The under reporting of recruitment strategies in research with children with life-threatening illnesses: A systematic review

Briony F Hudson1,2, Linda JM Oostendorp1, Bridget Candy2, Victoria Vickerstaff2, Louise Jones2, Monica Lakhanpaul3, Myra Bluebond-Langner1 and Paddy Stone2

Abstract

Background: Researchers report difficulties in conducting research with children and young people with life-limiting conditions or life-threatening illnesses and their families. Recruitment is challenged by barriers including ethical, logistical and clinical considerations.

Aim: To explore how children and young people (aged 0–25 years) with life-limiting conditions or life-threatening illnesses and their families were identified, invited and consented to research published in the last 5 years.

Design: Systematic review.

Data sources: MEDLINE, PsycINFO, Web of Science, Sciences Citation Index and SCOPUS were searched for original English language research published between 2009 and 2014, recruiting children and young people with life-limiting conditions or life-threatening illness and their families.

Results: A total of 215 studies – 152 qualitative, 54 quantitative and 9 mixed methods – were included. Limited recruitment information but a range of strategies and difficulties were provided. The proportion of eligible participants from those screened could not be calculated in 80% of studies. Recruitment rates could not be calculated in 77%. A total of 31% of studies recruited less than 50% of eligible participants. Reasons given for non-invitation included missing clinical or contact data, or clinician judgements of participant unsuitability. Reasons for non-participation included lack of interest and participants’ perceptions of potential burdens.

Conclusion: All stages of recruitment were under reported. Transparency in reporting of participant identification, invitation and consent is needed to enable researchers to understand research implications, bias risk and to whom results apply. Research is needed to explore why consenting participants decide to take part or not and their experiences of research recruitment.

Keywords

Child, palliative care, research design, research report, patient selection, review

What is already known about this topic?

- Recruitment to research with children and young people with life-limiting conditions or life-limiting illnesses is challenged by factors including limited researcher access to participants, ethical considerations and characteristics of the population.

What this paper adds?

- Recruitment strategies are not consistently reported in current research with this population.
- Inadequate reporting of recruitment practices limits our capacity to judge study quality, risk of bias, representativeness of samples, generalizability of results and ultimately the applicability of findings.

1Louis Dundas Centre for Children’s Palliative Care, UCL Institute of Child Health, London, UK
2Marie Curie Palliative Care Research Department, UCL Division of Psychiatry, London, UK
3Population, Policy and Practice Programme, UCL Institute of Child Health, London, UK

Corresponding author:
Briony F Hudson, Marie Curie Palliative Care Research Department, UCL Division of Psychiatry, 6th Floor, Maple House, 149 Tottenham Court Road, London W1T 7NF, UK.
Email: b.hudson@ucl.ac.uk
Introduction

There is an international need for research of all types involving children and young people aged 0–25 years (CYP) with life-limiting conditions (LLC; conditions for which there is no reasonable hope of cure and from which children or young people will die) or life-threatening illnesses (LTI; conditions for which curative treatment may be feasible but can fail) and their families. Research is required to explore pain and symptom management, decision making about care and treatment, illness experience, and service development and delivery.

A number of challenges to recruitment for research with this group have been reported in the literature. These include clinical considerations such as the often unpredictable course of the illnesses, limited access to potential participants as a result of both logistical factors and paternalistic attitudes, the perceived potential burden on participants and difficulties securing ethical approval. As a result, recruitment to research may be slow and selective. This may affect the quality of research, risk of bias and the generalizability of findings.

There is debate about when and how, and even if, CYP with LLC/LTI and their families might be invited to take part in research. At the same time, there is also growing evidence that CYP with LLC/LTI and their families value opportunities to participate in research and that this can be a positive experience for them.

Internationally, the culture of research is changing as the importance of establishing robust evidence for care is increasingly understood. In the United Kingdom, the National Institute for Health Research (NIHR) has established a Clinical Research Network for Children, and CYP and their families are being encouraged to share their views and participate in the design, review and conduct of research to ensure that it is valid, feasible and acceptable to potential participants.

CYP with LLC/LTI are likely to receive a palliative approach to care, often alongside active treatments. Interest in research on both adult and children’s palliative care is gaining momentum and guidance has been developed for its design and conduct. The MORECare (Methods of Researching End of Life Care) statement provides best practice solutions specifically for planning and conducting palliative care research, as well as the reporting of attrition data. However, it appears that such guidance is not routinely adhered to and a lack of reporting of methodological information has been identified in research with CYP with LLC/LTI and their families.

In this systematic review, we explored the reported methods of participant identification, invitation and recruitment of CYP with LLC/LTI and their families to research, conducted internationally and published over the last 5 years (September 2009 to September 2014). We wished to understand the challenges to recruitment, how these differ between types of studies, identify areas of good practice and provide evidence for areas in which improvements might be made.

Aims and objectives

We aimed to explore methods of recruitment of CYP with LLC/LTI and their families to qualitative, quantitative and mixed-methods research reported in peer-reviewed journal articles.

Our objectives were as follows:

1. To document the procedures for identifying, inviting and consenting eligible CYP with LLC/LTI and their families to research;
2. To document recruitment rates;
3. To identify reasons given for non-enrolment, both reasons given by researchers and clinicians for not approaching eligible participants, and those given by potential participants for deciding not to take part;
4. To explore whether recruitment differs between types of studies;
5. To explore what barriers and facilitators to research recruitment were highlighted by the authors of reviewed studies.

Methods

Inclusion criteria

Primary studies of all methodologies (quantitative, qualitative and mixed-method design) were included. Eligible studies reported research recruiting CYP with LLC/LTI (and/or their family members), were written in English and...
published between September 2009 and September 2014. Conference abstracts were not included. We applied the United Nations definition of ‘young people’, extending from birth to 25 years. Studies recruiting parents, grandparents or siblings of CYP with LLC/LTI were also included. The Richard Hain Directory\textsuperscript{23} of International Classification of Diseases, 10th Revision (ICD-10) diagnoses was used to ascertain whether diagnoses could be considered life-limiting or life-threatening; where definitions were unclear, the opinion of a clinical expert in paediatric palliative care (M.C.) was sought.

**Search strategy**

A systematic search of the literature was performed in the following: MEDLINE, PsycINFO, Web of Science, Sciences Citation Index and SCOPUS in September 2014. A combination of indexed and free-text terms was used to reflect the three components forming the search strategy (CYP; LLC/LTI recruitment).

Descriptive terms that have been used previously by other systematic reviews\textsuperscript{24,25} relating to all stages of youth were used. Terms relating to palliative care, death, bereavement and recruitment practices were also included. The MEDLINE search strategy is shown in Appendix 1.

**Data screening**

Two reviewers (B.F.H. and L.J.M.O.) screened citations against the inclusion criteria. Disagreements regarding eligibility were resolved through reading full text articles and discussion.

**Data extraction**

The data extraction tool was piloted by B.F.H. and L.J.M.O., and minor adjustments were made. We sought to extract the following from each study: research design, recruitment location and setting, funding source and type of body providing ethical review, sample characteristics, and numbers of participants screened, identified as eligible, invited to participate and consented. We also extracted reasons for the non-invitation of eligible participants and reasons given by eligible participants for non-participation. Data were extracted independently by one of four reviewers (B.F.H., L.J.M.O., V.V. or B.C.). A sample of data extracted by each reviewer was checked for accuracy and consistency by another reviewer.

Where data that we wished to extract could not be found in the article, we categorized this as ‘not reported’. No attempts were made to obtain this information by contacting the authors. We did not assess, alongside data extraction, the overall quality of each study using recommended checklists as we wished to explore the quality of reporting of recruitment information, rather than quality of the research itself.

If provided, descriptions of researchers’ experiences of recruiting participants were also extracted. We were interested in strategies employed by authors that aimed to facilitate recruitment and any issues encountered which hindered the recruitment processes. This information was usually found in the discussion section of the included papers. These data were not available in all reviewed studies.

**Data analysis**

**Quantitative analysis**

We used descriptive statistics to summarize the type of studies included, the recruitment strategies employed and the recruitment rates achieved. If we had found sufficient levels of reporting of items of interest, we planned to conduct comparative statistical analyses to assess differences in recruitment rates between (1) studies with different types of aims, such as intervention studies and psychosocial studies and (2) studies using different participant invitation methods, for example, an invitation letter in comparison to inviting participants in person.

**Qualitative analysis**

Any text specifically on researchers’ experiences of recruiting their sample was identified and analysed. We applied thematic synthesis as outlined by Thomas and Harden\textsuperscript{26} and described in Langford et al.\textsuperscript{27} to these informal data. Thematic synthesis is an adaptation of thematic analysis and can be used to pool qualitative data across different studies. Three stages of the recruitment process were used as a coding (grouping) framework: (1) identifying or screening participants, (2) inviting or approaching them to participate and (3) obtaining consent. Codes relating to the barriers and facilitators experienced in relation to each of these stages emerged from the data. Coding was completed in NVivo 10 by one reviewer (B.F.H.) and reviewed by two further reviewers (B.C. and L.J.M.O.), disagreements were resolved through discussion.

**Results**

**Literature search**

Figure 1 presents a Preferred Reporting items for Systematic Reviews and Meta-Analyses (PRISMA) flow diagram of the inclusion process. Our initial search yielded 6419 citations, 215 of these met all criteria and were included in this review. Appendix 2 lists references for all included studies.

**Study types**

Table 1 summarizes the research aims of reviewed studies.
Context and setting. Nearly half of reviewed studies were conducted in North America (46%), a quarter in Europe (26%), 10% in Asia, 7% in Australia and New Zealand, 6% in Africa, 1% in South America and a minority across multiple countries (4%). The majority of research was conducted within hospitals (83%).

Sample. Over half of studies recruited only CYP (56%), over a third recruited only family members (35%) and the remaining 11% recruited both CYP and family members. Young people aged 12–25 years were the least represented with 7% of studies recruiting this age group exclusively. The ages of CYP were not reported in a consistent manner, making descriptions of the ages of participants difficult. Ages of CYP with LLC/LTI were reported in 76% of all studies. Mean ages reported ranged from 19 min of life to 21.1 years.

CYP with malignant and non-malignant conditions were roughly equally represented (malignant: 45%; non-malignant: 44%; and both conditions: 11%). The majority of studies recruited families whose child was alive at the
start of the study (79%). In almost half of the 45 studies (47%), recruiting bereaved families, recruitment occurred more than 1 year after bereavement (Table 2).

**Study design.** Most studies were quantitative (71%), 25% were qualitative, while mixed-methods studies were uncommon (4%). Over a quarter of studies (27%) used interviews, 27% used experimental methods, 23% used questionnaires, 19% recorded physiological measures and 4% of studies used other methods.

