment after referral to SPPC	Retrospective chart review to examine end-of-life care patterns for children before and after receiving SPPC	Symptom detection Documented end-of-life discussions Treatment recommendations	×
Qualitative studies (n = 5)							
De Clercq and colleagues[79,80] Switzerland	Oncologists, psychologists, nurses and social workers working in paediatric oncology *n* = 29	Five paediatric oncology group centres	The study explored all palliative care including 'institutional referral practices' to specialist services	To examine the understanding of and attitudes towards paediatric palliative care	Thematic analysis of five mixed focus groups (one per centre) exploring conceptual barriers to palliative care implementation	×	Attitudes about accessing SPPC
Dalberg et al.[75] United States	Paediatric oncology providers *n* = 31	One academic children's hospital	Hypothetical (paediatric palliative care team with early integration model proposed to groups)	To explore perceptions of and barriers and facilitators to early integration of SPPC	Constant comparative analysis of exploratory focus groups with nurses (*n* = 2), oncologists (*n* = 1) and social workers (*n* = 1)	×	Barriers to early SPPC model Facilitators for early SPPC model

(Continued)

Table 1. (Continued)

Study ID and country	Population and sample size	Setting	SPPC description	Study aim	Overview of relevant methods	Study investigated the following:	
						SPPC outcomes	Factors affecting SPPC referral/uptake
Hill et al.[82,83] United States	Paediatric oncology staff (physicians, nurses and social workers) *n* = 29	Bone marrow transplant, neuro-oncology and solid tumour teams in one children's hospital	Specialised paediatric palliative care team	To explore how uncertainty might influence palliative care referrals (part of study co-designing an intervention to address referral barriers)	Phenomenological analysis of 16 semi-structured individual interviews and field notes from co-design workshops to conceptualise how uncertainty influences referrals to SPPC	×	Uncertainty as a factor affecting SPPC access Perceptions about SPPC
Spencer and Battye[76] United Kingdom	Health and social care professionals with a role in supporting children with cancer *n* = 40	Primary, tertiary and community services in South East England	The study explored all palliative care including hospices and 'specialist community palliative services for children with cancer'	To examine how professionals viewed palliative care for children with cancer and to identify provision and future needs	Framework analysis of individual semi-structured interviews and group discussions covering palliative care definitions, provision and needs	×	Attitudes and views about SPPC
Szymczak et al.[77] United States	Paediatric oncology providers *n* = 16	One large children's hospital	PACT – hospital-based specialist multidisciplinary team	To explore how paediatric oncology providers perceived the SPPC service and how these perceptions may influence referral	Modified grounded theory analysis of individual semi-structured interviews with oncologists, nurses, psychologists and social workers	Provider satisfaction Reported benefits	Understanding and perception of SPPC service

SPPC: specialist paediatric palliative care; DNR: do-not-resuscitate; POLST: physician order for life-sustaining treatment; SCCC: Survey about Caring for Children with Cancer; ICU: intensive care unit; CPR: cardiopulmonary resuscitation; POST: physician scope of treatment; APOSW: Association of Paediatric Oncology Social Workers; AAP SOHPM: American Academy of Paediatrics Section on Hospice and Palliative Medicine; ASPHO WG: American Society of Paediatric Haematology and Oncology Palliative Care Working Group; AAHPM SIG: American Academy of Hospice and Palliative Medicine Paediatric Palliative Care Special Interest Group; SIOP SIG: Societe Internationale D'Oncologie Pediatrique Palliative Care Special Interest Group; ASPHO: American Society of Paediatric Haematology/Oncology; CAMO: Canadian Association of Medical Oncologists; CARO: Canadian Association of Radiation Oncologists; CSSO: Canadian Society of Surgical Oncology; SCT: stem cell treatment.

[a]Children were included in the SPPC group if they had received a palliative care consultation at least 1 day before death.

[b]Occurrence of at least one of the following: $\geqslant$1 session of intra-hospital intravenous chemotherapy $<$14 days from death; $\geqslant$1 hospitalisation in intensive care in the last 30 days of life; $>$1 emergency room admission in the last 30 days of life and $>$1 hospitalisation in an acute care unit in the last 30 days of life.

[c]Occurrence of at least one of the following: intubation and/or ventilation; CPR and haemodialysis in the last 30 days of life.

[d]Children were included in the SPPC group if they had received SPPC for at least 30 days before death.

[e]Any one of the following: intravenous chemotherapy within 14 days of death; more than one emergency department visit; more than one hospitalisation and any ICU admission within 30 days of death.

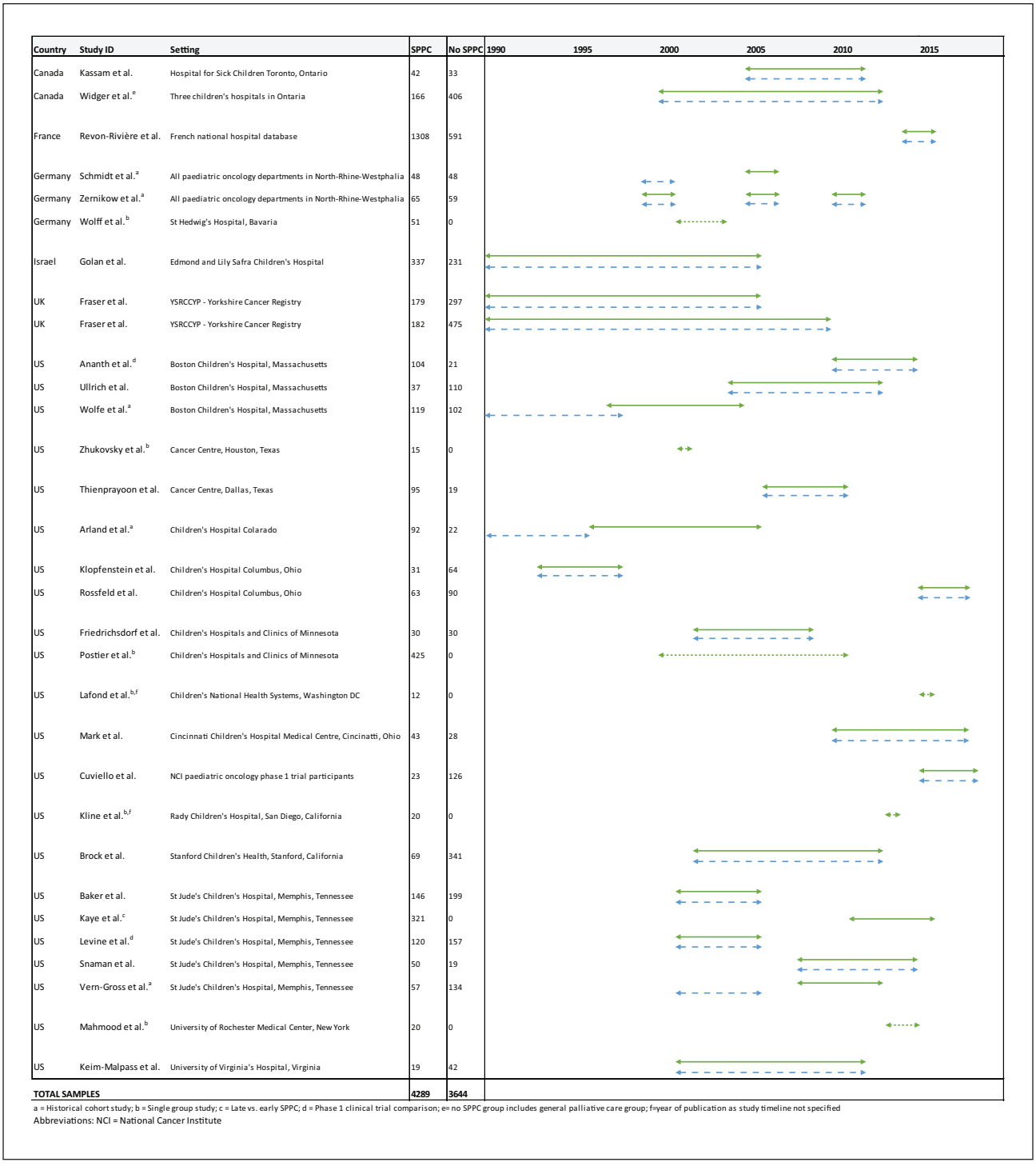
[f]Patients were considered high-risk if they had approached one of the following treatment changes: (1) SCT; (2) transition to end-of-life palliative care and (3) major treatment-related change.

[g]Transcendent comfort was conceptually defined as a state of ease and well-being influenced by the caring and actions of nursing, which lead to transcendence of the circumstances of symptom distress, functional status and quality of life.

[h]Patients were considered high-risk if they had (1) a newly diagnosed malignancy with estimated survival of less than 50%; (2) cancer requiring SCT and (3) relapsed, recurrent or progressive cancer.

[i]18 countries and 39 states.

[j]Triggers to consultation are defined as a diagnosis or prognosis predetermined to warrant consideration of an automatic referral to the palliative care subspecialty team.



Country	Study ID	Setting	SPPC	No SPPC
Canada	Kassam et al.	Hospital for Sick Children Toronto, Ontario	42	33
Canada	Widger et al.[e]	Three children's hospitals in Ontaria	166	406
France	Revon-Rivière et al.	French national hospital database	1308	591
Germany	Schmidt et al.[a]	All paediatric oncology departments in North-Rhine-Westphalia	48	48
Germany	Zernikow et al.[a]	All paediatric oncology departments in North-Rhine-Westphalia	65	59
Germany	Wolff et al.[b]	St Hedwig's Hospital, Bavaria	51	0
Israel	Golan et al.	Edmond and Lily Safra Children's Hospital	337	231
UK	Fraser et al.	YSRCCYP - Yorkshire Cancer Registry	179	297
UK	Fraser et al.	YSRCCYP - Yorkshire Cancer Registry	182	475
US	Ananth et al.[d]	Boston Children's Hospital, Massachusetts	104	21
US	Ullrich et al.	Boston Children's Hospital, Massachusetts	37	110
US	Wolfe et al.[a]	Boston Children's Hospital, Massachusetts	119	102
US	Zhukovsky et al.[b]	Cancer Centre, Houston, Texas	15	0
US	Thienprayoon et al.	Cancer Centre, Dallas, Texas	95	19
US	Arland et al.[a]	Children's Hospital Colarado	92	22
US	Klopfenstein et al.	Children's Hospital Columbus, Ohio	31	64
US	Rossfeld et al.	Children's Hospital Columbus, Ohio	63	90
US	Friedrichsdorf et al.	Children's Hospitals and Clinics of Minnesota	30	30
US	Postier et al.[b]	Children's Hospitals and Clinics of Minnesota	425	0
US	Lafond et al.[b,f]	Children's National Health Systems, Washington DC	12	0
US	Mark et al.	Cincinnati Children's Hospital Medical Centre, Cincinatti, Ohio	43	28
US	Cuviello et al.	NCI paediatric oncology phase 1 trial participants	23	126
US	Kline et al.[b,f]	Rady Children's Hospital, San Diego, California	20	0
US	Brock et al.	Stanford Children's Health, Stanford, California	69	341
US	Baker et al.	St Jude's Children's Hospital, Memphis, Tennessee	146	199
US	Kaye et al.[c]	St Jude's Children's Hospital, Memphis, Tennessee	321	0
US	Levine et al.[d]	St Jude's Children's Hospital, Memphis, Tennessee	120	157
US	Snaman et al.	St Jude's Children's Hospital, Memphis, Tennessee	50	19
US	Vern-Gross et al.[a]	St Jude's Children's Hospital, Memphis, Tennessee	57	134
US	Mahmood et al.[b]	University of Rochester Medical Center, New York	20	0
US	Keim-Malpass et al.	University of Virginia's Hospital, Virginia	19	42
TOTAL SAMPLES			**4289**	**3644**

a = Historical cohort study; b = Single group study; c = Late vs. early SPPC; d = Phase 1 clinical trial comparison; e= no SPPC group includes general palliative care group; f=year of publication as study timeline not specified
Abbreviations: NCI = National Cancer Institute

Figure 2. Data collection period for studies examining outcomes or characteristics of children receiving specialist paediatric palliative care.

and late interventions, included all children receiving specialist palliative care but defined early provision as that received for more than 30 days.[52,53] Only two studies explored how the duration of specialist paediatric palliative care exposure affected outcomes.[61,70]

Study quality

The quality and reporting of studies varied greatly among the 37 quantitative studies, with scores ranging from 0% to 100% (see Table 2).

There were concerns about the representativeness of samples in 14 of the quantitative studies, due to the single-site design of many studies, long study period and inappropriate participant selection or recruitment methods.[44,46,48–51,54,57,61,62,64,70,72,86,89] Assessment of representativeness was not possible in another seven studies because of poor reporting.[66,68,73,78,81,89,90] In eight studies, there were concerns about the comparability of groups (e.g. use of historical cohorts, different participant characteristics), which were not accounted for in the analysis.[44,50,51,54,55,57,58,62,90]

Although inappropriate measures were identified in only three studies,[50,51,56,66] wider concerns about measurement were evident across studies. These included how receipt of specialist palliative care was determined (e.g. from day of initial palliative care consultation); whether outcomes could be attributed to specialist provision or care from the primary oncology team; potential recall problems in studies using bereaved parent-reported outcomes and failure to take account of the chronology of variables. For example, having a do-not-resuscitate (DNR) order was a predictor of specialist palliative care in some studies[46,59,60] and an outcome in others.[54,58] Determining the extent of these limitations was hindered by poor reporting of study methods.

The five qualitative studies[75–77,79,83] used appropriate methods for sampling, data collection and analysis. However, there were limitations to the transferability of findings due to the single-site design in three studies,[75–77,83] and for the third, the time period elapsed since the study was conducted (published in 2001 when specialist paediatric palliative care for children with cancer was at an early stage of development).[76]

The impact of specialist paediatric palliative care

A total of 17 distinct outcome domains were identified and these were categorised under one of six overarching categories: advance care planning ($n = 9$ studies), end-of-life care provision ($n = 16$), location of death ($n = 18$), child's quality of life ($n = 6$), family support ($n = 3$) and service satisfaction ($n = 7$). The synthesis of results by *category* and *outcome domain* is summarised below and in detail in Table 3.

Advance care planning

The outcome domains were of *end of life discussions* and decisions about attempting resuscitation (*DNR orders*), and the timing of these. Provision of specialist palliative care was found to be associated with an increased likelihood of end-of-life discussions being documented in all six studies that measured this,[50,54,61,62,64,74] with evidence from two studies that these occurred earlier in the child's illness in those receiving specialist palliative care.[61,64] Six studies found that *DNR orders* occurred earlier in care trajectories in those receiving specialist palliative care.[46,54,55,58,62,64] However, the evidence about whether a DNR order was more likely was mixed.[46,54,55,61,62,64]

End-of-life care provision

There was evidence from the seven studies which measured *treatment intensity*[54,58,61,63,64,87,88] that children who received specialist paediatric palliative care were less likely to receive high-intensity treatments and to spend less time in an intensive care unit (ICU) during the end-of-life phase, compared to children who did not receive this. There was also evidence from the five studies which assessed *hospital admissions* (either the duration of stay or the number of admissions) that children who received specialist palliative care spent less time in the hospital than those who did not.[44,47,55,70,90] Only one study compared the types of admission; here specialist palliative care was only found to decrease the number of planned admissions.[47] Two studies examined the *cost of care*, also focusing on hospital admissions, but did not find any significant differences between before and after specialist input,[70] or between those who received specialist palliative care and those who did not.[54]

There was no evidence to suggest that *hospice care*, either enrolment or utilisation, changed as a result of specialist involvement from the four studies that explored this.[46,61,62,90] Outpatient care,[90] home-based care[57] and preferred location of care[51] were each assessed by a single study so no conclusions can be drawn about these.

Location of death

A total of 17 studies examined differences in *location of death* between children receiving and not receiving specialist palliative care. A consistent finding across studies was that children who received this were less likely to die in ICU.[49,52,54,57,58,61,64,84] However, studies varied in whether or not they found differences in the proportion of home or hospital deaths.[46,48,57,59,60,62,64,73,90] For example, the study comparing early and late involvement of a hospital team found that 'late access' children were nearly five times more likely to die in ICU than at home or in a hospice, but observed no differences in terms of non-ICU hospital versus home/hospice deaths.[52] However, they did find lower odds of hospital deaths when comparing hospice and no hospice involvement.[84] Just four studies investigated the impact of specialist palliative care on whether children died in their/families *preferred location of death*,[48,51,57,90] three of which found that congruence between preferred and actual place of death increased with specialist involvement.[48,51,90]

Table 2. Critical appraisal summary for included studies.