**Recruitment strategies – procedures used for identifying, inviting and consenting eligible patients and their families to research**

Information concerning the methods of participant recruitment was not often reported (Table 3).

**Identification of potential participants.** Over half of the studies did not report how potential participants were identified (64%). In the 78 studies reporting this, nearly one-third identified potential participants using medical records (29%) and a quarter used clinic attendance (26%).

Less than a quarter of studies reported the person responsible for identifying potential participants (24%). Physicians identified potential participants in nearly half of the studies providing this information (48%). Studies evaluating an intervention or observing the course of an illness were least likely to report how potential participants were identified, whereas those evaluating current practice were most likely to report their methods for participant identification.

**Invitation of participants.** Over three quarters of studies did not report who invited potential participants (76%). In research evaluating an intervention, including treatment for disease, 96% of studies did not provide this information. In studies reporting this, it was most often a researcher (40%) or a physician (20%). Across all studies, 68% did not report the methods used for participant invitation, over half of those reporting this invited participants by letter (54%) and around a third invited participants in person (32%).

**Consenting of participants.** All studies required informed consent prior to participation; however, specifically who provided consent was not reported in 22% of studies. The majority of studies recruiting both CYP and their family (that provided this information) obtained CYP consent/assent in additional to parental consent (78%).

**Recruitment rates and completeness of reporting**

Numbers of potential participants screened and the proportion found to be eligible. The numbers of participants...

---

**Table 2.** Summary of characteristics of reviewed studies, n (%).

| Sample, n (%) | All studies (n = 215) | Evaluating an intervention (n = 71, 33%) | Assessing quality of life (n = 22, 10%) | Observing course of illness (n = 36, 17%) | Exploring views or perspectives (n = 60, 28%) | Evaluating current practice (n = 26, 12%) |
|--------------|----------------------|------------------------------------------|----------------------------------------|------------------------------------------|------------------------------------------|------------------------------------------|
| CYP only     | 116 (56)             | 66 (93)                                  | 6 (27)                                 | 34 (95)                                  | 9 (16)                                   | 1 (4)                                   |
| Family members only | 75 (35)          | 2 (3)                                    | 11 (50)                                | 2 (5)                                    | 43 (71)                                  | 17 (65)                                 |
| Both CYP and family members | 24 (11)          | 3 (4)                                    | 5 (23)                                 | –                                        | 8 (13)                                   | 8 (31)                                  |
| CYP alive at recruitment | 170 (79)     | 70 (98)                                  | 17 (77)                                | 33 (92)                                  | 37 (62)                                  | 13 (50)                                 |
| Diagnosis, n (%) |                      |                                          |                                        |                                          |                                          |                                          |
| Malignant    | 96 (45)              | 33 (47)                                  | 8 (36)                                 | 12 (33)                                  | 27 (45)                                  | 16 (62)                                 |
| Non-malignant| 95 (44)              | 38 (53)                                  | 9 (41)                                 | 23 (64)                                  | 23 (38)                                  | 2 (8)                                   |
| Mixed malignant or non-malignant | 24 (11)    | –                                        | 5 (23)                                 | 1 (3)                                    | 10 (17)                                  | 8 (30)                                  |
| Design        |                      |                                          |                                        |                                          |                                          |                                          |
| Qualitative   | 54 (25)              | 2 (3)                                    | 3 (14)                                 | 1 (3)                                    | 41 (68)                                  | 7 (27)                                   |
| Quantitative  | 152 (71)             | 69 (97)                                  | 16 (73)                                | 35 (97)                                  | 16 (27)                                  | 16 (62)                                 |
| Mixed methods | 9 (4)                | –                                        | 3 (14)                                 | –                                        | 3 (5)                                    | 3 (11)                                   |
| Methodology, n (%) |                      |                                          |                                        |                                          |                                          |                                          |
| Interviews    | 57 (27)              | –                                        | 7 (32)                                 | 3 (8)                                    | 39 (65)                                  | 8 (31)                                   |
| Questionnaires| 49 (23)              | –                                        | 15 (68)                                | 2 (6)                                    | 17 (28)                                  | 15 (58)                                 |
| Physiological measures | 40 (19) | 10 (14)                                  | –                                      | 29 (80)                                  | –                                        | 1 (4)                                   |
| Experimental (randomized) | 29 (12) | 29 (41)                                  | –                                      | –                                        | –                                        | –                                       |
| Experimental (non-randomized) | 33 (15) | 32 (45)                                  | –                                      | 1 (3)                                    | –                                        | 1 (4)                                   |
| Other         | 8 (4)                | –                                        | –                                      | 1 (3)                                    | 4 (7)                                    | 1 (4)                                   |

CYP: children and young people.
screened for eligibility were not consistently reported; over three quarters of studies did not report this (77%) (Table 4).

It was not possible to calculate the proportion of eligible participants out of those screened in 80% of studies. Where this proportion could be calculated, 24% found less than half of those screened to be eligible and 76% found over half of screened to be eligible for participation.

Proportion approached out of those eligible. The proportion of eligible participants approached was not reported in nearly three quarters of studies (74%). Of the minority of studies providing this information (n = 55), all approached over 50% and the majority approached between 75% and 100% of individuals eligible for participation (93%).

Proportion consented out of those approached. The proportion of eligible participants consented out of those approached could not be calculated in over half of the studies (62%). Of the 82 studies providing this information, the majority consented more than half of approached participants (84%), while 16% recruited less than half those approached.

Recruitment rate (proportion recruited out of those eligible). Just over 10% of studies reported recruitment targets; of these, 71% met their target. Over three quarters (77%) of studies did not provide the necessary information to calculate recruitment rates. In the 49 studies where this was possible, around a third recruited less than 50% of eligible participants (31%).

Reasons for non-enrolment
A total of 19% of studies approached all eligible participants. In 7% of studies, all invited participants decided to...
Despite the majority of studies not approaching all eligible participants, 66% did not report reasons for this and 80% did not report reasons given by potential participants for deciding not to participate.

**Reasons for not approaching eligible participants (non-invitation).** Of the 20% of studies reporting reasons for not contacting eligible participants, the most commonly cited were missing participant data (either clinical or contact, \(n = 25\)), judgements from clinicians of participant unsuitability (\(n = 14\)), unanticipated death (\(n = 13\)) or caregivers denying approach (\(n = 9\)). Other reasons included logistical considerations (e.g. distance participants lived from hospital, \(n = 7\)), researchers’ perception of participant unsuitability (\(n = 2\)) and communication difficulties (\(n = 2\)).

**Reasons given by participants for non-enrolment (non-participation).** For studies in which not all eligible participants were recruited, 14% reported reasons given by potential participants for non-participation. Lack of interest was the most commonly reported reason (\(n = 10\)), followed by participants’ perception of practical (\(n = 9\)) and psychological burdens associated with research participation (\(n = 6\)). Personal reasons (\(n = 5\)), the child’s condition (\(n = 3\)), caregivers’ perceptions that their child needed their attention (\(n = 3\)) and refusal to consent to medical/surgical protocols (\(n = 1\)) or to randomization (\(n = 1\)) were also reported.

---

**Researchers’ experiences of recruitment**

Text regarding researchers’ experiences of recruitment were extracted from 58 studies (26%). This was coded thematically in relation to stages of participant recruitment.

**Identification of potential participants.** The unpredictable course and nature of illnesses and differing institutional policies and practices across research sites were reported as barriers to the identification of potentially eligible participants in four studies. Strategies used to facilitate the identification of eligible participants included widening inclusion criteria (e.g. changing the number of years since bereavement) or seeking support from lay steering groups (‘The parent advisors had particular interest in improving palliative care services and programs and volunteered to assist’).

**Invitation of participants.** Perceived barriers to the invitation of potential participants included gatekeeping from both professionals (‘staff … often chose not to approach families where the child was at the end of life’) and parents (‘In most cases the mother received the initial telephone call … This evolved as another layer of gatekeeping’). Logistical factors such as being unable to contact potential participants were also reported as barriers (‘Many young people were not consistently engaging with...')

---

**Table 4. Recruitment rates, numbers of participants screened, found to be eligible, approached and consented in all studies.**

| All studies, \(n(\%)\) | Evaluating an intervention \((n=71, 33\%)\) | Assessing quality of life \((n=22, 10\%)\) | Observing course of illness \((n=36, 17\%)\) | Exploring views or perspectives \((n=60, 28\%)\) | Evaluating current practice \((n=26, 12\%)\) |
|-------------------------|--------------------------------|--------------------------------|--------------------------------|--------------------------------|--------------------------------|
| **Numbers of potential participants screened** | | | | | |
| \(<50\) | 7 (3) | 4 (6) | – | 1 (3) | 1 (2) | 1 (4) |
| \(50–100\) | 8 (4) | 2 (3) | 1 (5) | 2 (6) | 1 (2) | 2 (8) |
| \(101–1000\) | 29 (14) | 10 (14) | 4 (18) | 5 (15) | 7 (12) | 3 (12) |
| \(>1000\) | 5 (2) | 2 (3) | – | 2 (6) | – | 1 (4) |
| Not reported | 166 (77) | 53 (75) | 17 (77) | 26 (72) | 51 (85) | 19 (73) |
| **Percentage of eligible participants out of those screened** | | | | | |
| \(0–50\) | 10 (5) | 5 (7) | 1 (5) | 2 (6) | 1 (2) | 2 (8) |
| \(51–100\) | 31 (15) | 10 (16) | 3 (14) | 6 (17) | 4 (7) | 7 (27) |
| Not reported | 173 (80) | 55 (77) | 18 (81) | 16 (44) | 55 (92) | 17 (65) |
| **Proportion approached out of those eligible (%)** | | | | | |
| \(0–50\) | – | – | – | – | – | – |
| \(51–100\) | 55 (26) | 18 (25) | 5 (23) | 8 (22) | 16 (26) | 8 (31) |
| Not reported | 160 (74) | 53 (75) | 17 (77) | 28 (78) | 44 (73) | 18 (69) |
| **Proportion consented out of those approached (%)** | | | | | |
| \(0–50\) | 13 (6) | 3 (4) | 1 (5) | – | 5 (9) | 4 (16) |
| \(51–100\) | 69 (32) | 24 (34) | 5 (23) | 13 (72) | 18 (29) | 8 (34) |
| Not reported | 133 (62) | 44 (62) | 16 (73) | 21 (58) | 37 (62) | 14 (54) |
| **Recruitment rate (percentage recruited out of those eligible)** | | | | | |
| \(0–50\) | 15 (7) | 8 (11) | 2 (10) | 2 (6) | 1 (2) | 2 (8) |
| \(51–100\) | 34 (16) | 12 (17) | 4 (18) | 7 (19) | 6 (10) | 5 (19) |
| Not reported | 166 (77) | 51 (72) | 16 (73) | 27 (75) | 53 (88) | 19 (73) |
Strategies to facilitate invitation and recruitment processes included considerations of the method and timing of invitations (‘6-24 months after bereavement was chosen to facilitate recall whilst being sensitive to the emotional requirements of parents’), outlining participants’ options during the invitation period (‘Providing opportunities for parents or formal carers to be present during meetings was welcomed by both the young people and parents/formal carers’30) or providing monetary incentives.