Quantitative non-randomised studies

Study ID	Overall score (%)[a]	Screening questions		Design-specific questions			
		Are there clear quantitative research questions?	Do the collected data address the research question?	Are participants (and/or organisations) recruited in a way that minimises selection bias?	Are measurements appropriate regarding the exposure/intervention and outcomes?	Are the participants comparable, or do researchers take into account the difference between groups?	Are there complete outcome data/an acceptable response rate/an acceptable follow-up rate?
Ananth et al.[43]	100	Yes	Yes	Yes	Yes	Yes	Yes
Arland et al.[44]	0	Yes	Yes	No	Can't tell	No	No
Baker et al.[45]	100	Yes	Yes	Yes	Yes	Yes	Yes
Brock et al.[46]	75	Yes	Yes	No	Yes	Yes	Yes
Cuviello et al.[78]	0	Yes	Yes	Can't tell	Can't tell	Can't tell	Can't tell
Fraser et al.[23]	100	Yes	Yes	Yes	Yes	Yes	Yes
Fraser et al.[47]	100	Yes	Yes	Yes	Yes	Yes	Yes
Friedrichsdorf et al.[48]	50	Yes	Yes	No	Yes	Yes	No
Golan et al.[49]	75	Yes	Yes	No	Yes	Yes	Yes
Kassam et al.[50]	0	Yes	Yes	No	No	No	No
Kaye et al.[84]	100	Yes	Yes	Yes	Yes	Yes	Yes
Keim-Malpass et al.[54]	50	Yes	Yes	No	No	Yes	Yes
Klopfenstein et al.[55]	50	No	Yes	Yes	Can't tell	No	Yes
Mark et al.[86]	25	Yes	Yes	No	Can't tell	Can't tell	Yes
Levine et al.[56]	75	Yes	Yes	Yes	No	Yes	Yes
Revon-Rivière et al.[87]	75	Yes	Yes	Yes	Yes	Yes	Can't tell
Rossfeld et al.[88]	100	Yes	Yes	Yes	Yes	Yes	Yes
Schmidt et al.[57]	0	Yes	Yes	No	Can't tell	Can't tell	No
Snaman et al.[58]	50	Yes	Yes	Yes	Yes	Can't tell	Can't tell
Thienprayoon et al.[60]	100	Yes	Yes	Yes	Yes	Yes	Yes
Ullrich et al.[61]	50	Yes	Yes	No	Yes	Yes	No
Vern-Gross et al.[62]	25	Yes	Yes	No	Yes	No	Can't tell
Widger et al.[63]	100	Yes	Yes	Yes	Yes	Yes	Yes
Wolfe et al.[64]	25	Can't tell	Can't tell	No	Can't tell	Yes	Can't tell
Zernikow et al.[90]	25	Yes	Yes	Can't tell	Yes	Can't tell	No

(Continued)

Table 2. (Continued)

Quantitative descriptive studies

Study ID	Overall score (%)	Screening questions		Design-specific questions			
		Are there clear quantitative research questions?	Do the collected data address the research question?	Is the sampling strategy relevant to address the research question?	Is the sample representative of the population under study?	Are measurements appropriate?	Is there an acceptable response rate?
Dalberg et al.[81]	25	Yes	Yes	Can't tell	Can't tell	Yes	No
Fowler et al.[65]	75	Yes	Yes	No	Yes	Yes	Yes
Kline et al.[66]	0	Yes	No	Can't tell	Can't tell	No	No
Lafond et al.[67]	100	Yes	Yes	Yes	Yes	Yes	Yes
Levine et al.[68]	75	Yes	Yes	Can't tell	Yes	Yes	Yes
Mahmood et al.[69]	100	Yes	Yes	Yes	Yes	Yes	Yes
Postier et al.[70]	0	Yes	Can't tell	No	No	Can't tell	Can't tell
Spruit et al.[89]	50	Yes	Yes	Yes	Can't tell	Yes	No
Weaver et al.[71]	75	Yes	Yes	Yes	Yes	Yes	Can't tell
Wentlandt et al.[72]	50	Yes	Yes	Yes	No	Can't tell	Yes
Wolff et al.[73]	0	Can't tell	Can't tell	Can't tell	Can't tell	Can't tell	No
Zhukovsky et al.[74]	75	Yes	Yes	Yes	Yes	Can't tell	Yes

Qualitative studies

Study ID	Overall score (%)	Screening questions		Design specific questions			
		Are there clear qualitative research questions?	Do the collected data address the research question?	Are the sources of qualitative data relevant to address the research question?	Is the process for analysing qualitative data relevant to address the research question?	Is appropriate consideration given to how findings relate to the context in which the data were collected?	Is appropriate consideration given to how findings relate to researchers' influence?
De Clercq et al.[79]	100	Yes	Yes	Yes	Yes	Yes	Yes
Dalberg et al.[75]	100	Yes	Yes	Yes	Yes	Yes	Yes
Hill et al.[83]	75	Yes	Yes	Yes	Yes	Yes	No
Spencer and Battye[76]	75	Yes	Yes	Yes	Yes	Yes	No
Szymczak et al.[77]	100	Yes	Yes	Yes	Yes	Yes	Yes

[a]Overall score calculated from the number of design-specific questions addressed (Can't tell classified as No for scoring): 0% = 0; 25% = 1; 50% = 2; 75% = 3 and 100% = 4 questions addressed.

Table 3. Impact of specialist paediatric palliative care by outcome domain.

Outcome domain	SPPC model	Study ID	Outcome descriptor	Summary finding[a]	Study design	Quality (%)
Advance care planning (n = 9 studies)						
DNR order (*n* = 7 studies)	Hospice service	Klopfenstein et al.[55]	DNR order in place	More likely in the SPPC group (no *p* value) and after SPPC implemented ($p < 0.05$)	Qn-NR	50
	Hospital team	Vern-Gross et al.[62]	DNR order in place	Higher rate in the SPPC vs non-SPPC group ($p < 0.001$)	Qn-NR[b]	25
	Hospital team	Brock et al.[46]	DNR order/POLST in place	No significant difference between the SPPC and non-SPPC groups ($p = 0.09$)	Qn-NR	75
	Hospital team	Ullrich et al.[61]	DNR order in place	Higher rate in the SPPC vs non-SPPC group ($p = 0.002$), but no association between DNR order and SPPC duration[c] ($p = 1.0$)	Qn-NR	50
	Hospital team	Wolfe et al.[64]	DNR order in place	No significant difference between the SPPC and non-SPPC groups ($p = 0.051$)	Qn-NR[b]	25
	PC consult	Keim-Malpass et al.[54]	DNR order in place	No significant difference between the SPPC and non-SPPC groups ($p = 0.876$)	Qn-NR	50
	Hospice service	Klopfenstein et al.[55]	Time order in effect	In effect for longer in the SPPC vs non-SPPC group ($p < 0.001$)	Qn-NR	50
	Hospital team	Vern-Gross et al.[62]	Time order in effect prior to death	In effect for longer in the SPPC vs non-SPPC group ($p = 0.001$)	Qn-NR[b]	25
	Hospital team	Wolfe et al.[64]	Time order in effect prior to death	In effect for longer in the SPPC (18 days) vs non-SPPC (12 days) group ($p = 0.031$)	Qn-NR[b]	25
	Hospital team	Brock et al.[46]	Time order in effect prior to death	In effect for longer in the SPPC vs non-SPPC group ($p = 0.05$)	Qn-NR	75
	Hospital team	Snaman et al.[58]	Time order in effect prior to death	In effect for longer in the SPPC vs non-SPPC group ($p = 0.008$)	Qn-NR	50
	PC consult	Keim-Malpass et al.[54]	Time order in effect prior to death	In effect for longer in the SPPC vs non-SPPC group ($p = 0.001$)	Qn-NR	50
EOL discussions (*n* = 6 studies)	Hospital team	Kassam et al.[50]	Numbers (11 EOL care elements)	Five elements more likely in the SPPC group ($p < 0.05$); no difference for the other six ($p \geqslant 0.05$)	Qn-NR	0
	PC consult	Keim-Malpass et al.[54]	Numbers (documented family meeting)	More meetings held in the SPPC (95%) vs non-SPPC group ($p = 0.036$)	Qn-NR	50
	Hospital team	Ullrich et al.[61]	Numbers (prognosis, DNR status)	More discussions in the SPPC group (prognosis: 97% vs 83%, $p = 0.04$; resuscitation: 88% vs 58%, $p = 0.002$), but no association with SPPC duration[c] ($p = 0.5$, $p = 1.0$)	Qn-NR	50
	Hospital team	Wolfe et al.[64]	Numbers (prognosis, hospice, DNR)	More hospice discussions in the SPPC (76% vs 54%, $p < 0.001$) group, others no difference	Qn-NR[b]	25
	Hospital team	Vern-Gross et al.[62]	Numbers (total number per patient)	More in the SPPC (median = 12) vs non-SPPC (median = 3) group ($p < 0.001$)	Qn-NR[b]	25
	Hospital team	Vern-Gross et al.[62]	Numbers (total pre- vs post-SPPC)	No significant difference in total number before vs after SPPC receipt (0.386)	Qn-NR[b]	25
	Hospital team	Ullrich et al.[61]	Timing (days before death)	Earlier in the SPPC group (prognosis: 8 vs 2 days, $p < 0.001$; resuscitation: 7 vs 2 days, $p < 0.001$)	Qn-NR	50
	Hospital team	Wolfe et al.[64]	Timing (days before death)	Earlier hospice discussions in the SPPC (52 vs 28 days, $p = 0.002$) group, others no difference	Qn-NR[b]	25
	Hospital team	Zhukovsky et al.[74]	Treatment recommendations by SPPC team	SPPC 'resulted in multiple treatment recommendations for . . . end of life care planning'	Qn-D	75
EOL care provision (n = 16 studies)						
Home-based care (*n* = 1 study)	Home-based service	Schmidt et al.[57]	Home-based care received by families	Higher in the SPPC (65%) vs non-SPPC (35%) group ($p = 0.007$)	Qn-NR[b]	0
Hospital admissions (*n* = 6 studies)	Home-based service	Postier et al.[70]	Admission rates (total number 12 months pre- vs post-SPPC) by duration of SPPC exposure	Children with the least SPPC exposure (under 3 months) experienced a significant decrease in the total number of admissions, whereas those with the highest SPPC exposure (12 months or more) experienced a significant increase. Others experienced no significant difference (*p* values missing)	Qn-D	0
	Home-based service	Zernikow et al.[90]	Admission to hospital during the last month of life	Fewer children in the SPPC group stayed in hospital (40%) compared to those in the non-SPPC group (84%) ($p < 0.0001$)	Qn-NR	25
	Hospice service	Fraser et al.[47]	Admission rates (emergency, planned and overall)	Lower planned admission rates in children referred to SPPC (IRR = 0.60, CI = 0.43–0.85, $p = 0.004$); no significant effect on overall or emergency admission rates	Qn-NR	100
	Integrated service	Arland et al.[44]	Admission rates (number of patients admitted, total number of admissions)	Fewer patients in the SPPC (29%) vs non-SPPC (54%) group admitted to hospital ($p < 0.05$) and 46% fewer admissions in SPPC vs non-SPPC patients (*p* value missing)	Qn-NR[b]	0

(Continued)

Table 3. (Continued)

Outcome domain	SPPC model	Study ID	Outcome descriptor	Summary finding[a]	Study design	Quality (%)
	PC consult	Keim-Malpass et al.[54]	Cost of hospital stays	No significant difference in cost between the SPPC and non-SPPC groups ($p = 0.925$)	Qn-NR	50
	Home-based service	Postier et al.[70]	Cost of hospital admissions 12 months pre- vs post-SPPC	No significant difference in total hospital charges pre- and post-SPPC initiation (p value missing)	Qn-D	0
	Home-based service	Postier et al.[70]	Length of stay (mean days)	No significant difference in the length of stay pre- and post-SPPC initiation (p value missing)	Qn-D	0
	Hospice service	Klopfenstein et al.[55]	Length of last hospital stay (days)	Shorter stay in the SPPC vs non-SPPC group (p value missing)	Qn-NR	50
	Integrated service	Arland et al.[44]	Length of stay (mean days, total days)	Shorter average length of stay in the SPPC (3.03 days) vs non-SPPC (4.05 days) group and fewer admission days in SPPC vs non-SPPC patients (1.25 vs 3.68) (p values missing)	Qn-NR[b]	0
Hospice care ($n = 5$ studies)	Home-based service	Zernikow et al.[90]	Hospice utilisation (time spent in hospice)	No significant difference between the SPPC and non-SPPC groups ($p \geqslant 0.723$)	Qn-NR	25
	Hospital team	Brock et al.[46]	Hospice enrolment numbers	No significant difference between the SPPC and non-SPPC groups ($p = 0.66$)	Qn-NR	75
	Hospital team	Ullrich et al.[61]	Hospice enrolment numbers	No significant difference between the SPPC and non-SPPC groups ($p = 0.6$) although children who received SPPC for longer ($\geqslant$1 month)[c] were more likely to receive hospice care (41% vs 5%) ($p = 0.01$)	Qn-NR	50
	Hospital team	Vern-Gross et al.[62]	Hospice enrolment numbers	Higher in the SPPC (71%) vs non-SPPC (46%) group ($p = 0.002$)	Qn-NR[b]	25
	Hospital team	Vern-Gross et al.[62]	Hospice enrolment timing (days to death)	No significant difference between the SPPC and non-SPPC groups ($p = 0.277$)	Qn-NR[b]	25
	Hospital team	Brock et al.[46]	Hospice utilisation (time spent at hospice)	No significant difference between the SPPC and non-SPPC groups ($p = 0.95$)	Qn-NR	75
Outpatient care ($n = 1$ study)	Home-based service	Zernikow et al.[90]	Outpatient treatment or daycare	No significant differences between the SPPC and non-SPPC groups (both $p \geqslant 0.723$)	Qn-NR	25
Preferred location of care ($n = 1$ study)	Hospital team	Kassam et al.[51]	Congruence between preferred and actual location of EOL care	Congruence not associated with SPPC involvement ($p = 0.07$)[d]	Qn-NR	0
Treatment intensity ($n = 7$ studies)	Hospital team	Revon-Rivière et al.[87]	Acute care unit (short-term medical treatment for acute illnesses) admission in the last 30 days of life	No significant difference between the SPPC and non-SPPC groups (multivariate analysis, $p = 0.058$, significance in univariate analysis not reported but $p < 0.2$)	Qn-NR	75
	Hospital team	Snaman et al.[58]	CPR attempts (in LMOL)	No significant difference between the SPPC and non-SPPC groups ($p = 0.203$)	Qn-NR	50
	Hospital team	Ullrich et al.[61]	CPR attempts	Fewer in the SPPC (3%) vs non-SPPC (20%) group ($p = 0.03$), but no association between CPR attempts and SPPC duration[c] ($p = 1.0$)	Qn-NR	50
	PC consult	Keim-Malpass et al.[54]	CPR attempts	No significant difference between the SPPC and non-SPPC groups ($p = 0.759$)	Qn-NR	50
	Hospital team	Snaman et al.[58]	Dialysis (in LMOL)	No significant difference between the SPPC and non-SPPC groups ($p = 0.232$)	Qn-NR	50
	Hospital team	Revon-Rivière et al.[87]	Emergency room admission in the last 30 days of life	No significant difference between the SPPC and non-SPPC groups ($p > 0.2$)	Qn-NR	75
	Hospital team	Revon-Rivière et al.[87]	High-intensity end-of-life (HI-EOL) care (composite – see Table 1)	Odds of receiving HI-EOL care were lower in the SPPC (51%) vs non-SPPC (83%) group (multivariate analysis, OR = 0.31, $p < 0.001$). Early (>1 month before death) vs late (within the month of death) SPPC was also associated with less HI-EOL care ($p < 0.001$)	Qn-NR	75
	Hospital team	Revon-Rivière et al.[87]	High-intensity end-of-life care – most invasive (MI-EOL) (composite – see Table 1)	Odds of receiving MI-EOL care were lower in the SPPC vs non-SPPC group (multivariate analysis, OR = 0.14, $p < 0.001$)	Qn-NR	75
	Hospital team	Widger et al.[63]	HIT (composite – see Table 1)	Odds of receiving HIT were lower in the SPPC vs non-SPPC group (OR = 0.2, $p < 0.001$)	Qn-NR	100
	PC consult	Keim-Malpass et al.[54]	ICU length of stay	No significant difference between the SPPC and non-SPPC groups ($p = 0.979$)	Qn-NR	50
	PC consult	Keim-Malpass et al.[54]	ICU stay (all patients died as inpatients)	Less likely in the SPPC (31%) vs non-SPPC (66%) group ($p = 0.007$)	Qn-NR	50

(Continued)

Table 3. (Continued)

Outcome domain	SPPC model	Study ID	Outcome descriptor	Summary finding[a]	Study design	Quality (%)
	Hospital team	Revon-Rivière et al.[87]	ICU stay in the last 30 days of life	Odds of ICU stay lower in the SPPC vs non-SPPC group (multivariate analysis, OR = 0.16, $p < 0.001$)	Qn-NR	75
	Hospital team	Rossfeld et al.[88]	ICU stay within a hospital admission	Receipt of SPPC within 1 day of hospital admission was associated with 79% lower odds of being in the ICU (OR = 0.21, $p < 0.001$). If SPPC was initiated further into a hospital stay, the association weakened (e.g. on day 7 the odds changed to 74%)	Qn-NR	100
	Hospital team	Widger et al.[63]	ICU stay within 30 days of death	Odds of ICU admission lower in the SPPC vs non-SPPC group (OR = 0.2, $p < 0.001$)	Qn-NR	50
	Hospital team	Ullrich et al.[61]	Intubations in the last 24 h	Fewer in the SPPC (42%) vs non-SPPC (66%) group ($p = 0.02$), but no association between intubations and SPPC duration[c] ($p = 0.9$)	Qn-NR	50
	Hospital team	Wolfe et al.[64]	Intubations in last 24 h	No significant difference between the SPPC and non-SPPC groups ($p = 0.303$)	Qn-NR[b]	25
	Hospital team	Snaman et al.[58]	Invasive medical procedures (LMOL)	Fewer received in the SPPC (median = 1) vs non-SPPC (median = 3) group ($p = 0.009$)	Qn-NR	50
	Hospital team	Snaman et al.[58]	Mechanical ventilation (LMOL)	Fewer mechanical ventilations in the SPPC group (34% vs 63%, $p = 0.028$)	Qn-NR	50
	Hospital team	Widger et al.[63]	Mechanical ventilation within 14 days of death	Odds of ventilation lower in the SPPC vs non-SPPC group (OR = 0.2, $p < 0.001$)	Qn-NR	100
	Hospital team	Snaman et al.[58]	Received benzodiazepines or opioids (LMOL)	No significant difference between the SPPC and non-SPPC groups ($p = 0.207$, $p = 1.00$)	Qn-NR	50
	Hospital team	Revon-Rivière et al.[87]	Received chemotherapy <14 days from death	No significant difference between the SPPC and non-SPPC groups (multivariate analysis, $p = 0.183$, significance in univariate analysis not reported but $p < 0.2$)	Qn-NR	75
	Hospital team	Snaman et al.[58]	Received chemotherapy (LMOL)	No significant difference between the SPPC and non-SPPC groups ($p = 0.731$)	Qn-NR	50
	Hospital team	Wolfe et al.[64]	Timing of stopping cancer-directed treatment	No significant difference between the SPPC and non-SPPC groups ($p = 0.163$)	Qn-NR[b]	25
Location of death (n = 18 studies)						
Location of death ($n = 17$ studies)	Home-based service	Friedrichsdorf et al.[48]	Home deaths	More in the SPPC (93%) vs non-SPPC (20%) group ($p < 0.001$)	Qn-NR	50
	Home-based service	Schmidt et al.[57]	Home deaths	No significant difference between the SPPC vs non-SPPC groups (p value missing)	Qn-NR[b]	0
	Home-based service	Zernikow et al.[90]	Home deaths	More in the SPPC (78%) vs non-SPPC (19%) group ($p < 0.005$)	Qn-NR	25
	Hospital team	Brock et al.[46]	Home deaths	No significant difference between the SPPC and non-SPPC groups ($p = 0.77$)	Qn-NR	75
	Hospital team	Wolfe et al.[64]	Home deaths	No significant difference between the SPPC and non-SPPC groups (p value missing)	Qn-NR[b]	25
	Hospital team	Wolff et al.[73]	Home deaths	More home deaths in the SPPC (69%) vs non-SPPC (18%) group ($p = 0.049$)	Qn-D	0
	PC consult	Thienprayoon et al.[60]	Home deaths	No significant association between PC consult and home death ($p = 0.61$)	Qn-NR	100
	Hospice service	Brock et al.[46]	Home or hospice deaths	More in the SPPC (75%) vs non-SPPC (5%) group (OR = 60, $p < 0.0001$)	Qn-NR	75
	Hospice service	Kaye et al.[84]	Home/hospice vs hospital (non-ICU) deaths	Lower odds (OR = 0.12) of hospital (non-ICU) death with hospice involvement ($p < 0.001$)	Qn-NR	100
	Hospice service	Kaye et al.[84]	Home/hospice vs ICU deaths	Lower odds (OR = 0.02) of ICU death with hospice involvement ($p < 0.0001$)	Qn-NR	100
	Hospital team	Kaye et al.[84]	Home/hospice vs hospital (non-ICU) deaths	Similar odds of dying in hospital (non-ICU) for the early SPPC[e] and late SPPC groups ($p = 0.855$)	Qn-NR	100
	Hospital team	Kaye et al.[84]	Home/hospice vs ICU deaths	Higher odds (OR = 4.7) of ICU death in the late SPPC[e] vs early SPPC group ($p < 0.0001$)	Qn-NR	100
	Hospital team	Ullrich et al.[61]	Home vs hospital (non-ICU) vs ICU deaths; home vs hospital (all)	No significant difference in numbers of home, hospital or ICU deaths ($p = 0.06$) or between numbers who died at home or hospital ($p = 0.5$), and no association with SPPC duration[c] ($p = 0.1$, $p = 0.08$)	Qn-NR	50
	Hospital team	Vern-Gross et al.[62]	Home vs home hospital vs inpatient vs other vs unknown	No significant differences between the SPPC and non-SPPC groups ($p = 0.06$) and similar proportions (56% vs 54%) died at home	Qn-NR[b]	25

(Continued)

Table 3. (Continued)

Outcome domain	SPPC model	Study ID	Outcome descriptor	Summary finding[a]	Study design	Quality (%)
	Hospice service	Thienprayoon et al.[59]	Home vs hospital (non-ICU) vs ICU vs other	More home deaths with SPPC (61% vs 0%); fewer hospital (15% vs 47%) and ICU (5% vs 47%) deaths ($p < 0.001$)	Qn-NR	100
	Integrated unit	Golan et al.[49]	Home vs hospital (non-ICU) vs ICU deaths	Declines in hospital ($p < 0.001$) and home ($p = 0.003$)[f] deaths after opening due to children dying in a new unit, although no change in ICU deaths	Qn-NR	75
	Hospice service	Klopfenstein et al.[55]	Hospital deaths	Fewer hospital deaths in the SPPC (19%) vs non-SPPC (78%) group ($p < 0.0001$)[g]	Qn-NR	50
	Hospice service	Mark et al.[86]	Hospital deaths	Odds of hospital death lower in the SPPC vs non-SPPC group (remained significant in multivariate analysis, OR = 58.8, $p = 0.0011$)	Qn-NR	25
	Hospital team	Widger et al.[63]	Hospital deaths	Odds of in-hospital death lower in the SPPC vs non-SPPC group (OR = 0.2, $p < 0.001$)	Qn-NR	100
	Integrated service	Arland et al.[44]	Hospital deaths	No significant difference between the SPPC and non-SPPC groups ($\chi^2 = 0.642$, df = 1, $p < 0.05$)	Qn-NR[b]	0
	Hospital team	Wolfe et al.[64]	ICU/external hospital (of hospital deaths)	Fewer in the SPPC (22%) vs non-SPPC (38%) group ($p = 0.024$)	Qn-NR[b]	25
	Hospital team	Snaman et al.[58]	ICU deaths	Fewer ICU deaths in the SPPC (38%) vs non-SPPC (68%) group ($p = 0.024$)	Qn-NR	50
	Integrated unit	Golan et al.[49]	ICU deaths	Of all ICU deaths, 8% had used SPPC compared to 92% who had not ($p < 0.001$)	Qn-NR	75
	Home-based service	Schmidt et al.[57]	ICU deaths (as the proportion of hospital deaths)	No significant difference between the SPPC and non-SPPC groups ($p = 0.107$)	Qn-NR[b]	0
	Hospital team	Ullrich et al.[61]	ICU deaths (as the proportion of hospital deaths)	Fewer in the SPPC (58%) vs non-SPPC (80%) group ($p = 0.03$), but no association between ICU deaths and SPPC duration[c] ($p = 0.5$)	Qn-NR	50
	PC consult	Keim-Malpass et al.[54]	ICU deaths (as the proportion of hospital deaths)	Fewer in the SPPC (11%) vs non-SPPC (48%) group ($p = 0.005$, OR = 2.83, CI = 1.14–6.95)	Qn-NR	50
Preferred location of death ($n = 4$ studies)	Home-based service	Friedrichsdorf et al.[48]	Proportion of parents who were able to plan the location of death	No significant difference between the SPPC and non-SPPC groups ($p = 0.08$)	Qn-NR	50
	Home-based service	Friedrichsdorf et al.[48]	Proportion of children who died at the planned location	No significant difference between the SPPC and non-SPPC groups ($p = 0.14$)	Qn-NR	50
	Home-based service	Friedrichsdorf et al.[48]	Congruence between home as a preferred and actual location of death	Congruence increased with SPPC involvement: the vast majority of children who died at home were in the SPPC group as per their wishes (93%) compared with the non-SPPC group (20%) ($p < 0.001$)	Qn-NR	50
	Home-based service	Schmidt et al.[57]	Preferred vs actual location of death	No significant difference in the actual vs preferred location between groups (p value missing)	Qn-NR[b]	0
	Home-based service	Zernikow et al.[90]	Proportion of parents who were able to plan the location of death	Families who received SPPC were more likely to plan the location of death than those who did not receive SPPC (75% vs 48%, $p = 0.003$)	Qn-NR	25
	Home-based service	Zernikow et al.[90]	Proportion of children who died at the planned location	Congruence in the actual vs planned place of death increased with SPPC involvement (92% of children died in the planned place vs 64%, $p = 0.001$)	Qn-NR	25
	Hospital team	Kassam et al.[51]	Congruence between the preferred and the actual location of death	Congruence in the preferred vs actual location of death increased with SPPC involvement ($p = 0.03$) (remained significant in the multivariate analysis)	Qn-NR	0
Child's quality of life ($n = 8$ studies)						
Comfort ($n = 1$ study)	Hospital team	Lafond et al.[67]	Transcendent comfort (child report)	No significant change in comfort over time (p value missing)	Qn-D	100
	Hospital team	Lafond et al.[67]	Transcendent comfort (parent report)	Comfort significantly increased from time of treatment to discharge ($p = 0.008$); in addition, all parents reported that SPPC was very helpful (80%) or helpful (20%) in helping their child to be comfortable	Qn-D	100

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Table 3. (Continued)

Outcome domain	SPPC model	Study ID	Outcome descriptor	Summary finding[a]	Study design	Quality (%)
Quality of life (*n* = 2 studies)	Home-based service	Friedrichsdorf et al.[48]	Amount of fun (parent report)	More fun in the SPPC vs non-SPPC group ($p = 0.03$)	Qn-NR	50
	Home-based service	Friedrichsdorf et al.[48]	Felt peaceful (parent report)	No significant difference between the SPPC and non-SPPC groups ($p = 0.63$)	Qn-NR	50
	Home-based service	Friedrichsdorf et al.[48]	How often afraid (parent report)	No significant difference between the SPPC and non-SPPC groups ($p = 0.15$)	Qn-NR	50
	Home-based service	Friedrichsdorf et al.[48]	Event adding meaning (parent report)	More events in the SPPC vs non-SPPC group ($p = 0.02$)	Qn-NR	50
	Integrated service	Kline et al.[66]	Parents asked if SPPC improved child's QoL	70% strongly agreed that SPPC improved child's quality of life	Qn-D	0
Symptoms (*n* = 5 studies)	Home-based service	Friedrichsdorf et al.[48]	Suffering from symptoms (parent report)[h]	More parents reporting 'a great deal/lot of' suffering from fatigue in the SPPC (93%) vs non-SPPC (63%) group ($p = 0.007$); no difference in suffering across other 12 symptoms measured[h]	Qn-NR	50
	Home-based service	Schmidt et al.[57]	Suffering from symptoms (parent report)[i]	More parents reporting 'a great deal/lot of' suffering from fatigue in the SPPC (50%) vs non-SPPC (25%) group ($p = 0.01$); no difference in suffering across other six symptoms measured[i]	Qn-NR[b]	0
	Home-based service	Zernikow et al.[90]	Suffering from symptoms (parent report)[i]	The proportion of parents reporting symptoms as distressing for their child did not differ between the three cohorts (years 2000, 2005 and 2010, all $p \geqslant 0.029$)	Qn-NR[b]	25
	Hospital team	Wolfe et al.[64]	Suffering from symptoms (parent report)[j]	Fewer parents reporting 'a great deal/lot' of suffering of pain and dyspnoea in the SPPC vs non-SPPC group (pain 47% vs 66%, $p = 0.018$; dyspnoea 37% vs 58%, $p = 0.020$). For the other two, there were no significant differences[j]	Qn-NR[b]	25
	Home-based service	Friedrichsdorf et al.[48]	Symptom prevalence (parent report)[h]	More constipation in the SPPC (70%) vs non-SPPC (36%) group ($p = 0.01$); no difference across the other 12 symptoms measured[h]	Qn-NR	50
	Home-based service	Schmidt et al.[57]	Symptom prevalence (parent report)[i]	More nausea in the SPPC (65%) vs non-SPPC (42%) group ($p = 0.024$); no difference across other six symptoms measured[i]	Qn-NR[b]	0
	Home-based service	Zernikow et al.[90]	Symptom prevalence (parent report)[i]	The rate of symptom occurrence during the EOL period did not differ between the three cohorts (years 2000, 2005 and 2010, all $p \geqslant 0.082$)	Qn-NR[b]	25
	Hospital team	Snaman et al.[58]	Symptom prevalence (median documented in medical record)	No significant difference in the number of total documented symptoms ($p = 0.49$), or physical (0.78), psychosocial (0.12) or refractory (for which interventions had not worked) (0.47) symptoms	Qn-NR	50
	Hospital team	Wolfe et al.[64]	Symptom prevalence (parent report)[j]	No significant difference in the prevalence of four symptoms measured[j]	Qn-NR[b]	25
	Home-based service	Schmidt et al.[57]	Treatment of symptoms (parent report)[i]	More anxiety treatment in the SPPC group (35.7% vs 8.7%, $p = 0.035$); no difference for the other six symptoms	Qn-NR[b]	0
	Home-based service	Zernikow et al.[90]	Treatment of symptoms (parent report)[i]	There was a significant increase in receiving treatment across the cohorts (years 2000, 2005 and 2010) for constipation ($p < 0.001$) and anxiety ($p = 0.044$); no difference for the other five symptoms	Qn-NR[b]	25
	Hospital team	Zhukovsky et al.[74]	Treatment of symptoms by the SPPC team	SPPC 'resulted in the detection of pain & other multiple symptoms' (median of three per patient) and 'multiple treatment recommendations for symptom control'	Qn-D	75
	Home-based service	Friedrichsdorf et al.[48]	Treatment success (parent report)[h]	No significant difference between the SPPC and non-SPPC groups across symptoms	Qn-NR	50
	Home-based service	Schmidt et al.[57]	Treatment success (parent report)[i]	No significant difference between the SPPC and non-SPPC groups across symptoms	Qn-NR[b]	0
	Home-based service	Zernikow et al.[90]	Treatment success (parent report)[i]	No significant differences between the cohorts (years 2000, 2005 and 2010) ($p \geqslant 0.242$)	Qn-NR[b]	25
Family support (n = 3 studies)						
Bereavement support (*n* = 1 study)	Hospital team	Vern-Gross et al.[62]	Bereavement support provided	Higher rate in the SPPC (96%) vs non-SPPC (50%) group ($p < 0.0001$)	Qn-NR[b]	25
	Hospital team	Vern-Gross et al.[62]	Sibling counselling provided	Higher rate in the SPPC (50%) vs non-SPPC (16%) group ($p < 0.0001$)	Qn-NR[b]	25
Parent preparedness (*n* = 2 studies)	Hospital team	Wolfe et al.[64]	Parent prepared for medical problems	Parents felt more prepared in the SPPC (56%) vs non-SPPC (27%) group ($p < 0.001$)	Qn-NR[b]	25
	Hospital team	Wolfe et al.[64]	Parent prepared for EOL circumstances	Parents felt more prepared in the SPPC (49%) vs non-SPPC (25%) group ($p = 0.002$)	Qn-NR[b]	25
	Integrated service	Kline et al.[66]	Understanding of child's condition	80% strongly agreed that SPPC helped understand the child's condition	Qn-D	0