Consenting of participants. Barriers to obtaining consent included overcoming participants’ attitudes or preconceptions towards research (‘Some parents felt that their son/daughter would be unable to participate as they were either non-verbal or had severe learning disabilities’30), logistical factors and CYP characteristics (‘Symptoms such as fatigue can keep children in critical condition from participating in research’31)

Methods found to facilitate the consenting process included incorporating a degree of flexibility (‘Rather than press for a decision on participation, they [participants] were advised they would be contacted after a week or so to discuss the study further’32), the attributes of the research team (‘Recruitment to the study depended on the appointment of an appropriately qualified and experienced research officer’39) and attitudes of potential participants (‘The majority of families treated in other hospitals … approached the physician themselves to discuss the possibility of an autopsy for research aims’33).

Discussion

This review explored systematically how CYP with LLC/LTI and their families have been identified, invited and consented to research, internationally, in the last 5 years.

Statement of principle findings

The majority of reviewed studies did not describe the methods employed in the identification, invitation or consenting of participants or the numbers considered or approached at each stage of the recruitment process. The lack of reporting observed that the proportion of eligible participants recruited (recruitment rates) could not be calculated in over three quarters of studies. This hindered our exploration of differences in recruitment practices between different types of research and to identify areas of good practice.

Where such information was available, we found that the documented reasons for non-invitation included missing clinical or contact data, or clinician judgements of participant unsuitability. The documented reasons for eligible patients deciding not to take part included lack of interest and participants’ perceptions of potential burdens.

The observed lack of reporting practices has implications for the interpretation and generalizability of the current evidence base underpinning the care and treatment of CYP with LLC/LTI and their families. Research with this population is open to the potential for bias for a number of reasons, whether this is due to characteristics of the population, the environment in which it is conducted or the different parties involved in their care. Without knowing who was considered and ultimately included or excluded from research, we cannot be sure that the results obtained are valid, generalizable and relevant for the populations that we are interested in. Moving forward research needs to be reported transparently in order for readers to be able to draw their own conclusions about how to use the information available.

Relationship with previous research and current reporting standards

Clinicians were most often reported to be responsible for the identification of potentially eligible participants and there is potential for this process to be influenced by clinician gatekeeping. As 80% of reviewed studies did not report how many screened participants were identified as eligible, we cannot expand further on this based on the findings of this review.

Strategies to overcome barriers to the invitation of participants reported included obtaining advice from steering groups and parent advisory committees. This is in line with current guidance advocating for the involvement of patients and the public in research design and conduct.16

Guidance and standards are in place for the conduct and reporting of research44 including the CONSORT (Consolidated Standards of Reporting Trials) statement,35 the TREND (Transparent Reporting of Evaluations with Nonrandomized Designs) guidelines,36 STROBE (STrengthening the Reporting of OBservational studies in Epidemiology) guidelines37 and for research with palliative populations, the MORECare Statement.21,38

While the CONSORT statement asks authors to provide information about the numbers of participants screened, identified as eligible and to provide reasons for excluding participants, not all journals require authors to follow reporting guidelines. Indeed information required by the CONSORT is not consistently reported in paediatric randomized controlled trials.39,40 This lack of adherence to guidelines could go some way to explaining the inconsistencies and inadequacies of reporting evidenced in this review.

The impact of inadequate reporting in research

Much research conducted with CYP with LLC/LTI and their families acknowledges its limited generalizability,
but fails to report precisely to whom the research is generalizable to. Without the provision of more transparent information about the recruitment of CYP with LLC/LTI, it will be difficult to develop a sound understanding of the nature of barriers to research and for solutions to be generated and shared.

Our objective to explore what could be learnt from current research regarding recruitment practices was limited by a lack of reporting. Such learning is important to enhance success of future projects, thereby reducing waste and unnecessary exposure of CYP to suboptimal practices. Inadequate reporting of recruitment practices limits our capacity to judge study quality, risk of bias, representativeness of samples, generalizability of results and ultimately the applicability of findings. This has worrying implications for the policies which are underpinned by the current evidence base.

**Strengths and limitations**

The lack of reporting about recruitment processes meant that while we were able to conduct a narrative synthesis of the data, and a thematic synthesis of text from the discussion section of included papers provided, it was not possible to conduct an analysis of the association between research aim, recruitment strategies and recruitment rates.

We recognize that one potential explanation for the lack of information about recruitment processes may be related to journal constraints and word limits. In addition, for some variables, such as reasons for non-participation, researchers may not have access to this information, or may not have had the ethical approvals to collect and record data on non-participation. Nonetheless, the rate of recruitment reporting is lower than we had anticipated.

Our review was limited to studies published in English over the last 5 years, which may have introduced the potential for a language or publishing bias; however, studies from a range of countries were included. We restricted ourselves to the last 5 years for both practical reasons (limited resources) and in order to focus on the most current reporting practices. We did not include studies with mixed populations (studies recruiting CYP with LLC/LTI plus healthy controls or professionals involved in their care). Given the uniqueness of this population, we wanted to focus exclusively on CYP with LLC/LTI and their families. We are unable to comment on the reporting of recruitment practices in studies recruiting more heterogeneous populations.

The inclusion and analysis of informal data in systematic reviews are not common but have been successfully implemented in a few pioneering studies. Informal evidence provided a richer, fuller picture of recruitment than formal evidence alone. However, as relevant text was extracted from just over a quarter of reviewed studies (26%), the potential for biases within the data reviewed exists.

A potential criticism of this review could be the grouping of both medical (e.g. drug trials) and psychosocial studies under the research aim ‘evaluating an intervention’. We acknowledge that requirements and methodologies of studies within this category vary, yet these are the only studies for which there is the potential for direct physical participant benefit. The majority of studies within this category evaluated a medical intervention and from the data reported, there appears to be minimal differences between research with different aims in terms of recruitment methodologies used and recruitment rates achieved.

**Recommendations for reporting of recruitment**

In order to strengthen the evidence base and inform the development of future research and policies to improve the care and treatment of CYP with LLC/LTI and their families, greater clarity is needed in the reporting of research conducted with this population.

Future research should endeavour to provide transparent accounts of participant recruitment. In order to achieve this goal, we would urge authors to report recruitment methods and practices and we would encourage journals to make this part of the manuscript submission requirements. The use of online supplementary material facilities should be used where authors are constrained by word limits.

**Implications for future research**

Based on the content of recently published papers, we cannot judge the impact of different recruitment strategies or the extent of impact of the barriers reported due to the inadequacies of current reporting practices. Explorations of the effectiveness of different identification, invitation and recruitment strategies and the impact of flexibility in recruitment may serve to strengthen the evidence base and advance the care and treatment of CYP with LLC/LTI and their families. Further reviews of existing literature, including studies with mixed populations (those recruiting CYP with LLC/LTI plus healthy controls or professionals involved in their care), could prove illuminating.

This review has pooled the experiences of researchers’ recruiting CYP with LLC/LTI and their families to research in an attempt to explore recruitment practices employed, barriers encountered and steps taken to overcome them. Expanding this approach and further drawing upon both researchers’ and participants’ experiences and expertise could enable solutions to be generated, developed, implemented and shared among the research community. Researchers’ recommendations for facilitating the recruitment of this population spoke to the importance of listening to the views and preferences of potential participants with regard to research participation, and accommodating these preferences where possible.

We cannot presume to know what constitutes an unacceptable burden to research participation from the perspective of CYP with LLC/LTI and/or their families. Providing...
consenting participants, in research of all designs, with the opportunity to share their reasons for deciding to participate, and their views on what constitutes a burden or barrier to research could provide meaningful and useful insights into the research experience of this population and could inform the development of future research design and recruitment.

Acknowledgements

The authors acknowledge Margaret Comac (advice regarding whether diagnoses could be considered life-limiting or life-threatening), Lizzie Chambers (advice on interests and concerns of organizations using data about CYP with LLC/LTI and/or their families), Doug Hall (PPI), Kate Hall (PPI), Thines Ganeshamoorthy (PPI) and Grazia Manzotti (assistance with search strategy).

Author contribution

B.F.H. designed and ran the search strategy, screened the articles, extracted data, analysed the results, drafted and approved the final manuscript as submitted. L.J.M.O and B.C. designed the search strategy, screened the articles, extracted data, analysed the results, reviewed the manuscript and approved the final manuscript as submitted. V.V. designed the search strategy, extracted data and approved the final manuscript as submitted. L.J., M.L., M.B.-L. and P.S. conceptualized the study, reviewed the manuscript and approved the final manuscript as submitted.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

The author(s) disclosed receipt of the following financial support for the research, authorship, and/or publication of this article: B.F.H.’s post is supported by The Health Foundation (grant code G25 512821 2LAAB), L.J.M.O.’s post is supported by Great Ormond Street Children’s Charity (G25 513947 2LGC), M.B.-L.’s post is supported by funding from The True Colors Trust (grant code G25 511830 2LGA), M.L. was supported by the National Institute for Health Research (NIHR) Collaboration for Leadership in Applied Health Research and Care (CLAHRCs) North Thames at Bart’s Health NHS Trust. The views expressed are those of the author(s) and not necessarily those of the NHS, the NIHR or the Department of Health. B.C., V.V., L.J. and P.S.’ posts are supported by Marie Curie core funding to the Marie Curie Palliative Care Research Department, UCL, grant MCCC-FCO-11-U.