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Table 3. (Continued)

Outcome domain	SPPC model	Study ID	Outcome descriptor	Summary finding[a]	Study design	Quality (%)
Service satisfaction (n = 7 studies)						
Parent satisfaction (*n* = 5 studies)	Home-based service	Zernikow et al.[90]	Parents rated the quality of care during their child's EOL period and the tailoring of care to their child's needs	There was a significant increase in satisfaction between earlier and later cohorts (2000 compared to 2005 and 2010) regarding quality of care ($p < 0.001$) and tailoring of care to child needs ($p < 0.001$)	Qn-NR	25
	Hospital team	Lafond et al.[67]	Six family satisfaction questions answered on a five-point Likert-type scale (1 = low, 5 = high satisfaction), with an overall score being calculated	'Overall, families were very satisfied with integration of palliative care services' – mean scores ranged from 4.6 to 5 across the questions, with a mean overall satisfaction score of 29 (out of possible 30). All parents reported at least 90% satisfaction with the SPPC service	Qn-D	100
	Integrated service	Kline et al.[66]	Evaluation included a series of structured questions asking parents how helpful the service was, treatments provided and some open-ended questions	Overall, parents reported that the SPPC service was helpful (at least 70% of parents agreed or strongly agreed across the items). 85% parents agreed that the SPPC service covered adequate treatment options and 80% agreed that the treatment plan developed with the SPPC team was followed. In open-ended responses, parents also responded positively about the service, what it provided and who delivered it	Qn-D	0
	Hospital team	Mahmood et al.[69]	Brief satisfaction survey	All (*n* = 16) reported being satisfied with the PC team service	Qn-D	100
	Hospital team	Wolff et al.[73]	Satisfaction with the information received, medical care and PC, rated on a Likert-type scale (1 = very good, 6 = unsatisfactory)	Overall satisfaction across the items was very good, with a mean rating of 1.6 (1 = very good, 6 = unsatisfactory). There were no significant differences in satisfaction ratings between parents whose child died at home (which was preferred by most families) or in hospital	Qn-D	0
Provider satisfaction (*n* = 3 studies)	Hospital team	Lafond et al.[67]	Six provider satisfaction questions answered on a five-point Likert-type scale (1 = low, 5 = high satisfaction), with an overall score being calculated	Overall providers indicated satisfaction, with a mean score of 4.4. 'Clinicians indicated that the PC team was helpful in managing symptoms and other stressors and in improving access to services for patients and families'	Qn-D	100
	Hospital team	Szymczak et al.[77]	Paediatric oncology providers were asked about their perceptions of SPPC service and how these may influence timing of referral	The SPPC service was highly regarded by oncology providers, who identified the following impacts: the SPPC team being able to spend time with families and provide emotional support and explore wider needs, expertise in pain and symptom management, supporting transition from hospital to home, around-the-clock support, support for parents and siblings, and helping families to clarify goals	QL	100
	All SPPC models	Wentlandt et al.[72]	Paediatric oncologists asked about the quality of SPPC services in a survey	More than 83% of participants agreed or strongly agreed that they were satisfied with the quality of SPPC services	Qn-D	100

SPPC: specialist paediatric palliative care; DNR: do-not-resuscitate; Qn-NR: quantitative non-randomised; Qn-D: quantitative descriptive; POLST: physician order for life-sustaining treatment; EOL: end of life; IRR: incidence rate ratio; CI: confidence interval; CPR: cardiopulmonary resuscitation; LMOL: last month of life; OR: odds ratio; HIT: high-intensity treatment; ICU: intensive care unit; PC: palliative care; QL: qualitative.

[a]Where results are reported as significant, this refers to statistical not clinical significance.

[b]Historical cohort study.

[c]Compared children who received SPPC for less than a month and for at least a month.

[d]*p* value reported as 0.06 in text and 0.07 in the table.

[e]This study compared early and late SPPC involvement, with late defined as <30 days before death.

[f]*p* value reported as 0.03 in text and 0.003 in the figure.

[g]Fisher's exact test performed on raw data reported in the paper.

[h]Symptoms measured: pain, poor appetite, nausea/vomiting, constipation, diarrhoea, breathing difficulty, energy loss/fatigue, sleep disturbance, sadness/depression, anxiety/nervousness, fear, bleeding episodes and seizures/convulsions.

[i]Symptoms measured: fatigue, pain, loss of appetite, dyspnoea, anxiety, constipation and nausea.

[j]Symptoms measured: fatigue, pain, dyspnoea and anxiety.

Child's quality of life

This theme included three outcome domains: *quality of life, comfort* and *symptoms*. Data were primarily from parent-reported measures collected after a child had died. Two studies measured quality of life but did not use validated measures; one found that children receiving specialist paediatric palliative care had more fun and more events adding meaning compared to children not receiving this, but found no differences in how afraid or peaceful they felt.[48] In the second, 70% of parents strongly agreed that these specialist services improved their child's quality of life, but there was no comparator group.[66] The study which measured *comfort*, also with no comparator, found an increase in comfort levels over time reported by parents but no change reported by children. This was the only study that used a child-reported measure.[67] Although three studies suggested that treatment of symptoms increased with specialist involvement,[57,74,90] there was little evidence that the extent of symptom control/suffering from physical and emotional *symptoms* differed between children receiving and not receiving specialist palliative care. Indeed, high levels of suffering from symptoms were described by the four studies which assessed this.[48,57,64,90]

Family support

Only three studies investigated whether specialist paediatric palliative care affected provision of support to family members, or the impact of that support. One study found that use of *bereavement support* for parents and siblings was more likely if a child had received specialist input.[62] Two studies found that specialist palliative care increased *parent preparedness* for the end-of-life phase of care.[64,66]

Service satisfaction

All seven studies (six surveys[66,67,69,72,73,90] and one qualitative[77]) assessing this reported high levels of family and professional satisfaction with specialist palliative care, with reported benefits including expertise in pain and symptom management, time to plan end-of-life care with families and meeting psychosocial and family needs. None of these studies included a comparator group, although one reported increasing parental satisfaction in line with increasing provision of specialist services.

Factors affecting specialist paediatric palliative care access

The synthesis of studies exploring factors affecting specialist paediatric palliative care access identified four overarching categories each containing several linked themes: sociodemographics (n = 14 studies), disease profile (n = 22 studies), end-of-life care characteristics (n = 18 studies) and acceptability of specialist paediatric palliative care (n = 17 studies). The synthesis of results by *category* and *theme* are summarised below and presented in detail in Table 4.

Sociodemographics

Multiple quantitative studies which investigated the sociodemographic profile of children and/or their families receiving and not receiving specialist palliative care consistently showed that access was not associated with a child's *gender*[47,49,53,59,61,63,78] or *ethnicity*.[45–47,49,50,53,59,61,78,85] There was mixed evidence about whether a *child's age*,[47,49,50,53,55,61,63,78] or the socioeconomic status of their family (e.g. *deprivation, education*),[47,50,59,63,71,87,89] influenced access. Only one or two studies investigated *language*,[46,59] *rurality*,[55,63] *religion*[46,59] and *distance to treatment centre*,[63] so it was difficult to draw conclusions about these factors. Just two of the studies exploring staff attitudes assessed sociodemographic factors, focusing on lack of insurance coverage.[71,89] The majority of participants in both studies did not view this as a barrier to referral.

Disease profile

Out of 12 studies examining *type of cancer*, 10 found that children with solid tumours were more likely to receive specialist palliative care than children with haematological malignancies.[46,47,49,50,53–55,59,61,63,78,86] Six studies investigated whether *disease status* (e.g. prognosis, relapse) was associated with access; no consistency was found across these, with some studies reporting conflicting results.[46,49,50,54,55,61] In contrast, all the studies investigating staff-reported practices consistently identified children with a poor prognosis as those most likely to be referred.[65,71,72,75,77,79,81,83,89] Staff in two studies believed that this could result in referrals that were too late for children to benefit from specialist input.[71,79]

Uncertainty about a child's prognosis and about the benefits of introducing specialist palliative care earlier in the disease trajectory (e.g. around diagnosis) were identified as key barriers to a timely referral,[65,71,75,77,79,81,83,89] although non-physician professionals[75,81] and families[68] were more receptive to early integration than physicians. Automatic referral triggers were used by the majority of providers in one study. However, in line with reported practice, most encouraged referrals for children during the end-of-life phase of care.[71]

End-of-life care characteristics

The end-of-life care provided to children was found to influence whether or not children received specialist palliative care. Four studies consistently found that *advance*

Table 4. Factors affecting access to specialist paediatric palliative care by theme.

Theme	SPPC model	Study ID	Factor descriptor	Summary finding	Study design	Quality (%)
Sociodemographics (n = 14 studies)						
Child's age (*n* = 8 studies)	Hospice service	Klopfenstein et al.[55]	Median age at death	Children receiving SPPC were older (12.4) than those not receiving SPPC (9.0) ($p = 0.013$)	Qn-NR	50
	Hospice service	Fraser et al.[47]	Age at diagnosis (0–4, 5–9, 10–14, 15–19)	Fewer young adults (age 15–19) referred to SPPC compared to other groups (*p* value missing)	Qn-NR	100
	Hospital team	Kaye et al.[84]	Age at death (0–5, 6–12, 13 and older)	Compared to older children (13 and above), the youngest group (age 5 and younger) had higher odds of late (<30 days before death) vs early SPPC involvement (OR = 2.02, $p = 0.03$)	Qn-NR[a]	100
	Hospital team	Ullrich et al.[61]	Median age (time of SCT, age at death)	No significant differences between the SPPC and non-SPPC groups ($p = 0.1$ for both age variables)	Qn-NR	50
	Hospital team	Kassam et al.[50]	Mean age at death	Children receiving SPPC were younger (8.8) than those not receiving SPPC (13.2) ($p = 0.02$)[b]	Qn-NR	0
	Hospital team	Widger et al.[63]	Age at death (0–4, 10–14, 15–18 vs 5–9 (ref.))	Age of death not associated with SPPC involvement ($p = 0.91$ (age 0–4), $p = 0.76$ (10–14), $p = 0.75$ (15–18))	Qn-NR	100
	Integrated service	Golan et al.[49]	Mean age at admission to the SPPC unit	Children receiving SPPC were older (10.8) than those not receiving SPPC (9.3) (*p* value missing)	Qn-NR	75
	PC consult	Cuviello et al.[78]	Age at death (<18, 18+)	No significant differences between the SPPC and non-SPPC groups (*p* value missing)	Qn-NR	0
Deprivation (*n* = 7 studies)	Hospice service	Fraser et al.[47]	Townsend score 1 (least) to 5 (most) deprived	No significant differences between the SPPC group and the whole sample (*p* value missing)	Qn-NR	100
	Hospice service	Thienprayoon et al.[59]	Payor status (Medicaid/private/other)	No significant differences between the SPPC and non-SPPC groups ($p = 0.76$)	Qn-NR	100
	Hospital team	Kassam et al.[50]	Parental income < $50,000	No significant differences between the SPPC and non-SPPC groups ($p = 0.88$)	Qn-NR	0
	Hospital team	Revon-Rivière et al.[87]	Social disadvantage using deprivation index (FDep99 index)	Those living in socially disadvantaged areas had lower access to SPPC (68%) compared to those with no social disadvantage (74%) ($p = 0.003$)	Qn-NR	75
	Hospital team	Widger et al.[63]	Income quintile (Q1 lowest to Q5 highest)	SPPC was less likely for the most deprived (compared to the least deprived: OR = 0.4, $p = 0.01$)	Qn-NR	100
	All SPPC models	*Spruit et al.*[89]	*Paediatric oncology providers (mixed group) asked about barriers to involving SPPC*	*Lack of insurance coverage was identified as a barrier by 3% of nurses and physicians, and 11% of advanced practice professionals (e.g. nurse practitioners, physician assistants)*	*Qn-D*	*50*
	All SPPC models	*Weaver et al.*[71]	*Staff representatives of oncology settings were asked about barriers to referral (ever and most important barrier)*	*Lack of insurance coverage was identified as a barrier by 6% of staff participants and no one rated this as the most important barrier*	*Qn-D*	*75*
Distance to treatment centre	Hospital team	Widger et al.[63]	Long vs short distance (based on the 75th percentile of all distances)	Children who lived a long distance from the treatment centre were less likely to receive SPPC (OR = 0.5, $p < 0.001$) compared to children who lived a short distance	Qn-NR	100
Education	Hospital team	Kassam et al.[50]	Parent had university education	No significant differences between the SPPC and non-SPPC groups ($p = 0.51$)	Qn-NR	0
Ethnicity/race (*n* = 9 studies)	Hospice service	Baker et al.[45]	Black, White	No significant differences between the SPPC and non-SPPC groups ($p = 0.64$)	Qn-NR	100
	Hospice service	Fraser et al.[47]	White, mixed, South Asian, Black, other	No significant difference between the SPPC and non-SPPC group (*p* value missing)	Qn-NR	100
	Hospice service	Thienprayoon et al.[59]	Latino, non-Latino White, other	Latinos were more likely to receive SPPC compared to non-Latino Whites (OR = 5.961, *p* value missing): 94% of Latinos vs 76% of non-Latino Whites vs 73% Other received SPPC ($p = 0.02$)	Qn-NR	100
	Hospice service	Brock et al.[46]	Race: White, Asian, Black, other; ethnicity: Latino, non-Latino	No significant differences between the SPPC and non-SPPC groups ($p = 0.42$ race, $p = 0.60$ ethnicity)	Qn-NR	75
	Hospice service	Kaye et al.[84]	Ethnicity: non-Hispanic, Hispanic	Ethnicity not associated with hospice involvement ($p = 0.459$)	Qn-NR	100

(Continued)

Table 4. (Continued)