References

1. Fraser LK, Miller M, Hain R, et al. Rising national prevalence of life-limiting conditions in children in England. Pediatrics 2012; 129(4): e923–e929.
2. Baum D. A guide to the development of children’s palliative care services (Report of a Joint Working Party of the Association for Children with Life-Threatening or Terminal Conditions and Their Families and the Royal College of Paediatrics and Child Health). Bristol: Association for Children with Life-Threatening or Terminal Conditions and their Families, 1997.
3. Keeley PW. Improving the evidence base in palliative medicine: a moral imperative. J Med Ethics 2008; 34(10): 757–760.
4. Duke S and Bennett H. Review: a narrative review of the published ethical debates in palliative care research and an assessment of their adequacy to inform research governance. Palliat Med 2010; 24(2): 111–126.
5. Addington-Hall J. Research sensitivities to palliative care patients. Eur J Cancer Care 2002; 11(3): 220–224.
6. Steele R, Cadell S, Siden H, et al. Impact of research participation on parents of seriously ill children. J Palliat Med 2014; 17(7): 788–796.
7. Hinds PS, Burghen EA and Pritchard M. Conducting end-of-life studies in pediatric oncology. Western J Nurs Res 2007; 29(4): 448–465.
8. Ewing G, Rogers M, Barclay S, et al. Recruiting patients into a primary care based study of palliative care: why is it so difficult? Palliat Med 2004; 18(5): 452–459.
9. Stevens MM, Lord BA, Proctor M-T, et al. Research with vulnerable families caring for children with life-limiting conditions. Qual Health Res 2010; 20(4): 496–505.
10. Tomlinson D, Bartels U, Hendershot E, et al. Challenges to participation in paediatric palliative care research: a review of the literature. Palliat Med 2007; 21(5): 435–440.
11. Hynson JL, Aroni R, Bauld C, et al. Research with bereaved parents: a question of how not why. Palliat Med 2006; 20(8): 805–811.
12. Scott DA, Valery PC, Boyle FM, et al. Does research into sensitive areas do harm? Experiences of research participation after a child’s diagnosis with Ewing’s sarcoma. Med J Aust 2002; 177(9): 507–510.
13. Olcree ME and Mack JW. Research participation experiences of parents of children with cancer who were asked about their child’s prognosis. J Palliat Med 2012; 15(3): 269–273.
14. Hellemans MA, Gurka KK and Hayden GF. A review of the Journal of Pediatrics: the first 75 years. J Pediatr 2009; 155(1): 16–20.
15. Kumar SP. Reporting of pediatric palliative care: a systematic review and quantitative analysis of research publications in palliative care journals. Indian J Palliat Care 2011; 17(3): 202–209.
16. Modi N, Vohra J, Preston J, et al. Guidance on clinical research involving infants, children and young people: an update for researchers and research ethics committees. Arch Dis Child 2014; 99: 887–891.
17. Nuffield Council on Bioethics. Children and clinical research: ethical issues. Nuffield Council on Bioethics: London, 2015.
18. Medical Research Council (MRC). MRC ethics guide. London: MRC, 2004.
19. Royal College of Paediatrics, Child Health: Ethics Advisory Committee. Guidelines for the ethical conduct of medical research involving children. Arch Dis Child 2000; 82(2): 177–182.
20. Bluebond-Langner M, Belasco JB, Goldman A, et al. Understanding parents’ approaches to care and treatment
of children with cancer when standard therapy has failed. *J Clin Oncol* 2007; 25(17): 2414–2419.
21. Higginson I, Evans C, Grande G, et al. Evaluating complex interventions in end of life care: the MORECare Statement on good practice generated by a synthesis of transparent expert consultations and systematic reviews. *BMC Med* 2013; 11(1): 111.
22. Gross CP, Mallory R, Heiat A, et al. Reporting the recruitment process in clinical trials: who are these patients and how did they get there? *Ann Intern Med* 2002; 137(1): 10–16.
23. Hain R, Devins M, Hastings R, et al. Paediatric palliative care: development and pilot study of a ‘Directory’ of life-limiting conditions. *BMC Palliat Care* 2013; 12(1): 43.
24. Beecham E, Candy B, Howard R, et al. Pharmacological interventions for pain in children and adolescents with life-limiting conditions. *Cochrane Database Syst Rev* 2015; 3, http://onlinelibrary.wiley.com/doi/10.1002/14651858.CD001075.pub2/abstract
25. Bluebond-Langner M, Beecham E, Candy B, et al. Preferred place of death for children and young people with life-limiting and life-threatening conditions: a systematic review of the literature and recommendations for future inquiry and policy. *Palliat Med* 2013; 27(8): 705–713.
26. Thomas J and Harden A. Methods for the thematic synthesis of qualitative research in systematic reviews. *BMC Med Res Methodol* 2008; 8: 45.
27. Langford R, Bonell C, Jones H, et al. Obesity prevention and the Health promoting Schools framework: essential components and barriers to success. *Int J Behav Nutr Phys Act* 2015; 12(1): 15.
28. Eilegard A, Steineck G, Nyberg T, et al. Bereaved siblings’ perception of participating in research – a nationwide study. *Psychooncology* 2013; 22(2): 411–416.
29. Robert R, Zhukovsky DS, Mauricio R, et al. Bereaved parents’ perspectives on pediatric palliative care. *J Soc Work End Life Palliat Care* 2012; 8(4): 316–338.
30. Mitchell W and Sloper P. Making choices in my life: listening to the ideas and experiences of young people in the UK who communicate non-verbally. *Children Youth Serv Rev* 2011; 33: 521–527.
31. Gaab EM, Owens RG and MacLeod RD. The voices of young New Zealanders involved in pediatric palliative care. *J Palliat Care* 2013; 29: 186–192.
32. Bradford N, Young J, Armfield NR, et al. A pilot study of the effectiveness of home teleconsultations in paediatric palliative care. *J Telemed Telecare* 2012; 18(8): 438–442.
33. Caretti V, Jansen MH, van Vuurden DG, et al. Implementation of a multi-institutional diffuse intrinsic pontine glioma autopsy protocol and characterization of a primary cell culture. *Neuropathol Appl Neurobiol* 2013; 39: 426–436.
34. APA Publications and Communications Board Working Group on Journal Article Reporting Standards. Reporting standards for research in psychology: why do we need them? What might they be? *Am Psychol* 2008; 63(9): 839–851.
35. Altman DG, Schulz KF, Moher D, et al. The revised CONSORT statement for reporting randomized trials: explanation and elaboration. *Ann Intern Med* 2001; 134(8): 663–694.
36. Des Jarlais DC, Lyles C and Crepaz N. Improving the reporting quality of nonrandomized evaluations of behavioral and public health interventions: the TREND statement. *Am J Public Health* 2004; 94(3): 361–366.
37. Von Elm E, Altman DG, Egger M, et al. The Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) statement: guidelines for reporting observational studies. *BMJ* 2007; 45(4): 247–251.
38. Gysels M, Evans CJ, Lewis P, et al. MORECare research methods guidance development: recommendations for ethical issues in palliative and end-of-life care research. *Palliat Med* 2013; 27(10): 908–917.
39. Brooks R, Higgins G and Webster A. Systematic review of randomized controlled trial quality in pediatric kidney transplantation. *Pediatr Nephrol* 2010; 25(12): 2383–2392.
40. DeMauro SB, Giaccone A, Kirpalani H, et al. Quality of reporting of neonatal and infant trials in high-impact journals. *Pediatrics* 2011; 128(3): e639–e644.
41. Sutcliffe K, O’Mara A, Caird J, et al. Pediatric medication error: a systematic review of the extent and nature of the problem in the UK and international interventions to address it. London: EPPI-Centre, Social Science Research Unit, UCL Institute of Education, University of London, 2014.

**Appendix 1**

**MEDLINE search terms**

1. Children and young people

Neonate* or newborn or new born or infant* or child* or adolescent* or pediatric or paediatric* or baby or babies or toddler* or juvenile* or boy* or girl* OR ‘child’ [MeSH Terms]) OR ‘infant’ [MeSH Terms]) OR ‘adolescent’ [MeSH Terms]) OR ‘pediatrics’ [MeSH Terms].

2. Life-limiting conditions and life-threatening illnesses

palliative) OR life threatening) OR life limiting) OR end of life) OR terminal care) OR terminal illness) OR bereave*) OR death) OR dying) OR ‘palliative care’ [MeSH Terms]) OR ‘terminally ill’ [MeSH Terms]) OR ‘death’ [MeSH Terms]) OR ‘terminal care’ [MeSH Terms]) OR ‘bereavement’ [MeSH Terms].

3. Invitation and recruitment practices

(participat* or recruit* or enrol* or invit* or select* or attitude* to research or accrual or enlist* or non-participat*)) OR (‘patient selection’ [MeSH Terms]) OR ‘patient participation’ [MeSH Terms] OR ‘research subjects’ [MeSH Terms] OR ‘researcher-subject relations’ [MeSH Terms] OR ‘refusal to participate’ [MeSH Terms]).
Appendix 2

References of reviewed studies

Studies aiming to evaluate an intervention

Adams MS, Khan NZ, Begum SA, et al. Feeding difficulties in children with cerebral palsy: low-cost caregiver training in Dhaka, Bangladesh. *Child Care Health Dev* 2012; 38(6): 878–888.

Adzick NS, Thom EA, Spong CY, et al. A randomized trial of prenatal versus postnatal repair of myelomeningocele. *N Engl J Med* 2011; 364: 993–1004.

Akber A, Portale AA and Johansen KL. Use of pedometers to increase physical activity among children and adolescents with chronic kidney disease. *Pediatr Nephrol* 2014; 29(8): 1395–1402.

Albers E, Donahue BS, Milne G, et al. Perioperative plasma F(2)-Isoprostanes levels correlate with markers of impaired ventilation in infants with single-ventricle physiology undergoing stage 2 surgical palliation on the cardiopulmonary bypass. *Pediatr Cardiol* 2012; 33: 562–568.

Almond CS, Morales DL, Blackstone EH, et al. Berlin Heart EXCOR pediatric ventricular assist device for bridge to heart transplantation in US children. *Circulation* 2013; 127(16): 1702–1711.

Altarabshie SE, Dearani JA, Burkhart HM, et al. Outcome of septal myectomy for obstructive hypertrophic cardiomyopathy in children and young adults. *Ann Thorac Surg* 2013; 95: 663–669.

Ancora G, Maranela E, Grandi S, et al. Early predictors of short term neurodevelopmental outcome in asphyxiated cooled infants. A combined brain amplitude integrated electroencephalography and near infrared spectroscopy study. *Brain Dev* 2013; 35(1): 26–31.

Azzopardi DV, Strohm B, Edwards AD, et al. Moderate hypothermia to treat perinatal asphyxial encephalopathy. *N Engl J Med* 2009; 361(14): 1349–1358.

Baker DL, Schmidt ML, Cohn SL, et al. Outcome after reduced chemotherapy for intermediate-risk neuroblastoma. *N Engl J Med* 2010; 363(14): 1313–1323.

Black AK, Sadanala UK, Mascio CE, Hornung CA and Keller BB. Challenges in implementing a pediatric cardiovascular home telehealth project. *Telemed J E Health* 2014; 20: 858–867.

Cefalo G, Massimino M, Ruggiero A, et al. Temozolomide is an active agent in children with recurrent medulloblastoma/ primitive neuroectodermal tumor: an Italian multi-institutional phase II trial. *Neuro Oncol* 2014; 16(5): 748–753.