Theme	SPPC model	Study ID	Factor descriptor	Summary finding	Study design	Quality (%)
	Hospice service	Kaye et al.[84]	Race: White, Black, other	Race not associated with hospice involvement ($p = 0.894$)	Qn-NR	100
	Hospital team	Brock et al.[46]	Race: White, Asian, Black, other; ethnicity: Latino, non-Latino	No significant differences between the SPPC and non-SPPC groups ($p = 0.22$ race, $p = 0.49$ ethnicity)	Qn-NR	75
	Hospital team	Kassam et al.[50]	Parental White race	No sigifnicant difference between the SPPC and non-SPPC groups ($p = 0.49$)	Qn-NR	0
	Hospital team	Kaye et al.[84]	Ethnicity: non-Hispanic, Hispanic/ Latino	Ethnicity not associated with the timing of SPPC involvement (early vs late) ($p = 0.35$) or days between SPPC initiation and death ($p = 0.578$)	Qn-NR[a]	100
	Hospital team	Kaye et al.[84]	Race: White, Black/African American, other	Race not associated with the timing of SPPC involvement (early vs late) ($p = 0.73$ Black, $p = 0.27$ Other) or days between SPPC initiation and death ($p = 0.488$)	Qn-NR[a]	100
	Hospital team	Ullrich et al.[61]	Race: White; ethnicity: non-Hispanic	No significant differences between the SPPC and non-SPPC groups ($p = 0.4$ race, $p = 0.8$ ethnicity)	Qn-NR	50
	Integrated service	Golan et al.[49]	Jewish, Muslim, Christian	No significant differences between the SPPC and non-SPPC groups (p value missing)	Qn-NR	75
	PC consult	Cuviello et al.[78]	White, Asian, Black, Hispanic, multiracial	No significant differences between the SPPC and non-SPPC groups (p value missing)	Qn-NR	0
Gender ($n = 7$ studies)	Hospice service	Fraser et al.[47]	Male, female	No significant difference between the SPPC and non-SPPC groups (p value missing)	Qn-NR	100
	Hospice service	Thienprayoon et al.[59]	Male, female	No significant difference between the SPPC and non-SPPC groups ($p = 0.62$)	Qn-NR	100
	Hospital team	Kaye et al.[84]	Male, female	Gender not associated with the timing of SPPC involvement (early vs late) ($p = 0.73$)	Qn-NR[a]	100
	Hospital team	Ullrich et al.[61]	Male, female	No significant difference between the SPPC and non-SPPC groups ($p = 0.5$)	Qn-NR	50
	Hospital team	Widger et al.[63]	Male, female	Gender not associated with SPPC involvement ($p = 0.99$)	Qn-NR	100
	Integrated service	Golan et al.[49]	Male, Female	No significant difference between the SPPC and non-SPPC groups (p value missing)	Qn-NR	75
	PC consult	Cuviello et al.[78]	Male, female	No significant differences between the SPPC and non-SPPC groups (p value missing)	Qn-NR	0
Language ($n = 2$ studies)	Hospice service	Brock et al.[46]	English vs non-English speakers	No significant difference between the SPPC and non-SPPC groups ($p = 0.45$)	Qn-NR	75
	Hospice service	Thienprayoon et al.[59]	English/Spanish/other (primary)	No significant differences between the SPPC and non-SPPC groups ($p = 0.07$)	Qn-NR	100
	Hospice service	Thienprayoon et al.[59]	Limited English proficiency	More families with limited English proficiency accessed SPPC (30% vs 5%, $p = 0.02$)	Qn-NR	100
	Hospital team	Brock et al.[46]	English vs non-English speakers	No significant difference between the SPPC and non-SPPC groups ($p = 0.07$)	Qn-NR	75
Religion ($n = 2$ studies)	Hospice service	Brock et al.[46]	Christian/Catholic, no religion, other religion	Christian/Catholic children utilised hospice (55%) at a rate similar to those with no religious preference (57%), but more commonly than children with other religious beliefs (33%) ($p = 0.03$)	Qn-NR	75
	Hospice service	Thienprayoon et al.[59]	Catholic, Christian, Protestant, other	No significant difference between the SPPC and non-SPPC groups ($p = 0.10$)	Qn-NR	100
Rurality ($n = 2$ studies)	Hospice service	Klopfenstein et al.[55]	Rural, urban, suburban county	No significant difference between the SPPC and non-SPPC groups (p value missing)	Qn-NR	50
	Hospital team	Widger et al.[63]	Rural, urban	Rurality not associated with SPPC involvement ($p = 0.96$)	Qn-NR	100
Disease profile ($n = 22$ studies)						
Cancer diagnosis ($n = 12$ studies)	Hospice service	Brock et al.[46]	Solid, brain, haematologic, leukaemia/lymphoma	Children with brain or solid tumours received SPPC more frequently than those with other cancers ($p < 0.0001$). No children with haematologic malignancies were enrolled in SPPC	Qn-NR	75
	Hospice service	Fraser et al.[47]	ICCC diagnostic categories[c]	Greater proportion of children with CNS (39.8%) received SPPC compared to the proportions of children with leukaemia (15.6%) and lymphoma (6.6%) (p value missing)	Qn-NR	100
	Hospice service	Klopfenstein et al.[55]	No details of categories used for analysis	Children with solid tumours were more likely to be receiving SPPC ($p < 0.01$)	Qn-NR	50
	Hospice service	Thienprayoon et al.[59]	Leukaemia, lymphoma and SCT vs brain and solid tumours	Children with leukaemia, lymphoma or SCT were significantly less likely to receive SPPC vs children with brain and solid tumours (OR = 0.166, p value missing): 67% compared to 90% ($p = 0.02$)	Qn-NR	100

(Continued)

Table 4. (Continued)

Theme	SPPC model	Study ID	Factor descriptor	Summary finding	Study design	Quality (%)
	Hospice service	Mark et al.[86]	Leukaemia/lymphoma, solid tumours, brain tumours	Young adults with leukaemia/lymphoma were less likely to be enrolled in hospice care (26.7%) compared to those with solid tumours (66.7%) and brain tumours (71.4%) (*p* value missing)	Qn-NR	25
	Hospital team	Brock et al.[46]	Solid, brain, haematologic, leukaemia/lymphoma	Diagnosis was not associated with SPPC involvement ($p = 0.31$)	Qn-NR	75
	Hospital team	Kassam et al.[50]	Haematologic malignancy	Fewer children with haematologic malignancy in the SPPC (14.3%) vs non-SPPC group (39.4%) ($p = 0.01$)[b]	Qn-NR	0
	Hospital team	Kaye et al.[84]	Solid, brain, haematologic	Children with a haematologic malignancy (OR = 3.24, $p = 0.001$) or a brain tumour (OR = 2.69, $p = 0.003$) had higher odds of late SPPC involvement, compared to children with a solid tumour	Qn-NR[a]	100
	Hospital team	Ullrich et al.[61]	Solid/brain, haematologic	No significant difference between the SPPC and non-SPPC groups ($p = 0.0511$)	Qn-NR	50
	Hospital team	Widger et al.[63]	Solid, CNS, haematologic	SPPC involvement is significantly less likely for children with haematologic malignancies compared to those with solid tumours (OR = 0.3, $p < 0.001$). No difference for children with CNS	Qn-NR	100
	Integrated service	Golan et al.[49]	Sarcoma, low/high-grade brain tumour, low/high-risk neuroblastoma, leukaemia, lymphoma, Wilms, haematologic, other	Those in the SPPC group (compared to the non-SPPC group) were more likely to have high-grade brain tumours, sarcoma or high-risk neuroblastoma (17%, 25% and 5%, vs 8%, 12% and 1%, respectively) (*p* value missing). No reported differences for other diagnoses	Qn-NR	75
	PC consult	Keim-Malpass et al.[54]	Solid, brain, CNS, haematologic	Children with solid tumours more likely to receive SPPC (66%) than those with CNS (38%) or haematologic malignancies (25%) ($p = 0.02$)[d]	Qn-NR	50
	PC consult	Cuviello et al.[78]	Leukaemia, sarcoma, NF-1, neuroblastoma, glioblastoma, DIPG, mantle cell lymphoma, melanoma	No significant differences between the SPPC and non-SPPC groups (*p* value missing) (all patients were enrolled on phase 1 trial)	Qn-NR	0
Disease status ($n = 16$ studies)	Hospice service	Klopfenstein et al.[55]	Death caused by progressive disease or therapy related	All the children who died from progressive disease were referred to SPPC compared to none of the children who died from therapy-related complications ($p < 0.001$)	Qn-NR	50
	Hospital team	Ullrich et al.[61]	Death caused by relapse or treatment-related toxicity	Greater proportion of children died from treatment-related toxicity in the SPPC (76%) vs non-SPPC (54%) group ($p = 0.03$)	Qn-NR	50
	Hospital team	Kassam et al.[50]	Disease duration (years, diagnosis to death)	Children with a shorter disease duration were more likely to be referred to SPPC (2.12 years in the SPPC group vs 3.55 years in the non-SPPC group) ($p < 0.01$)[b]	Qn-NR	0
	PC consult	Keim-Malpass et al.[54]	Disease duration (months, diagnosis to death)	Children who received SPPC had a longer disease duration (20 vs 13 months) ($p = 0.02$)[d]	Qn-NR	50
	Hospital team	Ullrich et al.[61]	Disease duration (months, diagnosis to death)	There was no difference in disease duration between children receiving and not receiving SPPC ($p = 0.5$)	Qn-NR	50
	Hospital team	Brock et al.[46]	Relapsed disease or not	No significant difference between the SPPC and non-SPPC groups ($p = 0.47$)	Qn-NR	75
	Integrated service	Golan et al.[49]	Disease progression[e]	Disease progression was more common among children receiving SPPC (33% vs 4%) (*p* value missing)	Qn-NR	75
	Integrated service	Golan et al.[49]	Poor prognosis vs intent to cure (>30% of cure)	Children with an overall poor prognosis were more likely to receive SPPC (91%) compared to 49% of children with >30% chance of cure (*p* value missing)	Qn-NR	75
	Palliative care team	*De Clercq et al.*[79]	*Paediatric oncology providers' perceptions about obstacles to SPPC implementation*	*The majority of participants insisted that SPPC should be provided when there is no response to curative treatment, and viewed palliative care as non-curative care. Despite this, there were uncertainties and disagreements about when to initiate SPPC, and nurses in particular believed that these uncertainties led to late involvement of SPPC, and sometimes too late for children to benefit*	*QL*	*100*

(Continued)

Table 4. (Continued)

Theme	SPPC model	Study ID	Factor descriptor	Summary finding	Study design	Quality (%)
	Palliative care team – hypothetical	*Dalberg et al.*[75]	*Paediatric oncology providers' perceptions of early integration of SPPC service (around diagnosis)*	*Physician participants held the view that SPPC is inconsistent with curative intent but indicated that early integration of SPPC would benefit patients with a survival of <40%–50%. Other participants (nurses, social workers) did not hold this view and believed that basing referral on prognosis alone would exclude some patients in need: 'is there another 40% that could benefit from our extra care that would focus on their coping ability', 'there can be kids with a good prognosis and the family still isn't coping well'. Non-physician participants were more receptive to integrating SPPC within the first month of diagnosis, regardless of prognosis*	*QL*	*100*
	Palliative care team	*Dalberg et al.*[81]	*Paediatric oncology providers' perceptions of the barriers and facilitators to early integration of SPPC*	*Although few participants (5%) stated that they would not refer any patients within the first month of diagnosis, nearly half of the physicians (48%) and social workers (45%) stated that they would limit referrals to patients with a poorer prognosis (compared to 28% of nurses and 31% of nurse practitioners)*	*Qn-D*	*25*
	Hospice service	*Fowler et al.*[65]	*Paediatric oncologists were asked about their referral practices and attitudes*	*Participants most frequently reported referring patients late in the disease course; 44% at the time of progressive disease, 20% when death was imminent and 26% when no additional therapy options were available. Only 2.5% reported referring at the time of relapse. 38% of respondents identified 'extended prognosis' as a reason for not referring children to SPPC.*	*Qn-D*	*75*
	Hospital team	*Hill et al.*[83]	*Paediatric oncology providers' perceptions and uncertainties about SPPC*	*From the results presented, SPPC introduction was associated with a poor prognosis, and prognostic uncertainty was a key barrier to SPPC involvement. This was not just about whether a child would live or die, but was linked to informational uncertainty about diagnosis and treatments, and the uncertainty held by others including families, team members and others who were involved in treating a child. These uncertainties were believed to delay SPPC involvement until late in a child's illness, in other words when there was greater certainty about their prognosis*	*QL*	*100*
	Palliative care team – hypothetical	*Levine et al.*[68]	*Parent and young person dyad attitudes towards early integration of SPPC*	*Few parents and young people expressed opposition to early SPPC involvement (6% and 2%, respectively). At the same time, just over a quarter of both groups were unsure whether they would have accepted a referral at this time. There were varying opinions between and within participant groups about the optimal timing for SPPC; 59% of young people and 50% of parents agreed from the beginning of cancer therapy, 32% and 20% throughout a child's cancer care, 49% and 32% if cancer got worse/came back and 42% and 33% at the end of life. Only a small proportion of participants (11% of young people, 7% of parents) believed that SPPC would help with making initial treatment decisions*	*Qn-D*	*75*
	All SPPC models	*Spruit et al.*[89]	*Paediatric oncology providers (mixed group) asked about who should be offered SPPC and what the barriers to SPPC involvement were*	*The most common indications for SPPC involvement were situations that occur late in the disease trajectory, and only 31% felt that all children with cancer should have SPPC. One in five participants defined SPPC as a service offered to patients at the 'end of life' or 'when death becomes inevitable', and difficulties with prognostication were identified as a barrier to SPPC involvement by 14% of physicians. These different understandings about who SPPC is for were identified as the barrier to involvement by 46% of participants (56% of nurses compared to 25% of physicians)*	*Qn-D*	*50*
	Hospital team	*Szymczak et al.*[77]	*Paediatric oncology providers were asked about their perceptions of SPPC service and how these may influence timing of referral*	*Participants believed that early involvement of SPPC in the care of children with advancing cancer was beneficial, and while no participants explicitly mentioned prognosis or disease status as a trigger, all participants described the referral of SPPC as being linked to the shift from curative to palliative treatment: 'in your mind you bring another service in. It's a really very clear shift from curative thinking to palliative thinking' (physician)*	*QL*	*100*

(Continued)

Table 4. (Continued)

Theme	SPPC model	Study ID	Factor descriptor	Summary finding	Study design	Quality (%)
	All SPPC models	*Weaver et al.*[71]	*Staff representatives of oncology settings were asked about the timing and triggers for SPPC introduction and barriers to referral (ever a barrier and most important barrier)*	*Several automatic triggers for SPPC referral were related to disease status: diagnosis of refractory disease (reported by 32% of participants), diagnosis of recurrent disease (31%) and 'low likelihood' of anticipated event-free survival above certain percent (22%). Only 4% reported that new cancer diagnosis was an automatic trigger. 48% of participants reported that the introduction of palliative care concepts to families was also 'prognosis' specific. Late referrals, which were defined as a 'patient's disease is too advanced for them to benefit significantly from a referral', were identified as a barrier to referral by 76% of participants and the most important barrier by 16% of participants. This was the second most common and important barrier*	*Qn-D*	*75*
	All SPPC models	*Wentlandt et al.*[72]	*Paediatric oncologists were asked about their referral practices and attitudes*	*83% of participants reported always or usually referring terminally ill patients, and for most participants the life expectancy of patients at the point of referral was 6 months or less (85%). 65% of participants identified the diagnosis of incurable cancer as the ideal time for referral; only 8% identified the time of cancer diagnosis regardless of prognosis as an ideal time*	*Qn-D*	*100*
End-of-life care characteristics (n = 18)						
Advance care planning (*n* = 4 studies)	Hospital team	Kaye et al.[84]	Advance directive in place and length of time in effect (7 days or less vs greater than 7 days before death)	Advance directive not associated with timing SPPC involvement (early vs late) ($p = 0.14$); children with advance directive in place for 7 days or less before death had higher odds of late SPPC involvement (OR = 4.81, $p < 0.0001$)	Qn-NR[a]	100
	Hospice service	Brock et al.[46]	DNR order/POLST in place and median days in effect before death	More children in the SPPC (64%) vs non-SPPC (52%) group had DNR order ($p = 0.02$). Orders in place for longer for children who accessed SPPC (30 days) compared to those who did not (3 days) ($p < 0.0001$)	Qn-NR	75
	Hospital team	Brock et al.[46]	DNR order/POLST in place and median days in effect before death	No significant difference in the proportion of children with a DNR order/POLST ($p = 0.09$); however, orders in place for longer for children who accessed SPPC (15 days) vs to those who did not (7 days) ($p < 0.05$)	Qn-NR	75
	Hospice service	Klopfenstein et al.[55]	DNR order in place (for >1 day) and length of time in effect before death	DNR orders more likely to occur in children with a hospice referral ($p < 0.05$). Orders were in place for longer for children who accessed SPPC compared to those who did not ($p < 0.001$)	Qn-NR	50
	Hospice service	Thienprayoon et al.[59]	DNR status – Yes/No/Withdrawal of Care	More children had a DNR order (No) in place in the SPPC (43%) vs non-SPPC (11%) group ($p < 0.001$)	Qn-NR	100
	Hospital team	Kaye et al.[84]	Enrolled in hospice at time of death	Children receiving hospice care at the time of death had lower odds of late SPPC involvement (hospital team) as compared to those not enrolled in hospice (OR = 0.29, $p < 0.001$)	Qn-NR[a]	100
	All SPPC models	*Weaver et al.*[71]	*Staff representatives of oncology settings were asked about barriers to referral (ever a barrier, most important barrier)*	*Providers varied in terms of whether they introduced palliative care concepts to families (e.g. 46% sometimes did this and 32% usually did). There was no significant association between the introduction of palliative care concepts and the number of SPPC referrals (p = 0.88). However, the odds that palliative care principles were introduced to families in settings with automatic triggers for SPPC referral were 3.41 (1.52–7.69) greater than those in settings which did not use referral triggers ($p < 0.003$)*	*Qn-D*	*75*
Identifying needs for specialist input (*n* = 10 studies)	*Palliative care team*	*De Clercq et al.*[79]	*Paediatric oncology providers' perceptions about obstacles to SPPC implementation*	*Nurse participants felt that children's needs were not always taken into consideration because of late referral practices, although they did identify 'very poor quality of life' and 'presence of pain' as triggering implementation. One participant believed that there was not enough need for a specialist service for children with cancer because of the small numbers of children who die.*	*QL*	*100*

(Continued)

Table 4. (Continued)

Theme	SPPC model	Study ID	Factor descriptor	Summary finding	Study design	Quality (%)
	Palliative care team – hypothetical	*Dalberg et al.*[75]	*Paediatric oncology providers' perceptions of early integration of SPPC service (around diagnosis)*	*Non-physician participants believed that patient needs for palliative care during active treatment often go unmet, and that care provided by the oncology team did not always attend to symptoms (particularly psychosocial), resulting in a lower quality of life for children: 'why would we wait a month to say, you're pain and symptom management and issues of quality of life are important . . . how can we help you?' Some nurse participants suggested that this was to do with oncologists' different perceptions of children's needs, and others explained that nurses were more likely to 'hear a lot of the behind the scenes stuff' (nurse) and were therefore more aware of families needs. Some participants believed that having prognostic-based triggers for SPPC referral could exacerbate this problem*	*QL*	*100*
	Palliative care team	*Dalberg et al.*[81]	*Paediatric oncology providers' perceptions of the barriers and facilitators to early integration of SPPC*	*Non-physicians expressed more concern than physicians that quality of life is often overlooked in the face of cancer treatment ($p < 0.01$). Nurses were also more likely to believe than physicians that parents avoid addressing symptoms with their child's oncologist for fear of disappointing him/her ($p < 0.05$). In addition, 74% of social worker participants felt that parents worried that talking about end of life would lead their oncologist to 'give up' on the child (the views among other participant groups were divided about this). These findings suggest that parents may avoid discussing palliative care needs with their child's oncologist*	*Qn-D*	*25*
	Hospice service	*Fowler et al.*[65]	*Paediatric oncologists were asked about their referral practices*	*Hospice referrals were more likely among participants who worked in centres with larger numbers of children receiving new diagnoses each year (OR = 1.8 (1.2–2.8), $p < 0.02$)*	*Qn-D*	*75*
	Palliative care team – hypothetical	*Levine et al.*[68]	*Parent and young person dyad attitudes towards early integration of SPPC*	*Young people were more likely than parents to identify when pain or symptom management was a problem as the optimal timing for SPPC involvement (49% vs 34%, $p = 0.01$). Young people who rated their quality of life as poor or fair were more likely to recommend early SPPC involvement (73%) than those who rated their quality of life as good (61%), and very good or excellent (52%)*	*Qn-D*	*75*
	Not defined – hypothetical	*Spencer and Battye*[76]	*Staff (mixed group) views about the potential role of SPPC for children with cancer*	*Participants believed that the palliative care needs of children with cancer were similar to those of children with other life-threatening diseases and could therefore be supported by generic services (e.g. children's community nursing teams). This, combined with the small numbers of children with advanced cancer who were already being supported by existing services, was not felt to justify a dedicated local service. At the same time, participants believed that children's hospices, which were part of existing provision, were not appropriate for the majority of children with cancer because of the perception that they primarily provide respite care for children with long-term degenerative conditions. They identified specific needs that could be met by a children's hospice, for example, providing an alternative place of care for families who did not want to be at home or in hospital during the end of life, providing respite at home to support transition to home*	*QL*	*75*
	All SPPC models	*Spruit et al.*[89]	*Paediatric oncology providers (mixed group) asked about desire to involve SPPC services*	*High symptom burden was one of the top four reasons to involve SPPC services*	*Qn-D*	*50*

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Table 4. (Continued)

Theme	SPPC model	Study ID	Factor descriptor	Summary finding	Study design	Quality (%)
	Hospital team	*Szymczak et al.*[77]	*Paediatric oncology providers were asked about their perceptions of SPPC service and how these may influence timing of referral*	*Oncologist participants admitted that they did not always have time to meet the emotional and psychosocial needs of families, and that the SPPC team addressed this unmet need: 'they have time to sit for an hour, hear the fears, hear the problems, partner with that family, cry with that family. I've got 17 other patients to see . . . I don't have time . . . and I cannot deliver that to you in an effective way . . . that is immensely valuable because they are providing something that I should but can't'. However, non-physician participants believed that oncologists were not always able to identify the psychosocial needs of their own patients and sometimes needed some encouragement to refer*	*QL*	*100*
	All SPPC models	*Weaver et al.*[71]	*Staff representatives of oncology settings were asked about the timing and triggers for palliative care introduction*	*56% of participants reported that their sites used automatic 'triggers' to prompt timely palliative care referrals, several of which were related to specific palliative care needs: difficult to manage symptoms or high symptom burden (reported by 37% of participants), patients needing discussion of advanced directives (24%) and difficult social situation or family having difficulty coping (23%). However, 65% of participants identified a lack of perceived patient need among staff as a barrier to SPPC referral; this was identified as the most important barrier by 11% of participants*	*Qn-D*	*75*
	All SPPC models	*Wentlandt et al.*[72]	*Paediatric oncologists were asked about referral practices*	*Participants frequently referred terminally ill patients (prognosis less than 1 year) for symptom control (90%) and discharge planning (75%) but less frequently for social (23%), psychological (30%) or spiritual concerns (21%). Only 50% of participants referred patients who were symptom free*	*Qn-D*	*100*
Treatment intensity (n = 11 studies)	Hospital team	Kaye et al.[84]	CPR receipt	CPR receipt not associated with the timing of SPPC involvement (early vs late) ($p = 0.26$)	Qn-NR[a]	100
	Hospital team	Kaye et al.[84]	ICU admission (numbers)	ICU admissions not associated with the timing of SPPC involvement ($p > 0.05$ for all ORs calculated)	Qn-NR[a]	100
	Hospital team	Kaye et al.[84]	Cancer-directed therapy during the last month of life	Children who received cancer-directed therapy had higher odds of late SPPC involvement compared to those who did not (OR = 5.52, $p < 0.0001$)	Qn-NR[a]	100
	Hospice service	Brock et al.[46]	Phase 1 trial enrolment	More children enrolled in phase 1 trial received SPPC (80% vs 49%) (OR = 4.0, $p < 0.0001$)	Qn-NR	75
	Hospice service	Levine et al.[56]	Phase 1 trial enrolment	Phase 1 trial enrolment not associated with involvement ($p = 0.15$) or timing ($p = 0.23$) of SPPC	Qn-NR	75
	Hospital team	Kaye et al.[84]	Phase 1 trial enrolment	Phase 1 trial enrolment not associated with the timing of SPPC involvement (early vs late) ($p = 0.11$)	Qn-NR[a]	100
	PC consult	Ananth et al.[43]	Phase 1 trial enrolment	Children enrolled in phase 1 trial received SPPC later (median 58 days before death) than those not enrolled in phase 1 trial (85 days) ($p = 0.04$). No difference in SPPC receipt ($p = 0.40$)	Qn-NR	100
	Hospice service	Brock et al.[46]	SCT recipient	Fewer children who had undergone SCT received SPPC (32% vs 60%) (OR = 0.3, $p < 0.0001$)	Qn-NR	75
	Hospital team	Kassam et al.[50]	SCT recipient	Fewer children who had undergone SCT receive SPPC (27% vs 53%) ($p = 0.02$)[b]	Qn-NR	0
	Hospital team	Ullrich et al.[61]	SCT type	More allogeneic unrelated donor transplants in the SPPC (68%) vs non-SPPC group (39%) ($p = 0.02$)	Qn-NR	50
	Palliative care team – hypothetical	*Dalberg et al.*[75]	*Paediatric oncology providers' perceptions of early integration of SPPC service (around diagnosis)*	*SPPC was perceived as not consistent with active treatment: 'The palliative care team's idea of the patient's quality of life is to not do chemotherapy and to let them die of their life-threatening disease' (oncologist). Oncologists believed that they themselves were best placed to manage treatment-related symptoms, but were more likely to refer to SPPC for disease-related symptoms*	*QL*	*100*

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Table 4. (Continued)

Theme	SPPC model	Study ID	Factor descriptor	Summary finding	Study design	Quality (%)
	Hospice service	*Fowler et al.*[65]	*Paediatric oncologists were asked about referral practices*	*57% of respondents identified 'continued therapy' as a reason for not referring children to SPPC. 45% of SPPC services were reported to accept patients on chemotherapy, 68% for patients receiving transfusions and 57% for patients receiving total parenteral nutrition (TPN). When asked for suggestions to increase hospice referrals, participants believed having more facilities allowing TPN, chemotherapy and blood product support would help*	*Qn-D*	*75*
	Palliative care team – hypothetical	*Levine et al.*[68]	*Parent and young person dyad attitudes towards early integration of SPPC*	*Very few participants (2% of young people and 2% of parents) believed that early involvement of SPPC would interfere with their cancer treatment*	*Qn-D*	*75*
	All SPPC models	*Weaver et al.*[71]	*Staff representatives of oncology settings were asked about referral triggers for SPPC*	*31% of settings had automatic palliative care referral triggers for children receiving SCT. Only 9% of settings had triggers for children referred to a phase 1 trial*	*Qn-D*	*75*
	All SPPC models	*Wentlandt et al.*[72]	*Paediatric oncologists were asked if SPPC accepted children receiving certain treatments and about their referral practices*	*The majority of respondents reported that the services available to them accepted patients on chemotherapy (64%) and those receiving transfusions (79%). Although the majority of participants referred children late on in their disease, only 13% reported waiting until after chemotherapy or transfusions had been stopped*	*Qn-D*	*100*
We already do palliative care (*n* = 7 studies)	*Palliative care team*	*De Clercq et al.*[79]	*Paediatric oncology providers' perceptions about obstacles to SPPC implementation*	*Some participants believed that the primary oncology team could offer all the necessary palliative care to children.*	*QL*	*100*
	Palliative care team – hypothetical	*Dalberg et al.*[75]	*Paediatric oncology providers' perceptions of early integration of SPPC service (around diagnosis)*	*Participants in all provider groups reported concern about overlap in roles between the oncology and SPPC team: 'the definition of what a palliative team's role are exactly what the primary oncologist is doing' (physician). However, there were differences in opinion between groups; oncologists in particular believed that patients' needs for palliative care were already being met and perceived treatment-related symptom management and discussions of diagnosis, prognosis and treatment options as their responsibility (some perceived a role for SPPC in managing disease-related symptoms). However, nurse and social worker participants reported that children's wider psychosocial and quality-of-life needs were not always met by the oncology team*	*QL*	*100*
	Palliative care team	*Dalberg et al.*[81]	*Paediatric oncology providers' perceptions of the barriers and facilitators to early integration of SPPC*	*Over half of all participants agreed with the concern that the overlapping role between the oncology team and SPPC is a barrier to referral. Nurses were more likely to agree to this statement than physicians and social workers ($p < 0.05$)*	*Qn-D*	*25*
	Hospice service	*Fowler et al.*[65]	*Paediatric oncologists were asked how comfortable they were doing palliative care*	*The majority of participants reported being comfortable in managing end-of-life pain (86%) and end-of-life psychosocial issues (67%). 43% of respondents identified 'access to resources' as a reason for not referring children to SPPC*	*Qn-D*	*75*
	All SPPC models	*Spruit et al.*[89]	*Paediatric oncology providers (mixed group) asked about barriers to involving SPPC*	*Very few participants (<5%) agreed that 'our team already provides the services that SPPC offers' was a barrier to involvement of SPPC*	*Qn-D*	*50*
	All SPPC models	*Weaver et al.*[71]	*Staff representatives of oncology settings were asked about barriers to referral (ever a barrier and most important barrier)*	*The perception that paediatric oncology providers already provide adequate palliative care was the most commonly reported barrier to SPPC referral (76% of participants) and identified as the most important barrier by more participants than any other barrier (29%)*	*Qn-D*	*75*
	All SPPC models	*Wentlandt et al.*[72]	*Paediatric oncologists were asked about their attitudes towards SPPC referral*	*68% of participants enjoyed treating patients at the end of life; however, only 37% were comfortable providing palliative care*	*Qn-D*	*100*

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Table 4. (Continued)

Theme	SPPC model	Study ID	Factor descriptor	Summary finding	Study design	Quality (%)
Acceptability of SPPC (n = 17 studies)						
Added value of SPPC (*n* = 8 studies)	*Palliative care team*	*De Clercq et al.*[79]	*Paediatric oncology providers' perceptions about obstacles to SPPC implementation*	*Some participants believed that promoting the value of SPPC could help address barriers to implementation. However, the uncertainty about when to introduce SPPC was linked to not knowing what the 'added value' would be. Some participants identified the benefits associated with an external palliative care team, which included easing the emotional burden on the primary team and offering an additional perspective on a child's needs*	*QL*	*100*
	Palliative care team – hypothetical	*Dalberg et al.*[75]	*Paediatric oncology providers' perceptions of early integration of SPPC service (around diagnosis)*	*Non-physician participants believed that early involvement of SPPC would ensure a focus on addressing quality-of-life issues which they reported as being overlooked during active treatment; would address symptoms and suffering better than current practice; and lead to enhanced communication and documentation about families' needs and concerns, which some participants believed families did not always feel able to discuss with their oncologist. However, the lack of evidence about the benefits of early involvement of SPPC was viewed as a barrier to referral, and physician participants were not convinced about the added value for patients receiving active treatment: 'I don't know that the palliative care team has a lot more to offer to a patient during their therapy than the primary oncology team' (oncologist)*	*QL*	*100*
	Palliative care team	*Dalberg et al.*[81]	*Paediatric oncology providers' perceptions of the barriers and facilitators to early integration of SPPC*	*The majority of participants believed that early integration of SPPC would provide greater attention to symptom management for children with cancer and improve interdisciplinary communication. However, there were more mixed views about whether early integration of SPPC for all patients diagnosed with cancer would decrease patient suffering. For example, 62% of nurses moderately/strongly agreed with this statement compared to 48% of physicians and 43% of social workers. All participants agreed that evidence about the benefits of early integration was needed*	*Qn-D*	*25*
	Hospital team	*Hill et al.*[83]	*Paediatric oncology providers' perceptions and uncertainties about SPPC*	*Some participants believed that viewing SPPC in terms of the added value for families (e.g. 'I'm offering this family another layer of support') could help overcome barriers to referral, such as concerns about what families might think*	*QL*	*100*
	Palliative care team – hypothetical	*Levine et al.*[68]	*Parent and young person dyad attitudes towards early integration of SPPC*	*40% of young person participants compared to 18% of parents believed that early involvement of SPPC would have been helpful for treating symptoms ($p < 0.001$). However, not all participants (36% of young people and 40% parents) believed that early SPPC involvement would have been a positive addition to the overall care experience*	*Qn-D*	*75*
	All SPPC models	*Spruit et al.*[89]	*Paediatric oncology providers (mixed group) asked about desire to involve SPPC services*	*Nearly all participants (99%) felt that involving SPPC services benefits children and their families, through improved symptom management (95%), enhanced patient and family outcomes (93%) and enhanced family support (92%)*	*Qn-D*	*50*

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Table 4. (Continued)