Chagalukka G, Stanley C, Banda K, et al. Kaposi’s sarcoma in children: an open randomised trial of vincristine, oral etoposide and a combination of vincristine and bleomycin. *Eur J Cancer* 2014; 50: 1472–1481.

Cohen KJ, Gibbs IC, Fisher PG, et al. A phase I trial of arsenic trioxide chemoradiotherapy for infiltrating astrocytomas of childhood. *Neuro Oncol* 2013; 15(6): 783–787.

Cohen KJ, Heideman RL, Zhou TN, et al. Temozolomide in the treatment of children with newly diagnosed diffuse intrinsic pontine gliomas: a report from the Children’s Oncology Group. *Neuro Oncol* 2011; 13(4): 410–416.

Cotton MF, Violari A, Otewome K, et al. Early time-limited antiretroviral therapy versus deferred therapy in South African infants infected with HIV: results from the children with HIV early antiretroviral (CHER) randomised trial. *Lancet* 2013; 382(9904): 1555–1563.

Cuvelier GD, Baker TJ, Peddie EF, et al. A randomized, double-blind, placebo-controlled clinical trial of megestrol acetate as an appetite stimulant in children with weight loss due to cancer and/or cancer therapy. *Pediatr Blood Cancer* 2014; 61(4): 672–679.

Dilli D, Aydin B, Zenciroglu A, et al. Treatment outcomes of infants with cyanotic congenital heart disease treated with synbiotics. *Pediatrics* 2013; 132(4): E932–E938.

Dingley J, Tooley J, Liu X, et al. Xenon ventilation during therapeutic hypothermia in neonatal encephalopathy: a feasibility study. *Pediatries* 2014; 133(5): 809–818.

Dworzak MN, Gaipa G, Schumich A, et al. Modulation of antigen expression in B-cell precursor acute lymphoblastic leukemia during induction therapy is partly transient: evidence for a drug-induced regulatory phenomenon. Results of the AIEOP-BFM-ALL-FLOW-MRD-Study Group. *Cytometry B Clin Cytom* 2010; 78: 147–153.

Foster C, McDonald S, Frize G, et al. ‘Payment by Results’ – financial incentives and motivational interviewing, adherence interventions in young adults with perinatally acquired HIV-1 infection: a pilot program. *AIDS Patient Care STDS* 2014; 28(1): 28–32.

Gohar SF, Comito M, Price J, et al. Feasibility and parent satisfaction of a physical therapy intervention program for children with acute lymphoblastic leukemia in the first 6 months of medical treatment. *Pediatr Blood Cancer* 2011; 56(5): 799–804.

Granger M, Grupp SA, Kletzel M, et al. Feasibility of a tandem autologous peripheral blood stem cell transplant regimen for high risk neuroblastoma in a cooperative group setting: a Pediatric Oncology Group study: a report from the Children’s Oncology Group. *Pediatr Blood Cancer* 2012; 59(5): 902–907.

Hauser PM, Bernard T, Greub G, et al. Microbiota present in cystic fibrosis lungs as revealed by whole genome sequencing. *PLoS ONE* 2014; 9(3): e90934.

Hayes-Jordan A, Green H, Ludvig J, et al. Toxicity of hyperthermic intraoperative chemotherapy (HIPEC) in pediatric patients with sarcomatosis/carcinomatosis: early experience and phase 1 results. *Pediatr Blood Cancer* 2012; 59(2): 395–397.

Hessissen L, Khtar R, Madani A, et al. Improving the prognosis of pediatric Hodgkin lymphoma in developing countries: a moroccan society of pediatric hematology and oncology study. *Pediatr Blood Cancer* 2013; 60(9): 1464–1469.

Hickey EJ, Caldarone CA, Blackstone EH, et al. Biventricular strategies for neonatal critical aortic stenosis: high mortality associated with early reintervention. *J Thorac Cardiovasc Surg* 2012; 144(2): 409–41.

Hoving MA, van Raak EPM, Spincemaille G, et al. Early predictors of short term neurodevelopmental outcome after reduced craniospinal radiotherapy: a children’s oncology group phase I/II study. *J Clin Oncol* 2012; 30(21): 2648–2653.
Jozwiak M, Harasymczuk P, Koch A, et al. Incidence and risk factors of hip joint pain in children with severe cerebral palsy. *Disabil Rehab* 2011; 33(15–16): 1367–1372.

Karaman S, Vural S, Yildirim K, et al. Comparison of piperacillin tazobactam and cefepirone sulbactam monotherapy in treatment of febrile neutropenia. *Pediatr Blood Cancer* 2012; 58(4): 579–583.

Keating JJ, Simsic JM, Kogon BE, et al. Impact of early fundoplication or gastrostomy tube on midterm outcomes for patients with single ventricle. *J Thorac Cardiovasc Surg* 2012; 143: 891–895.

Kishnani PS, Corzo D, Leslie ND, et al. Early treatment with alglucosidase alpha prolongs long-term survival of infants with Pompe disease. *Pediatr Res* 2009; 66(3): 329–335.

Kumar R, Tripathi P, Baranwal V, et al. Randomized, controlled trial of oral ribavirin for Japanese encephalitis in children in Uttar Pradesh, India. *Clin Infect Dis* 2009; 48: 400–406.

Latus H, Binder W, Kerst G, et al. Right ventricular-pulmonary arterial coupling in patients after repair of tetralogy of Fallot. *J Thorac Cardiovasc Surg* 2013; 146(6): 1366–1372.

Lindsey JC, Hughes MD, Violari A, et al. Predictors of virologic and clinical response to nevirapine versus lopinavir/ritonavir-based antiretroviral therapy in young children with and without prior nevirapine exposure for the prevention of mother-to-child HIV transmission. *Pediatr Infect Dis J* 2014; 33(8): 846–854.

Lipshultz SE, Scully RE, Lipsitz SR, et al. Assessment of dexrazoxane as a cardioprotectant in doxorubicin-treated children with high-risk acute lymphoblastic leukaemia: long-term follow-up of a prospective, randomised, multicentre trial. *Lancet Oncol* 2010; 11(10): 950–961.

MacDonald TJ, Vezina G, Stewart CF, et al. Phase II study of cilengitide in the treatment of refractory or relapsed high-grade gliomas in children: a report from the Children’s Oncology Group. *Neuro Oncol* 2013; 15(10): 1438–1444.

McJunkin JE, Nahata MC, Reyes E, et al. Safety and pharmacokinetics of ribavirin for the treatment of la crosse encephalitis. *Pediatr Infect Dis J* 2011; 30(10): 860–865.

Meinhardt A, Burkhardt B, Zimmermann M, et al. Phase II window study on rituximab in newly diagnosed pediatric mature B-cell non-Hodgkin’s lymphoma and Burkitt leukemia. *J Clin Oncol* 2010; 28(19): 3115–3121.

Ndeezi G, Tumwine JK, Bolann BJ, et al. Zinc status in HIV infected Ugandan children aged 1–5 years: a cross sectional baseline survey. *BMC Pediatr* 2010; 10: 68.

Nolbris MJ and Ahlström BH. Siblings of children with cancer – their experiences of participating in a person-centered support intervention combining education, learning and reflection: pre- and post-intervention interviews. *Ear J Otolaryngol Nurs* 2014; 18(3): 254–260.

Novak I, Cusick A and Lannin N. Occupational therapy home programs for cerebral palsy: double-blind, randomized, controlled trial. *Pediatrics* 2009; 124(4): e606–e614.

Ohman A, Stromvall-Larsson E, Nilsson B, et al. Pulse oximetry home monitoring in infants with single-ventricle physiology and a surgical shunt as the only source of pulmonary blood flow. *Cardiol Young* 2013; 23: 75–81.

Okomo U, Tojgun T, Oko F, et al. Mortality and loss to programme before antiretroviral therapy among HIV-infected children eligible for treatment in The Gambia, West Africa. *AIDS Res Ther* 2012; 9: 28.

Pasquali SK, Ohye RG, Lu MM, et al. Variation in perioperative care across centers for infants undergoing the Norwood procedure. *J Thorac Cardiovasc Surg* 2012; 144(4): 915–921.

Pession A, Masetti R, Rizzari C, et al. Results of the AIEOP AML2002/01 multicenter prospective trial for the treatment of children with acute myeloid leukemia. *Blood* 2013; 122: 170–178.

Pickering D, Horrocks LM, Visser KS, et al. ‘Every picture tells a story’: interviews and diaries with children with cerebral palsy about adapted cycling. *J Paediatr Child Health* 2013; 49(12): 1040–1044.

Prado JG, Prendergast A, Thobakgale C, et al. Replicative capacity of human immunodeficiency virus type 1 transmitted from mother to child is associated with pediatric disease progression rate. *J Virol* 2010; 84(1): 492–502.

Radbill AE, Friedman JK, Berul CI, et al. Prospective evaluation of defibrillation threshold and postshock rhythm in young ICD recipients. *Pacing Clin Electrophysiol* 2012; 35(12): 1487–1493.

Ridola V, Barone G, Lazzareschi I, et al. Feasibility study of 21-day-on/7-day-off temozolomide in children with brain tumors. *J Neurooncol* 2011; 103(1): 147–153.

Sakzewski L, Carlson S, Shields N, et al. Impact of intensive upper limb rehabilitation on quality of life: a randomized trial in children with unilateral cerebral palsy. *Dev Med Child Neurol* 2012; 54(5): 415–423.

Salzer WL, Asselin B, Supko JG, et al. Erwinia asperaginase achieves therapeutic activity after pegaspargase allergy: a report from the Children’s Oncology Group. *Blood* 2013; 122(4): 507–514.

Salzer WL, Jones TL, Devidas M, et al. Modifications to induction therapy decrease risk of early death in infants with acute lymphoblastic leukemia treated on Children’s Oncology Group P9407. *Pediatr Blood Cancer* 2012; 59(5): 834–839.

Sandlund JT, Pui CH, Zhou Y, et al. Effective treatment of advanced-stage childhood lymphoblastic lymphoma without prophylactic cranial irradiation: results of St Jude NHL13 study. *Leukemia* 2009; 23: 1127–1130.

Shankaran S, Pappas A, McDonald SA, et al. Predictive value of an early amplitude integrated electroencephalogram and neurologic examination. *Pediatrics* 2011; 128(1): E112–E120.

Simbruner G, Tumwine JK, Bolann BJ, et al. Zinc status in HIV infected Ugandan children aged 1–5 years: a cross sectional baseline survey. *BMC Pediatr* 2010; 10: 68.

Souid AK, Dubowy RL, Ingle AM, et al. A pediatric phase I trial and pharmacokinetic study of ispinesib: a Children’s Oncology Group P9407 trial and pharmacokinetic study of ispinesib: a Children’s Oncology Group phase I consortium study. *Pediatr Blood Cancer* 2012; 55(7): 1323–1328.