Theme	SPPC model	Study ID	Factor descriptor	Summary finding	Study design	Quality (%)
	Hospital team	*Szymczak et al.*[77]	*Paediatric oncology providers were asked about their perceptions of SPPC service and how these may influence timing of referral*	*Participants believed that SPPC added value to what they as oncology providers offered ('it's value added to my medical care'), particularly bringing expertise in symptom and pain management, enabling and supporting home-based care for families, having time to provide superior emotional and social support for families around the clock and helping families with planning end-of-life care and clarifying goals. Participants also valued the communication skills of SPPC team members which some participants admitted were superior to their own: 'This was a family that was really doing everything to keep this poor kid alive . . . She just brought this mum around in a way that none of us could ever do' (physician). 'it's kind of reassurance that we're gonna take care of you and your child at home and we're not gonna let anything bad happen' (child life specialist)* Some participants reported that being able to outsource aspects of palliative care benefitted them as well as their patients, from knowing that their patients were receiving the emotional and social support that they themselves lacked time to provide, enabling them to emotionally distance themselves from families if they needed to: 'it doesn't impact the care that I give them but I feel I emotionally don't let myself get too involved', and helping them maintain a work–life balance (as a result of SPPC becoming the first point of contact around the clock)	*QL*	*100*
	All SPPC models	*Weaver et al.*[71]	*Staff (mixed group) were asked about barriers to referral (ever a barrier and most important barrier) from predefined list*	*Not perceiving a benefit from incorporating SPPC was commonly reported as a barrier to referral (65%) and identified as the most important barrier by 16% of participants. Not being aware of the benefits of SPPC involvement and not being aware of the scope of SPPC services were also commonly reported as barriers to referral (44% and 55%, respectively). These were identified as the most important barrier by 9% and 4% of participants*	*Qn-D*	*75*
Bringing in a new team (*n* = 7 studies)	*Palliative care team*	*De Clercq et al.*[79]	*Paediatric oncology providers' perceptions about obstacles to SPPC implementation*	*Concerns were raised about whether the two teams could successfully work together, and some participants were worried about possible interpersonal conflicts*	*QL*	*100*
	Palliative care team – hypothetical	*Dalberg et al.*[75]	*Paediatric oncology providers' perceptions of early integration of SPPC service (around diagnosis)*	*Some participants believed that having another team addressing aspects of PC might negatively affect their relationship with families. Physicians in particular indicated that outsourcing PC to another team could 'make the oncologist look bad'. Physicians also expressed concerns about sharing responsibility for their children with a new team: 'overlapping discussions will decrease trust' (oncologist)*	*QL*	*100*
	Palliative care team – hypothetical	*Levine et al.*[68]	*Parent and young person dyad attitudes towards early integration of SPPC*	*Very few participants (5% of young people and 4% of parents) believed that early involvement of SPPC would interfere with their relationship with their oncologist*	*Qn-D*	*75*
	Not defined – hypothetical	*Spencer and Battye*[76]	*Staff (mixed group) views about the potential role of SPPC for children with cancer*	*Participants expressed concerns that SPPC involvement for children who became palliative would undermine continuity of care, which was identified as the best model for supporting children with cancer*	*QL*	*75*
	All SPPC models	*Spruit et al.*[89]	*Paediatric oncology providers (mixed group) asked barriers to involving SPPC*	*Very few participants (<5%) agreed that the time it takes to involve a new team was a barrier to SPPC involvement. More physicians than nurses reported this as a barrier ($p = 0.009$)*	*Qn-D*	*50*

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Table 4. (Continued)

Theme	SPPC model	Study ID	Factor descriptor	Summary finding	Study design	Quality (%)
	Hospital team	*Szymczak et al.*[77]	*Paediatric oncology providers were asked about their perceptions of SPPC service and how these may influence timing of referral*	*Some participants believed that SPPC referral could alienate families with whom they had a good relationship, and also negatively affect relationships with families which were already strained: 'that family where your relationship is tenuous and this offer is so upsetting that it just impairs your relationship with them'. This was reported to delay timely introduction of SPPC for families*	*QL*	*100*
	All SPPC models	*Wentlandt et al.*[72]	*Paediatric oncologists were asked about their attitudes towards SPPC referral*	*Only 17% of participants indicated that involving SPPC would add too many care providers and very few (2%) believed that referring to SPPC is an abandonment of patients*	*Qn-D*	*100*
Clinician emotion (*n* = 6 studies)	*Palliative care team*	*De Clercq et al.*[79]	*Paediatric oncology providers' perceptions about obstacles to SPPC implementation*	*Some participants acknowledged that they have difficulty accepting a child's change in prognosis and described sometimes being too emotionally involved to start SPPC in a timely manner. The intervention of an external SPPC team was reported to lessen the burden*	*QL*	*100*
	Palliative care team – hypothetical	*Dalberg et al.*[75]	*Paediatric oncology providers' perceptions of early integration of SPPC service (around diagnosis)*	*Oncologists' emotions about their own patients (e.g. their hope for cure even when prognosis was poor, their fear of failure and responsibility for saving their patients) were reported to influence patient care and act as a barrier to SPPC referral: 'part of me that really wants to hold on and keep pressing the family to go for curative intent and it's maybe not the rational thing to do' (oncologist), 'we try to put a positive spin on a bad situation, which is the responsible thing to do' (oncologist)*	*QL*	*100*
	Palliative care team	*Dalberg et al.*[81]	*Paediatric oncology providers' perceptions of the barriers and facilitators to early integration of SPPC*	*Physician participants slightly agreed that the emotional relationship between physician and family was reported to influence what treatment options are offered, and moderately agreed that this influences how options are conveyed to families. Nurses agreed to a lesser extent ($p < 0.01$)*	*Qn-D*	*25*
	Hospital team	*Hill et al.*[83]	*Paediatric oncology providers' perceptions and uncertainties about SPPC*	*Some participants acknowledged sometimes waiting too long before consulting the SPPC team because they were emotionally involved with the family. As one participant explained: 'There are two patients I can think of . . . I would go batshit crazy trying anything on the planet to try and save the lives of those two children . . . I would need someone else to step in and have that conversation because I couldn't do it'. Accepting a child's change in prognosis could also generate feelings of failure, and worries about what families would think of them and how they would react, both of which impacted on referring children to SPPC. Some participants also worried about how this discussion would impact on their relationship with families. Introduction of SPPC was perceived as 'emotionally risk'*	*QL*	*100*
	All SPPC models	*Spruit et al.*[89]	*Paediatric oncology providers (mixed group) asked about barriers to SPPC involvement*	*Just over a third of the participants identified discomfort discussing SPPC with families as a barrier to involvement. More nurses (47%) than physicians (13%) and advanced practice professionals (e.g. nurse practitioners, physician assistants) (26%) identified this as a barrier*	*Qn-D*	*50*

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Table 4. (Continued)

Theme	SPPC model	Study ID	Factor descriptor	Summary finding	Study design	Quality (%)
	Hospital team	*Szymczak et al.*[77]	*Paediatric oncology providers were asked about their perceptions of SPPC service and how these may influence timing of referral*	*Some delays in referring to SPPC were reported to be about professionals not being ready rather than families: 'We say it is the family [who isn't ready for SPPC] but it's a little bit us. We say "they can't hear it. They're not ready". Which sometimes means "I can't hear it. I'm not ready"' (physician). Introducing the service to families was described as difficult and emotional work: 'If you're going to sit with a family and discuss the merits, the benefits, why we think it's important, you have to first explain where we're at. Why am I telling you this today? So it is recapping everything, where we've been, what we're going through, why I'm nervous today, why I think we need to get another team involved' (physician), 'you know this is his second relapse. But his family is like in a totally different direction. . .And this is my primary patient, we email all the time and they tell me they love me and we're like, I'm like part of the family. But I still haven't gone there (SPPC referral) so I feel responsible for that too . . . it is hard to not alienate them, to be honest with them without alienating them' (physician), . . . 'I can only imagine it is very hard to shift the conversation from 'my child is dying' to 'how can I make life the best for the time remaining'. That cannot happen in one conversation' (psychologist)*	*QL*	*100*
Clinical ownership (*n* = 4 studies)	*Palliative care team – hypothetical*	*Dalberg et al.*[75]	*Paediatric oncology providers' perceptions of early integration of SPPC service (around diagnosis)*	*Nurse participants identified the physicians' need to control aspects of patient care and maintain ownership of their patients as a barrier to SPPC referral. 'I don't know if I'd trust the palliative care team to do as well as the way I would want it to be done' (oncologist)*	*QL*	*100*
	Palliative care team	*Dalberg et al.*[81]	*Paediatric oncology providers' perceptions of the barriers and facilitators to early integration of SPPC*	*Around half of the participants believed that paediatric oncologists' need to control all aspects of patient care was a barrier to SPPC. Non-physicians (56%) were more likely to report this as a barrier than physicians (45%) ($p < 0.01$)*	*Qn-D*	*25*
	Hospital team	*Hill et al.*[83]	*Paediatric oncology providers' perceptions and uncertainties about SPPC*	*Certain physicians were known to be more or less receptive to SPPC. This varying acceptability of SPPC among clinicians led to uncertainty about how, when and whether to suggest a referral for a child, and some participants described the practice of keeping quiet until the primary oncologist or the service treating a child at the time made a decision, even when they thought their opinion was wrong*	*QL*	*100*
	All SPPC models	*Spruit et al.*[89]	*Paediatric oncology providers (mixed group) asked about utilisation of SPPC services*	*Participants reported no pressure from their institution or colleagues about whether or not to involve SPPC*	*Qn-D*	*50*
Family readiness (*n* = 4 studies)	*Palliative care team – hypothetical*	*Dalberg et al.*[75]	*Paediatric oncology providers' perceptions of early integration of SPPC service (around diagnosis)*	*Nearly all participants expressed concern that introducing SPPC early (around diagnosis) could lead to additional parental burden. Physician participants in particular believed that families may not be ready for SPPC involvement during the diagnostic period. However, some non-physician participants expressed that the anxiety caused by early integration would be far less than the anxiety experienced if SPPC was not introduced until relapse/disease progression. As one nurse pointed out, 'it's going to be a burden regardless' of timing*	*QL*	*100*

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Table 4. (Continued)

Theme	SPPC model	Study ID	Factor descriptor	Summary finding	Study design	Quality (%)
	Palliative care team	*Dalberg et al.*[81]	*Paediatric oncology providers' perceptions of the barriers and facilitators to early integration of SPPC*	*Participants were divided about whether parents were ready for SPPC around diagnosis, with just under half agreeing that early involvement would increase parental anxiety. However, participants did agree that introducing SPPC would not create an additional burden for parents overall, and that the potential benefits would outweigh the risks. They also agreed that early integration as the standard of care would reduce anxiety associated with SPPC*	*Qn-D*	*25*
	Hospital team	*Hill et al.*[83]	*Paediatric oncology providers' perceptions and uncertainties about SPPC*	*Uncertainty about whether a family was ready to hear about SPPC and accept a change in their child's diagnosis was identified as a key barrier to referral, and some participants worried that having the conversation too early would impact on their relationship with the family. Some participants were also concerned about advocating too much for SPPC as they did not want families to stop treatments before they were ready. Uncertainty about what information a family had already received, that is, how ready they were to be introduced to SPPC, was also a barrier to referral.*	*QL*	*100*
	Hospital team	*Szymczak et al.*[77]	*Paediatric oncology providers were asked about their perceptions of SPPC service and how these may influence timing of referral*	*Most participants suggested that the reason SPPC is not consulted earlier is because they perceive that families are not emotionally ready for them: 'it's knowing that if I call at a certain point, the family is gonna reject [SPPC]' (physician), 'you fear that you would alienate the family and especially a family who is very much in denial' (physician). Some participants also believed that in these cases there could be negative consequences: 'they just feel like the child is dead. They don't understand that there is still time' (physician)*	*QL*	*100*
Family resistance (*n* = 6 studies)	*Palliative care team*	*De Clercq et al.*[79]	*Paediatric oncology providers' perceptions about obstacles to SPPC implementation*	*Family reluctance was identified as a barrier by healthcare staff participants, who believed that they associated SPPC with giving up*	*QL*	*100*
	Hospice service	*Fowler et al.*[65]	*Paediatric oncologists asked about their referral practices*	*24% of paediatric oncologist respondents identified 'family refusal' as a reason for not referring children to SPPC*	*Qn-D*	*75*
	Palliative care team – hypothetical	*Levine et al.*[68]	*Parent and young person dyad attitudes towards early integration of SPPC*	*Only 1% of young people and parent participants believed that SPPC should not be involved in a child's cancer treatment (regardless of timing)*	*Qn-D*	*75*
	All SPPC models	*Spruit et al.*[89]	*Paediatric oncology providers (mixed group) asked about barriers to SPPC involvement*	*38% of participants identified family resistance as a barrier to SPPC involvement*	*Qn-D*	*50*
	All SPPC models	*Weaver et al.*[71]	*Staff (mixed group) were asked about barriers to referral (ever a barrier and most important barrier) from predefined list*	*54% of staff participants identified parental negative perception of palliative care as a barrier to SPPC referral. This was the most important barrier for 9% of participants. In contrast, only 28% of participants identified patient (child/young person) negative perception as a barrier, and no one rated this as the most important barrier*	*Qn-D*	*75*
	All SPPC models	*Wentlandt et al.*[72]	*Paediatric oncologists were asked about their attitudes towards SPPC referral*	*60% of paediatric oncologist respondents believed that their patients had negative perceptions of SPPC; however, very few (4%) reported that their patients refused referral*	*Qn-D*	*100*
Intervention uptake (*n* = 8 studies)	Home-based services	Zernikow et al.[90]	Cohort years: 2000, 2005 and 2010	Proportion of children receiving SPPC significantly increased from 34.8% in 2000 to 64.6% in 2005, but the difference between 2005 and 2010 (60.7%) was not significant. This was in line with the increasing availability of SPPC during this time period	Qn-NR	25

(Continued)

Table 4. (Continued)

Theme	SPPC model	Study ID	Factor descriptor	Summary finding	Study design	Quality (%)
	Hospice service	Brock et al.[46]	Time periods (time of death): 2002–2004, 2005–2007, 2008–2010 and 2011–2014	No significant difference in hospice enrolments over time ($p = 0.55$)	Qn-NR	75
	Hospice service	Fraser et al.[23]	Time period (year of death and referral): 1990–1993, 1994–1997, 1998–2001 and 2002–2005	No significant difference in the number of referrals between time periods ($p = 0.43$)	Qn-NR	100
	Hospital team	Brock et al.[46]	Time periods (time of death): 2002–2004, 2005–2007, 2008–2010 and 2011–2014	Greater percentage of children accessed SPPC in recent quartiles ($p = 0.04$): 27% of children in 2011–2014 compared to <20% in other periods	Qn-NR	75
	Hospital team	Lafond et al.[67]	Referral rates to SPPC and clinician attitudes about referring to SPPC	All 12 eligible children were referred: oncology providers reporting being comfortable or very comfortable referring families to SPPC and were very likely to recommend the service to others	Qn-D	100
	Hospital team	Lafond et al.[67]	Uptake of SPPC and families' attitudes about accessing SPPC	All 12 families referred were enrolled and received SPPC: parents indicated that it was very important to offer SPPC and were very likely to recommend SPPC to others	Qn-D	100
	Hospital team	Mahmood et al.[69]	Whether primary oncologist agreed to SPPC	No oncologist asked for the initial palliative care consultation with families to be deferred	Qn-D	100
	Hospital team	Mahmood et al.[69]	Enrolment and uptake of SPPC (measured using the initial palliative care consultation)	Anticipated 75% enrolment among families; achieved 80% (20 of 25) enrolment and all 20 families received the initial palliative care consult	Qn-D	100
	Hospital team	Ullrich et al.[61]	Palliative care consultation rates over time	Rates of palliative care consultation increased over the study period from 5 over the first 3 years to 17 over the last 3 years (p value missing)	Qn-NR	50
	Hospital team	Widger et al.[63]	Time periods (time of death): early 2000–2004, mid-2005–2008 and late 2009–2012	Children were more likely to access SPPC later in the study period compared with earlier (OR = 43, $p = 0.01$): referrals up from 8.2% of children in 2000–2004 to 84.3% in 2009–2012	Qn-NR	100
	All SPPC models	*Spruit et al.*[89]	*Paediatric oncology providers (mixed group) asked about utilisation of SPPC services*	*Nearly all participants (95%) reported that SPPC services were available at their facility, but 56% stated that they never or rarely involved SPPC for their paediatric patients with cancer. Services were utilised more by staff working in children's hospitals than in oncology programmes within general hospitals ($p = 0.002$)*	*Qn-D*	*50*
Perceived availability ($n = 4$ studies)	*Hospice service*	*Fowler et al.*[65]	*Paediatric oncologists were asked if they had access to SPPC*	*86% reported having access to a local hospice facility, 75% to a hospice palliative care programme, but only 27% to an inpatient hospice facility. Hospice referrals were predicted by the availability of a hospice facility (OR = 5.6 (2.4–13.3), $p < 0.001$)*	*Qn-D*	*75*
	All SPPC models	*Spruit et al.*[89]	*Paediatric oncology providers (mixed group) asked about barriers to SPPC involvement*	*Although availability of SPPC services was high (95%), perceptions about available resources and access to SPPC was identified as a barrier by 17% of participants. Very few participants (around 5%) identified lack of community resources as a barrier. Staff from children's hospitals were less likely to identify these as barriers compared with staff from general hospitals ($p < 0.001$)*	*Qn-D*	*50*