Srinivasan A, Kasow KA, Cross S, et al. A pediatric phase I trial and pharmacokinetic study of ibratumomab: a Children’s Oncology Group phase I consortium study. *Pediatr Blood Cancer* 2010; 55(7): 1323–1328.

Srinivasan R, Asselin J, Gildengorin G, et al. A prospective study of ventilator-associated pneumonia in children. *Pediatrics* 2009; 123: 1108–1115.
Sugimoto A, Ota N, Ibuki K, et al. Risk factors for adverse neurocognitive outcomes in school-aged patients after the Fontan operation (dagger). Eur J Cardiothorac Surg 2013; 44: 454–461.

Sung L, Buxton A, Gamis A, et al. Life-threatening and fatal infections in children with acute myeloid leukemia: a report from the Children’s Oncology Group. J Pediatr Hematol Oncol 2012; 34: e30–e35.

Takenouchi T, Rubens EO, Yap VL, et al. Delayed onset of sleep-wake cycling with favorable outcome in hypoergic-treated neonates with encephalopathy. J Pediatr 2011; 159(2): 232–237.

Tomizawa D, Tawa A, Watanabe T, et al. Appropriate dose reduction in induction therapy is essential for the treatment of infants with acute myeloid leukemia: a report from the Japanese Pediatric Leukemia/Lymphoma Study Group. Int J Hematol 2013; 98(5): 578–588.

Uzark K, Wang Y, Rudd N, et al. Interstage feeding and weight gain in infants following the Norwood operation: can we change the outcome? Cardiol Young 2012; 22: 520–527.

Van Poppel M, Klimo P, Dewire M, et al. Resection of infantile brain tumors before neoadjuvant chemotherapy: the St. Jude experience Clinical article. J Neurosurg Pediatr 2011; 8(3): 251–256.

Violari A, Lindsey JC, Hughes MD, et al. Nevirapine versus ritonavir-boosted lopinavir for HIV-infected children. N Engl J Med 2012; 366(25): 2380–2389.

Vora A, Goulden N, Wade R, et al. Treatment reduction for children and young adults with low-risk acute lymphoblastic leukemia defined by minimal residual disease (UKALL 2003): a randomised controlled trial. Lancet Oncol 2013; 14(3): 199–209.

Widemann BC, Arceci RJ, Jayaprakash N, et al. Phase 1 trial and pharmacokinetic study of the farnesyl transferase inhibitor tipifarnib in children and adolescents with refractory leukemias: a report from the Children’s Oncology Group. Pediatr Blood Cancer 2011; 56(2): 226–233.

Zaky W, Dhall G, Ji L, et al. Intensive induction chemotherapy followed by myeloablative chemotherapy with autologous hematopoietic progenitor cell rescue for young children newly-diagnosed with central nervous system atypical teratoid/rhabdoid tumors: the Head Start III experience. Pediatr Blood Cancer 2014; 61(1): 95–101.

Zhou WH, Cheng GQ, Shao XM, et al. Selective head cooling with mild systemic hypothermia after neonatal hypoxic-ischemic encephalopathy: a multicenter randomized controlled trial in China. J Pediatr 2010; 157(3): 367–372, 372.e1–372.e3.

Jones JK, Kamani SA, Bush PJ, et al. Development and evaluation of an educational interactive CD-ROM for teens with cancer. Pediatr Blood Cancer 2010; 55(3): 512–519.

Studies assessing quality of life

Abu-Saad Huijer H, Doumit M, Abboud S, et al. Quality of palliative care. Perspective of Lebanese patients with cancer. J Med Liban 2012; 60(2): 91–98.

Al-Gamal E. Quality of life and anticipatory grieving among parents living with a child with cerebral palsy. Int J Nurs Prac 2013; 19(3): 288–294.

Andrinopoulous K, Clum G, Murphy DA, et al. Health related quality of life and psychosocial correlates among HIV-infected adolescent and young adult women in the US. AIDS Educ Prev 2011; 23: 367–381.

Bhattacharya M, Rajeshwari K and Saxena R. Demographic and clinical features of orphans and nonorphans at a pediatric HIV centre in North India. Indian J Pediatr 2010; 77(6): 627–631.

Bradford N, Young J, Armfield NR, et al. A pilot study of the effectiveness of home teleconsultations in paediatric palliative care. J Telemed Telecare 2012; 18(8): 438–442.

Byrne MW, Evan E, Goshin LS, et al. Parent self-efficacy for managing pain in seriously ill children and adolescents nearing end of life. Palliat Support Care 2011; 9: 137–147.

Cadell S, Kennedy K and Hemsworth D. Informing social work practice through research with parent caregivers of a child with a life-limiting illness. J Soc Work End Life Palliat Care 2012; 8(4): 356–381.

Caeymaex L, Jousselme C, Vlasescu C, et al. Perceived role in end-of-life decision making in the NICU affects long-term parental grief response. Arch Dis Child Fetal Neonatal Ed 2013; 98: F26–F31.

Ceravolo F, Mascaro I, Sestito S, et al. Home treatment in paediatric patients with Hunter syndrome: the first Italian experience. Italian J Pediatr 2013; 39: 53.

Engelen V, Koopman HM, Detmar SB, et al. Health-related quality of life after completion of successful treatment for childhood cancer. Pediatr Blood Cancer 2011; 56(4): 646–653.

Jalmell S, Onelov E, Steineck G, et al. Hematopoietic stem cell transplantation in children with cancer and the risk of long-term psychological morbidity in the bereaved parents. Bone Marrow Transplant 2011; 46(8): 1063–1070.

Knapp C, Madden V, Revicki D, et al. Health status and health-related quality of life in a pediatric palliative care program. J Palliat Med 2012; 15(7): 790–797.

Li HCW, Chung OKJ and Chiu SY. The impact of cancer on children’s physical, emotional, and psychosocial well-being. Cancer Nurs 2010; 33(1): 47–54.

O’Byrne ML, Mercer-Rosa L, et al. Morbidity in children and adolescents after surgical correction of truncus arteriosus communis. Am Heart J 2013; 166(3): 512–518.

Reilly D, Huws J, Hastings R, et al. Life and death of a child with down syndrome and a congenital heart condition: experiences of six couples. Intel Dev Disabil 2010; 48(6): 403–416.

Reisi-Dehkordi N, Baratian H and Zargham-Boroujeni A. Challenges of children with cancer and their mothers: a qualitative research. Iran J Nurs Midwifery Res 2014; 19(4): 334–339.

Rosenberg AR, Postier A, Osenga K, et al. Long-term psychosocial outcomes among bereaved siblings of children with cancer. J Pain Symptom Manage 2015; 49: 55–65.

Schoenmaker NJ, Haverman L, Tromp WF, et al. Children of non-Western origin with end-stage renal disease in the Netherlands, Belgium and a part of Germany have impaired health-related quality of life compared with Western children. Nephrol Dial Transplant 2014; 29: 448–457.

Steele R, Siden H, Cadell S, et al. Charting the territory: symptoms and functional assessment in children with progressive, non-curable conditions. Arch Dis Child 2014; 99: 754–762.
Syczewska M, Dembowska-Baginska B, Perek-Polnik M, et al. Gaits pathology assessed with Gillette Gaits Index in patients after CNS tumour treatment. *Gait Posture* 2010; 32: 358–362.

Tomlinson D, Hinds PS, Bartels U, et al. Parent reports of quality of life for pediatric patients with cancer with no realistic chance of cure. *J Clin Oncol* 2011; 29(6): 639–645.

Whittingham K, Wee D, Sanders MR, et al. Predictors of psychological adjustment, experienced parenting burden and chronic sorrow symptoms in parents of children with cerebral palsy. *Child Care Health Dev* 2013; 39(3): 366–373.

**Studies observing the course of illness**

Anga G, Barnabas R, Kaminiel O, et al. The aetiology, clinical presentations and outcome of febrile encephalopathy in children in Papua New Guinea. *Ann Trop Paediatr* 2010; 30(2): 109–118.

Aron R, Kerkar N, Davis MK, et al. Liver transplantation in children with metabolic diseases: the studies of pediatric liver transplantation experience. *Pediatr Transplant* 2010; 14(6): 796–805.

Badiei Z, Khalesi M, Alami MH, et al. Risk factors associated with life-threatening infections in children with febrile neutropenia: a data mining approach. *J Pediatr Hematol Oncol* 2011; 33(1): E9–E12.

Bonkowski JL, Nelson C, Kingston JL, et al. The burden of inherited leukodystrophies in children. *Neurology* 2010; 75(8): 718–725.

Bucuvalas J, Filipovich L, Yazigi N, et al. Immunophenotype predicts outcome in pediatric acute liver failure. *J Pediatr Gastroenterol Nutr* 2013; 56: 311–315.

Chen I, Khaki L, Lindsey JC, et al. Association of pol diversity with antiretroviral treatment outcomes among HIV-infected African children. *Plos ONE* 2013; 8(11): e81213.

Cheong JLY, Coleman L, Hunt RW, et al. Prognostic utility of magnetic resonance imaging in neonatal hypoxic-ischemic encephalopathy substudy of a randomized trial. *Arch Pediatr Adolesc Med* 2012; 166: 634–640.

Chiu SN, Wu MH, Su MJ, et al. Coexisting mutations/polymorphisms of the long QT syndrome genes in patients with repaired Tetralogy of Fallot are associated with the risks of life-threatening events. *Hum Genet* 2012; 131(8): 1295–1304.

Ciuca IM, Pop L, Tamas L, et al. Cystic fibrosis liver disease – from diagnosis to risk factors. *Rom J Morph Embryol* 2014; 55: 91–95.

Cortez MAA, Scrideli CA, Yunes JA, et al. mRNA expression profile of multidrug resistance genes in childhood acute lymphoblastic leukemia. Low expression levels associated with a higher risk of toxic death. *Pediatr Blood Cancer* 2009; 53(6): 996–1004.

Elgendy HM, Mekawy MA, Wahab S, et al. AC133 Expression in Egyptian children with acute leukemia: impact on treatment response and disease outcome. *J Pediatr Hematol Oncol* 2010; 32: 286–293.

Finkel RS, McDermott MP, Kaufmann P, et al. Observational study of spinal muscular atrophy type I and implications for clinical trials. *Neurology* 2014; 83(9): 810–817.

Fukushima H, Fukushima T, Sakai A, et al. Polymorphisms of MTHFR associated with higher relapse/death ratio and delayed weekly MTX administration in pediatric lymphoid malignancies. *Leukemia Res Treat* 2013; 2013: 238528.