(Continued)

Table 4. (Continued)

Theme	SPPC model	Study ID	Factor descriptor	Summary finding	Study design	Quality (%)
	All SPPC models	*Weaver et al.*[71]	*Staff (mixed group) were asked about access to SPPC and barriers to referral (ever a barrier and most important barrier) from predefined list*	*75% of participants reported having access to an SPPC service where they worked, 15% to an SPPC contact person but not a service and 10% had no SPPC services available to them (3% of these utilised adult providers). Views about whether capacity met demand varied and there was no significant association between perceived capacity and the number of SPPC referrals ($p = 0.97$). Lack of palliative care availability was only identified as a barrier by 1.5% of participants and was not rated as the most important barrier by anyone. However, a greater proportion of participants (36%) identified inadequate palliative care staffing as a barrier, and this was identified as the most important barrier by 6% of participants*	*Qn-D*	*75*
	All SPPC models	*Wentlandt et al.*[72]	*Paediatric oncologists were asked if they had access to different types of SPPC*	*96% of respondents reported that they had access to hospital-based SPPC (although availability of other types of SPPC was much lower) and 73% agreed or strongly agreed that they were satisfied with availability of SPPC*	*Qn-D*	*100*
What SPPC symbolises ($n = 9$ studies)	*Palliative care team*	*De Clercq et al.*[79]	*Paediatric oncology providers' perceptions about obstacles to SPPC implementation*	*The association of SPPC with 'death and dying', 'loss of hope' and 'giving up' was identified by many participants as a barrier to implementation. One participant described the term 'palliative' as the biggest enemy when addressing families, as one participant explained, 'we are afraid of pronouncing the word and at the same time we do not know how to tell it differently'. Some participants felt that palliative care was also associated with the elderly and therefore difficult to reconcile with children's lives. The different meaning of palliative care in other cultures was also identified as a potential barrier. Standardising the introduction of SPPC earlier in the care pathway, rebranding the name of SPPC and improving public perceptions and promoting its value were all identified as potential solutions*	*QL*	*100*
	Palliative care team – hypothetical	*Dalberg et al.*[75]	*Paediatric oncology providers' perceptions of early integration of SPPC service (around diagnosis)*	*Nearly all participants believed that early referral (around diagnosis) would lead to additional parental anxiety because of the association between the term 'palliative care' and end of life, although one nurse pointed out that 'a very small percentage of people even know what that word means'. Changing the service name to 'supportive care team' or 'quality of life team' was identified as a possible solution. Early integration of SPPC as the standard of care, which could focus on educating families about the role and benefit of SPPC, was also identified as something which might help address concerns about what a referral symbolises: 'if it's standard of care, it's very simple; it's the standard of care rather than meeting the death squad', 'hearing the definition of palliative care up front would be different than when you have a setback'.*	*QL*	*100*
	Palliative care team	*Dalberg et al.*[81]	*Paediatric oncology providers' perceptions of the barriers and facilitators to early integration of SPPC*	*Public and provider perceptions that palliative care is synonymous with end of life were identified as a barrier to early integration of SPPC. Interestingly, staff were more likely to report these beliefs to exist in professional groups other than their own (e.g. 73% of physicians believed that nurses associated palliative care with end of life compared to 50% of nurses). All providers agreed that changing the name of SPPC would help, along with education for parents and healthcare providers*	*Qn-D*	*25*
	Hospital team	*Hill et al.*[83]	*Paediatric oncology providers' perceptions and uncertainties about SPPC*	*Participants were worried that SPPC introduction would be associated with the loss of hope, and the team giving up on their child, because of its association with death. Because of the late involvement of SPPC, the conversation about SPPC involvement was also associated with telling a family bad news about their child's prognosis and therefore generated uncertainties about how a family would react. One participant described the introduction of SPPC as a potential 'bombshell'*	*QL*	*100*

(Continued)

Table 4. (Continued)

Theme	SPPC model	Study ID	Factor descriptor	Summary finding	Study design	Quality (%)
	Palliative care team – hypothetical	*Levine et al.*[68]	*Parent and young person dyad attitudes towards early integration of SPPC*	*The majority of parent (62%) and young person (98%) participants reported that they had not heard of the term palliative care, and of those familiar with it none reported a negative attitude. When learning that SPPC provides end-of-life care in addition to symptom management and quality-of-life directed care, 16% of young people and 15% of parents reported that they would have been less willing to meet with the team around diagnosis; however, 26% of young people and 18% of parents reported a greater willingness. Only 2% of young people and 8% of parents believed that SPPC referral would be associated with the loss of hope for a cure*	*Qn-D*	*75*
	Not defined – hypothetical	*Spencer and Battye*[76]	*Staff (mixed group) views about the potential role of SPPC for children with cancer*	*Participants believed that a separate SPPC team would be seen as 'death nurses'*	*QL*	*75*
	All SPPC models	*Spruit et al.*[89]	*Paediatric oncology providers (mixed group) asked about barriers to involving SPPC services.*	*The belief that SPPC involvement would be misinterpreted as 'giving up' was the most commonly cited barrier for oncology providers (49% of participants) when deciding whether to involve SPPC. In addition, 29% of participants believed that SPPC involvement led to loss of hope for families*	*Qn-D*	*50*
	Hospital team	*Szymczak et al.*[77]	*Paediatric oncology providers were asked about their perceptions of SPPC service and how these may influence timing of referral*	*Numerous participants explained that they believed families hear 'palliative' as 'death' and reported that parents of children who had been cared for at the hospital for a long time referred to the SPPC team as 'the death team'. Many participants believed that the term palliative care acted as a barrier to introducing the service: 'when a regular family hears the word palliative they just hear their kid is dying. That's it. They can't focus on anything else' (psychologist). For some, palliative care was also associated with the term 'hospice' for families, which was described as another term meaning death: 'I just think the association of the word palliative, particularly with families who've had experience, maybe like grandma or grandpa, somebody went home on hospice – I think that is an immediate trigger, just the word' (social worker)*	*QL*	*100*
	All SPPC models	*Wentlandt et al.*[72]	*Paediatric oncologists were asked about their attitudes towards SPPC referral*	*58% of participants indicated that they would refer earlier to SPPC if it were renamed 'supportive care', although only 6% reported feeling uncomfortable referring children who were not terminal*	*Qn-D*	*100*

Results in Italics indicate studies that sought the views of families or healthcare staff.
SPPC: specialist paediatric palliative care; Qn-NR: quantitative non-randomised; OR: odds ratio; SCT: stem cell transplant; Qn-D: quantitative descriptive; ICCC: International Classification of Childhood Cancer; CNS: central nervous system; NF-1: neurofibromatosis type 1; DIPG: diffuse intrinsic pontine glioma; QL: qualitative; DNR: do-not-resuscitate; POLST: Physician Order for Life-Sustaining Treatment; CPR: cardiopulmonary resuscitation; ICU: intensive care unit.
[a]Study comparing late versus early SPPC.
[b]In the multivariable analysis, the presence of a hematologic malignancy was the only factor that remained significant; SCT, disease duration and age did not.
[c]ICCC categories include leukaemia, lymphoma, CNS, neuroblastoma, retinoblastoma, renal, hepatic, bone, soft tissue, germ cell, other epithelial and other.
[d]The no-SPPC group included children who first received SPPC on the day of death.
[e]Progression was determined by whether children moved into a different medical category using four categories: IC – intent to cure, ICP – intent to cure palliative, NC – non-curable and TC – terminal care.

care planning (e.g. documented advance directives or DNRs, hospice referrals) was associated with an increased likelihood of specialist involvement, or earlier compared to late involvement.[46,53,55,59] Provision of palliative care by the oncology team (*we already do palliative care*) was also reported by oncology staff to influence referral practices[65,71,72,75,79] and to impede oncologists' abilities and willingness to *identify needs for specialist input* in their patients and practice, something that was reported to encourage referrals.[65,71,72,75–77,79,89] Similarly, young people were more likely to accept specialist input if they had specific unmet needs, for example, pain and poor quality of life,[68] although staff in one study were concerned that parents might not discuss their child's palliative care needs during active treatment.[81]

There was conflicting evidence about whether *treatment intensity* (e.g. phase 1 trial enrolment) influenced access to specialist palliative care,[43,46,50,53,56,61] and this mirrored varying beliefs among oncology staff and families about whether children receiving active treatments should or could be referred.[65,68,71,72,75]

Acceptability of specialist paediatric palliative care

Staff acceptability of specialist palliative care was reported to influence access in 16 studies. Family acceptability was identified in 13, although families' views were sought in only three of these.[67–69] Eight studies measured *intervention uptake* to explore acceptability.[23,46,61,63,67,69,89,90] These reported varying rates of referral and uptake of specialist palliative care, but provided evidence of increasing involvement over time.

Staff uncertainties about the benefits of specialist palliative care, and about how a specialist service differs from care provided within oncology (the *added value of specialist paediatric palliative care*), were identified as key barriers to referral.[68,75–77,79,81,83,89] Concerns that *bringing in a new team* could undermine continuity of care and impact on relationships with families were also reported to influence referral practices,[68,72,75–77,79,89] as were perceptions about availability and capacity of specialist palliative care services (*perceived availability*).[65,71,72]

Staff in eight studies identified the association of palliative care with end of life (*what specialist paediatric palliative care symbolises*) as a barrier to access.[72,75–77,79,81,83,89] A *clinician's emotion* about a family and their readiness to accept a child's prognosis and discuss this were identified as additional challenges,[75,77,79,81,83,89] and contributed to what was described as the emotional labour associated with introducing a service which staff participants referred to as 'death nurses',[76] 'the death team'[77] and 'the death squad'.[75] A perception that oncologists need to control patient care ('clinical ownership') was identified as a further barrier to specialist palliative care referral and could deter others involved in a child's care from recommending this for a family.[75,81,83,89]

Oncology staff reported that *family readiness*[75,77,81,83] and *family resistance*[65,71,72,79,89] could affect access, although views on this varied. For example, 60% of paediatric oncologist respondents in one survey believed that their patients had negative perceptions of specialist palliative care, but very few (4%) reported that patients refused a referral.[72] In another study, only 38% of staff participants identified family resistance as a barrier,[89] and the study that explored young people and parents' attitudes found mainly positive views, and very few participants (2% and 8%, respectively) believed that referral was associated with the loss of hope for a cure,[68] which was a fear commonly reported by staff.[72,75–77,79,83,89]

Discussion

Main findings of the review

This systematic review found evidence that children and young people with cancer who receive specialist paediatric palliative care are more likely to be engaged in advance care planning, receive less intensive care at the end of life and are less likely to die in hospital, compared to those who do not receive this. Some of the included studies also indicate that these differences may be more marked when children receive specialist input for a longer duration before they die. The review did not find that receipt of specialist palliative care is associated with improved quality of life or symptom control; however, no conclusions can be drawn because of the significant methodological limitations of the seven studies investigating this.[48,57,58,64,66,67] Importantly, only one study sought young people's views about the impact of these specialist services across the 28 studies which examined this, and this was a feasibility study with no comparator group.[67]

The review also found that the type of cancer and whether or not paediatric oncology teams themselves engage in palliative care practices may affect access to specialist palliative care services. Thus, studies reported fewer children with haematological malignancies receiving specialist palliative care, and involvement more likely where the oncology team were proactively addressing palliative care needs (e.g. evidence of advance care planning). There was no indication from the studies included about why children with certain cancers are less likely to receive specialist palliative care, and a recent review of barriers to access did not explore this.[31] Evidence from adult cancer[91] indicates that the remitting and relapsing trajectories of haematological malignancies, a more aggressive approach to treatment and greater difficulties predicting prognosis may contribute to the observed inequity of specialist palliative care involvement.[92] Evidence from our review regarding clinician uncertainty

about when to involve specialist palliative care, combined with the commonly reported practice of referring children with a poor prognosis,[27] suggests this might be the case.

Clinicians' views regarding the need for specialist palliative care input for their patients and their acceptability of specialist palliative care services were also consistently reported as affecting referral practices. More specifically, views on how these specialist services differ to palliative care provided by the oncology team, perceived drawbacks associated with involving a new team, readiness to accept a change in prognosis and the negative connotations associated with the term 'palliative care' were identified as barriers to referral. Concerns among clinicians about how families might react to the offer of a referral was a common theme in several studies, and although the study that explored young people and parents' views runs counter to this, wider work on this issue reports mixed opinions among young people[93,94] and parents[73,74] about palliative care and how this should be introduced.[95,96]

Strengths and limitations

This review is the first to systematically synthesise the available evidence about specialist paediatric palliative care for children and young people with cancer. Strengths of the review include a published protocol, robust search, independent screening and data extraction by two reviewers, and the use of appropriate mixed-methods techniques to synthesise the results. There are, however, limitations in the conclusions which can be drawn from this review due to the heterogeneity of study populations and interventions. This, and the substantial risk of potential bias identified in some of the studies and inconsistency of measurement across studies, meant it was inappropriate to aggregate the results statistically, or to compare results between the different approaches to providing specialist palliative care. In addition, the descriptions of specialist services and the palliative care provided by oncology teams were typically very poor, making interpretation of the differences between these challenging.

What this review adds

Our finding that end-of-life care is different for those who receive specialist paediatric palliative care compared to those who do not broadly aligns with three recent reviews about children with all life-limiting conditions.[19,28,29] Two of these reviews[19,29] concluded that specialist intervention appears to offer benefit in terms of improved quality of life. By focusing only on children and young people with cancer, our review highlights the lack of robust evidence pertaining to both quality of life and symptom burden in this population. Although the broader literature implies that other differences we observed, such as reduced hospital stays and more advance care planning, are indicative of better care,[63] there is no evidence that these changes in the delivery of care reflect family preferences,[48,51,57] or lead to reduced symptom burden, which studies continue to show is significant for children with cancer.[5,90,97]

The integration of findings from evaluation studies and those which have explored factors affecting access tells us that while end-of-life care may be different for children who receive specialist palliative care, children who receive this are also different to those who do not, particularly in terms of their disease profile and care processes. Synthesis of the qualitative studies go some way to explaining these differences, highlighting in particular the central role of clinicians' emotions, beliefs and attitudes in shaping referral practices, and the ongoing uncertainty about when to initiate palliative care and whether this should be provided by a specialist service or the oncology team. It also reveals that, in practice, decisions and discussions about no longer pursuing curative treatments and introducing specialist palliative care go hand in hand, and that families who are comfortable discussing an uncertain future or families cared for by clinicians who have this confidence may be more likely to receive specialist palliative care.[75,77,83]

Interventions that support clinicians to initiate palliative care with families and improve clinical acceptability of specialist palliative care services therefore offer the potential to address these key barriers to access.[82,98] Although there are various initiatives to support the delivery of palliative care within oncology services (e.g. palliative care training,[99,100] communication tools,[101,102] early integration models[26,67,69]), we know very little about whether these are being implemented in practice and how they might influence referral to specialist palliative care. There is also little available evidence about whether families play an active role in the initiation of palliative care, or whether clinicians' concerns about how families will respond to a referral are warranted.[68]

Future research should therefore investigate the effectiveness, delivery and acceptability of the different models of delivering palliative care for children and young people with cancer, particularly outside of North America. Future research also needs to examine factors affecting uptake from families' perspectives and to explore the role of socioeconomic factors. This conclusion is supported by the recent priority setting partnership results for teenagers and young adults with cancer,[103] which includes how best to support young people who have incurable cancer and their families. In order to undertake this research, we must first determine what outcomes are the most important to measure and develop appropriate tools to measure them.[104–107] Development of a core outcome set would meet this requirement.[108] This too will need to include the views of children and young people and their families, particularly if we are to address the methodological challenges that continue to affect the quality of research in this area, and the lack of evidence about whether specialist paediatric palliative care improves quality of life for children and their families.

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Author contributions

B.B., B.P. and L.F. conceived the original study. J.T., A.B., B.B., B.P., K.W. and L.F. contributed to the design and methods. K.W. prepared the search strategy. J.T., B.B., B.P., A.B. and L.F. screened studies for inclusion. J.T. and A.B. carried out data extraction. J.T., A.B. and L.F. conducted the critical appraisals. J.T. conducted the synthesis with input from A.B., B.B., B.P. and L.F. J.T. drafted the manuscript with contributions from A.B., B.B., B.P., K.W. and L.F. The final manuscript was approved by all authors.

Data management and sharing

Data are reported fully in the included tables and figures and are derived from published sources only.

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Supplemental material

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