Gamis AS, Alonzo TA, Gerbing RB, et al. Natural history of transient myeloproliferative disorder clinically diagnosed in Down syndrome neonates: a report from the Children’s Oncology Group Study A2971. *Blood* 2011; 118(26): 6752–6759.

Gavidia R, Fuentes SL, Vasquez R, et al. Low socioeconomic status is associated with prolonged times to assessment and treatment, sepsis and infectious death in pediatric fever in El Salvador. *PLoS ONE* 2012; 7(8): e43639.

Gnekwok AK, Falkenstein F, von Hornstein S, et al. Long-term follow-up of the multicenter, multidisciplinary treatment study HIT-LGG-1996 for low-grade glioma in children and adolescents of the German Speaking Society of Pediatric Oncology and Hematology. *Neuro Oncol* 2012; 14(10): 1265–1284.

Goergen SK, Ang H, Wong F, et al. Early MRI in term infants with perinatal hypoxic-ischaemic brain injury: interobserver agreement and MRI predictors of outcome at 2 years. *Clin Radiol* 2014; 69(1): 72–81.

Horn AR, Swingler GH, Myer L, et al. Early clinical predictors of a severely abnormal amplitude-integrated electroencephalogram at 48 hours in cooled neonates. *Acta Paediatrica* 2013; 102: e378–e384.

Lau DT, Hesson LB, Norris MD, et al. Prognostic significance of promoter DNA methylation in patients with childhood neuroblastoma. *Clin Cancer Res* 2012; 18(20): 5690–5700.

Liu FT, Xiong YY, Zhao Y, et al. Identification of aberrant microRNA expression pattern in pediatric gliomas by microarray. *Diagn Pathol* 2013; 8: 158.

Lufi F, Leibundgut K, Niggli FK, et al. Serious medical complications in children with cancer and fever in chemotherapy-induced neutropenia: results of the prospective multicenter SPOG 2003 FN study. *Pediatr Blood Cancer* 2012; 59: 90–95.

Massaro AN, Chang T, Kadom N, et al. Biomarkers of brain injury in neonatal encephalopathy treated with hypothermia. *J Pediatr* 2012; 161(3): 434–440.

Moodley A, Qin M, Singh KK, et al. Vitamin D-related host genetic variants alter HIV disease progression in children. *Pediatr Infect Dis J* 2013; 32: 1230–1236.

Ottosson-Wadlund A, Ceder R, Preta G, et al. Requirement of apoptotic protease-activating factor-1 for bortezomib-induced apoptosis but not for Fas-mediated apoptosis in human leukemic cells. *Mol Pharmacol* 2013; 83: 245–255.

Pietrzyk JJ, Bik-Multanowski M, Balwierz W, et al. Additional genetic risk factor for death in children with acute lymphoblastic leukemia: a common polymorphism of the MTHFR gene. *Pediatr Blood Cancer* 2009; 52: 364–368.

Pisani F, Orsini M, Braibanti S, et al. Development of epilepsy in newborns with moderate hypoxic-ischemic encephalopathy and neonatal seizures. *Brain Dev* 2009; 31: 64–68.

Radman M, Keller RL, Oishi P, et al. Preoperative B-type natriuretic peptide levels are associated with outcome after total cavopulmonary connection (Fontan). *J Thorac Cardiovasc Surg* 2014; 148: 212–219.

Rainwater-Lovett K, Nkamba HC, Mubiana-Mbewe M, et al. Immunologic risk factors for early mortality after starting antiretroviral therapy in HIV-infected Zambian children. *AIDS Res Hum Retroviruses* 2013; 29(3): 479–487.
Rowland M, Gallagher CG, O’Laoide R, et al. Outcome in cystic fibrosis liver disease. *Am J Gastroenterol* 2011; 106(1): 104–109.

Sanchez Mejia AA, Simpson KE, Hildebolt CF, et al. Tissue Doppler septal Tei index indicates severity of illness in pediatric patients with congestive heart failure. *Pediatr Cardiol* 2014; 35: 411–418.

Sundaram V, Dutta S, Ahluwalia J, et al. Score for neonatal acute physiology II predicts mortality and persistent organ dysfunction in neonates with severe sepsis. *Indian Pediatr* 2009; 46: 775–780.

Tzanetos DRT, Yu C, Hernanz-Schulman M, et al. Prospective study of the incidence and predictors of thrombus in children undergoing palliative surgery for single ventricle physiology. *Intensive Care Med* 2012; 38(1): 105–112.

Van Dijk JH, Sutcliffe GF, Munsanje B, et al. HIV-infected children in rural Zambia achieve good immunologic and virologic outcomes two years after initiating antiretroviral therapy. *PloS ONE* 2011; 6(4): e19006.

Westbom L, Bergstrand L, Wagner P, et al. Survival at 19 years of age in a total population of children and young people with cerebral palsy. *Dev Med Child Neurology* 2011; 53: 808–814.

Wong DTH, George K, Wilson J, et al. Effectiveness of serial increases in amino-terminal pro-B-type natriuretic peptide levels to indicate the need for mechanical circulatory support in children with acute decompensated heart failure. *Am J Cardiol* 2011; 107(4): 573–578.

Wood F, Simpson S, Barnes E, et al. Disease trajectories and ACT/RCPCH categories in paediatric palliative care. *Palliat Med* 2010; 24(8): 796–806.

**Studies exploring views and perspectives**

Alam R, Barrera M, D’Agostino N, et al. Bereavement experiences of mothers and fathers over time after the death of a child due to cancer. *Death Stud* 2012; 36(1): 1–22.

Anderzen-Carlsson A, Sorlie V and Kihlgren A. Dealing with fear – from the perspective of adolescent girls with cancer. *Death Stud* 2012; 36(1): 286–292.

Baker JN, Windham JA, Hinds PS, et al. Bereaved parents’ intentions and suggestions about research autopsies in children with lethal brain tumors. *J Pediatr* 2013; 163(2): 581–586.

Bally JMG, Duggleby W, Holtslander L, et al. Keeping hope possible: a grounded theory study of the hope experience of parental caregivers who have children in treatment for cancer. *Cancer Nurs* 2014; 37(5): 363–372.

Barling JA, Stevens J and Davis KM. Family members’ retrospective stories of the treatment stage of an adolescent or young adult who subsequently died of cancer. *Cancer Nurs* 2013; 36(5): E39–E48.

Bratt EL, Ostman-Smith I, Sparud-Lundin C, et al. Parents’ experiences of having an asymptomatic child diagnosed with hypertrophic cardiomyopathy through family screening. *Cardiol Young* 2011; 21(1): 8–14.

Bratt EL, Sparud-Lundin C, Ostman-Smith I, et al. The experience of being diagnosed with hypertrophic cardiomyopathy through family screening in childhood and adolescence. *Cardiol Young* 2012; 22(5): 528–535.

Brennan C, Hugh-Jones S and Aldridge J. Paediatric life-limiting conditions: coping and adjustment in siblings. *J Health Psychol* 2013; 18(6): 813–824.

Cataudella DA and Zelcer S. Psychological experiences of children with brain tumors at end of life: parental perspectives. *J Palliat Med* 2012; 15(11): 1191–1197.

Cote-Arsenault D and Denney-Koelsch E. ‘My baby is a person’: parents’ experiences with life-threatening fetal diagnosis. *J Palliat Med* 2011; 14: 1302–1308.

Dussel V, Joffie S, Hilden JM, et al. Considerations about hastening death among parents of children who die of cancer. *Arch Pediatr Adolesc Med* 2010; 164(3): 231–237.

El Mallia H, Kreieberg U, Steineck G, et al. Parental trust in health care – a prospective study from the Children’s Cancer Hospital in Egypt. *Psychooncology* 2013; 22(3): 548–554.

Foster TL, Gilmer MJ, Davies B, et al. Comparison of continuing bonds reported by parents and siblings after a child’s death from cancer. *Death Stud* 2011; 35(5): 420–440.

Gaab EM, Owens RG and MacLeod RD. Primary caregivers’ experiences living with children involved in pediatric palliative care in New Zealand. *Vulnerable Child Youth Stud* 2013; 8(1): 1–9.

Gaab EM, Owens RG and MacLeod RD. The voices of young New Zealanders involved in pediatric palliative care. *J Palliat Care* 2013; 29: 186–192.

Garvie PA, He J, Wang J, et al. An exploratory survey of end-of-life attitudes, beliefs, and experiences of adolescents with HIV/AIDS and their families. *J Pain Symptom Manage* 2012; 44(3): 373–385 e29.

Gerhardt CA, Fairclough DL, Grossenbacher JC, et al. Peer relationships of bereaved siblings and comparison classmates after a child’s death from cancer. *J Pediatr Psychol* 2012; 37(2): 209–219.

Granek L, Barrera M, Shaheed J, et al. Trajectory of parental hope when a child has difficult-to-treat cancer: a prospective qualitative study. *Psychooncology* 2013; 22(11): 2436–2444.

Guo J, Wilford BS, Farlow B, et al. Our children are not a diagnosis: the experience of parents who continue their pregnancy after a prenatal diagnosis of trisomy 13 or 18. *Am J Med Genet A* 2014; 164(2): 308–318.

Hexem KR, Mollen CJ, Carroll K, et al. How parents of children receiving pediatric palliative care use religion, spirituality, or life philosophy in tough times. *J Palliat Med* 2011; 14: 39–44.

Hinds PS, Oakes LL, Hicks J, et al. ‘Trying to Be a Good Parent’ As defined by interviews with parents who made phase I, terminal care, and resuscitation decisions for their children. *J Clin Oncol* 2009; 27(35): 5979–5985.

Hinojosa MS, Knapp CA, Madden VL, et al. Caring for children with life-threatening illnesses: impact on white, African American, and Latino families. *J Pediatr Nurs* 2012; 27(5): 500–507.

Hogwood J, Campbell T and Butler S. I wish I could tell you but I can’t: adolescents with perinatally acquired HIV and their dilemmas around self-disclosure. *Clin Child Psychol Psychiatry* 2013; 18(1): 44–60.

Hoven E, von Essen L and Norberg AL. A longitudinal assessment of work situation, sick leave, and household income of mothers and fathers of children with cancer in Sweden. *Acta Oncol* 2013; 52(6): 1076–1085.

Janvier A, Farlow B and Wilford BS. The experience of families with children with trisomy 13 and 18 in social networks. *Pediatrics* 2012; 130(2): 293–298.
Kars MC, Grypdonck MH, Beishuizen A, et al. Factors influencing parental readiness to let their child with cancer die. *Pediatr Blood Cancer* 2010; 54(7): 1000–1008.

Lagrange RD, Mitchell SJ, Lewis M, et al. Health protective behaviors among young people living with HIV/AIDS. *J AIDS Clin Res* 2012; 13: 7348.

Lannen P, Wolfe J, Mack J, et al. Absorbing information about a child’s incurable cancer. *Oncology* 2010; 78(3–4): 259–266.

Larsen HB, Heilmann C, Johansen C, et al. An analysis of parental roles during hematopoietic stem cell transplantation of their offspring: a qualitative and participant observational study. *J Adv Nurs* 2011; 67: 1458–1467.

Lathrop A and Vandeusse L. Affirming motherhood: validation and invalidation in women’s perinatal hospice narratives. *Birth* 2011; 38(3): 256–265.

Lazarus R, Struthers H and Violiari A. Starting HIV-positive babies on antiretroviral treatment: perspectives of mothers in Soweto, South Africa. *J Pediatr Health Care* 2010; 24(3): 176–183.

Lindahl Norberg A, Poder U and von Essen L. Early avoidance of disease- and treatment-related distress predicts post-traumatic stress in parents of children with cancer. *Eur J Oncol Nurs* 2011; 15(1): 80–84.

Malcolm C, Gibson F, Adams S, et al. A relational understanding of sibling experiences of children with rare life-limiting conditions: findings from a qualitative study. *J Child Health Care* 2014; 18(3): 230–240.

Matsuoka M and Narama M. Parents’ thoughts and perceptions on hearing that their child has incurable cancer. *J Palliat Med* 2012; 15(3): 340–346.

McCarthy MC, Clarke NE, Ting CL, et al. Prevalence and predictors of parental grief and depression after the death of a child from cancer. *J Palliat Med* 2010; 13(11): 1321–1326.

Medway M, Tong A, Craig JC, et al. Parental perspectives on the financial impact of caring for a child with CKD. *Am J Kidney Dis* 2015; 65: 384–393.

Menezes A. Moments of realization: life-limiting illness in childhood – perspectives of children, young people and families. *Int J Palliat Nurs* 2010; 16(1): 41–47.

Mitchell W and Sloper P. Making choices in my life: listening to the ideas and experiences of young people in the UK who communicate non-verbally. *Child Youth Serv Rev* 2011; 33: 521–527.

Montoya-Juárez R, García-Caro MP, Schmidt-Rio-Valle J, et al. Suffering indicators in terminally ill children from the parental perspective. *J Palliat Med* 2010; 13(6): 720–725.

Naftel RP, Tubergen E, Shannon CN, et al. Parental recognition of shunt failure: a prospective single-institution study. *Clinical article. J Neurosurg Pediatr* 2012; 9: 363–371.

Nasr AS and Rehm RS. Parental live liver donation: a transformational experience. *Prog Transplant* 2014; 24(1): 69–75.

Paisley MA, Kang TI, Insogna IG, et al. Complementary and alternative therapy use in pediatric oncology Patients with failure of frontline chemotherapy. *Pediatr Blood Cancer* 2011; 56(7): 1088–1091.

Pouset G, Bilsen J, De Wilde J, et al. Attitudes of adolescent cancer survivors toward end-of-life decisions for minors. *Pediatrics* 2009; 124(6): E1142–E1148.

Price J, Jordan J, Prior L, et al. Comparing the needs of families of children dying from malignant and non-malignant disease: an in-depth qualitative study. *BMJ Support Palliat Care* 2012; 2(2): 127–132.

Pritchard M, Srivastava DK, Okuma JO, et al. Bereaved parents’ perceptions about when their child’s cancer-related death would occur. *J Pain Symptom Manage* 2009; 38(4): 561–567.

Rahman B, Macciocca I, Sahhar M, et al. Adolescents with implantable cardioverter defibrillators: a patient and parent perspective. *Pacing Clin Electrophysiol* 2012; 35(1): 62–72.

Rapaport A, Shaheed J, Newman C, et al. Parental perceptions of forgoing artificial nutrition and hydration during end-of-life care. *Pediatrics* 2013; 131(5): 861–869.

Ravindran VP and Rempel GR. Grandparents and siblings of children with congenital heart disease. *J Adv Nurs* 2011; 67(1): 169–175.

Robert R, Zhukovsky DS, Mauricio R, et al. Bereaved parents’ perspectives on pediatric palliative care. *J Soc Work End Life Palliat Care* 2012; 8(4): 316–338.

Seth A, Gupta R, Chandra J, et al. Adherence to antiretroviral therapy and its determinants in children with HIV infection – experience from Paediatric Centre of Excellence in HIV Care in North India. *AIDS Care* 2014; 26: 865–871.

Tan JS, Docherty SL, Barfield R, et al. Addressing parental bereavement support needs at the end of life for infants with complex chronic conditions. *J Palliat Med* 2012; 15(5): 579–584.

Tomlinson D, Hesser T, Ether MC, et al. Complementary and alternative medicine use in pediatric cancer reported during palliative phase of disease. *Support Care Cancer* 2011; 19(11): 1857–1863.

Vles GF, Soudant DL, Hoving MA, et al. Long-term follow-up on continuous intrathecal Bacofoil therapy in non-ambulant children with intractable spastic Cerebral Palsy. *Eur J Paediatr Neurol* 2013; 17(6): 639–644.

Von Lutzau P, Otto M, Hechler T, et al. Children dying from cancer: parents’ perspectives on symptoms, quality of life, characteristics of death, and end-of-life decisions. *J Palliat Care* 2012; 28(4): 274–281.

Wells F, Ritchie D and McPherson AC. ‘It is life threatening but I don’t mind’. A qualitative study using photo elicitation interviews to explore adolescents’ experiences of renal replacement therapies. *Child Care Health Dev* 2013; 39: 602–612.

Yoshida S, Amano K, Ohta H, et al. A comprehensive study of the distressing experiences and support needs of parents of children with intractable cancer. *Jpn J Clin Oncol* 2014; 44: 1181–1188.

Yuen WY, Duipmans JC and Jonkman MF. The needs of parents with children suffering from lethal epidermolysis bullosa. *Br J Dermatol* 2012; 167: 613–618.

Zahmacioglu O, Yildiz CE, Koca B, et al. Coming from behind to win – a qualitative research about psychological conditions of adolescents who have undergone open-heart surgery for their offspring: a qualitative and participant observational study. *J Cardiothorac Surg* 2013; 6: 1181–1188.
lymphoblastic leukemia (ALL). *Pediatr Blood Cancer* 2011; 57(7): 1204–1209.

**Studies evaluating current practice**

Akard TF, Gilmer MJ, Miller K, et al. Factors affecting recruitment and participation of bereaved parents and siblings in grief research. *Prog Palliat Care* 2014; 22(2): 75–79.

Akard TF, Wray S and Gilmer MJ. Facebook advertisements recruit parents of children with cancer for an online survey of web-based research preferences. *Cancer Nurs* 2015; 38: 155–161.

Baker JN, Leek AC, Salas HS, et al. Suggestions from adolescents, young adults, and parents for improving informed consent in phase I pediatric oncology trials. *Cancer* 2013; 119(23): 4154–4161.

Beslow LA, Ichord RN, Gindville MC, et al. Pediatric intracerebral hemorrhage score: a simple grading scale for intracerebral hemorrhage in children. *Stroke* 2014; 45(1): 66–70.

Bingen K, Kupst MJ and Himelstein B. Development of the palliative care parental self-efficacy measure. *J Palliat Med* 2011; 14(9): 1009–1016.

Bona K, Bates J and Wolfe J. Massachusetts’ Pediatric Palliative Care Network: successful implementation of a novel state-funded pediatric palliative care program. *J Palliat Med* 2011; 14: 1217–1223.

Caretti V, Jansen MH, van Vuurden DG, et al. Implementation of a multi-institutional diffuse intrinsic pontine glioma autopsy protocol and characterization of a primary cell culture. *Neuropathol Appl Neurobiol* 2013; 39: 426–436.

Darbyshire P, Cleghorn A, Downes M, et al. Supporting bereaved parents: a phenomenological study of a telephone intervention programme in a paediatric oncology unit. *J Clin Nurs* 2013; 22(3–4): 540–549.

Davies B, Contro N, Larson J, et al. Culturally-sensitive information-sharing in pediatric palliative care. *Pediatrics* 2010; 125(4): E859–E865.

Dickinson HO, Rapp M, Arnaud C, et al. Predictors of drop-out in a multi-centre longitudinal study of participation and quality of life of children with cerebral palsy. *BMC Res Notes* 2012; 5: 300.

Eilegard A, Steineck G, Nyberg T, et al. Bereaved siblings’ perception of participating in research – a nationwide study. *Psychooncology* 2013; 22(2): 411–416.

Gilmer MJ, Foster TL, Bell CJ, et al. Parental perceptions of care of children at end of life. *Am J Hosp Palliat Care* 2013; 30(1): 53–58.

Groh G, Borasio GD, Nickolay C, et al. Specialized pediatric palliative home care: a prospective evaluation. *J Palliat Med* 2013; 16: 1588–1594.

Lyon ME, Jacobs S, Briggs L, et al. Family-centered advance care planning for teens with cancer. *JAMA Pediatr* 2013; 167(5): 460–467.

Miller KS, Vannatta K, Vasey M, et al. Health literacy variables related to parents’ understanding of their child’s cancer prognosis. *Pediatr Blood Cancer* 2012; 59(5): 914–918.

Miller VA, Baker JN, Leek AC, et al. Patient involvement in informed consent for pediatric phase I cancer research. *J Pediatr Hematol Oncol* 2014.

Morley TE, Cataudella D, Fernandez CV, et al. Development of the Pediatric Advanced Care Quality of Life Scale (PAC-QoL): evaluating comprehension of items and response options. *Pediatr Blood Cancer* 2014; 61(10): 1835–1839.

Olcese ME and Mack JW. Research participation experiences of parents of children with cancer who were asked about their child’s prognosis. *J Palliat Med* 2012; 15(3): 269–273.

Rosenberg AR, Dussel V, Orellana L, et al. What’s missing in missing data? Omissions in survey responses among parents of children with advanced cancer. *J Palliat Med* 2014; 17: 953–956.

Steele R, Cadell S, Siden H, et al. Impact of research participation on parents of seriously Ill children. *J Palliat Med* 2014; 17(7): 788–796.

Stevens MM, Lord BA, Proctor M-T, et al. Research with vulnerable families caring for children with life-limiting conditions. *Qual Health Res* 2010; 20(4): 496–505.

Van der Geest IMM, Darlington ASE, Streng IC, et al. Parents’ experiences of pediatric palliative care and the impact on long-term parental grief. *J Pain Symptom Manage* 2014; 47(6): 1043–1053.

Walsh KE, Roblin DW, Weingart SN, et al. Medication errors in the home: a multisite study of children with cancer. *Pediatrics* 2013; 131(5): E1405–E1414.

Wiener L, Sweeney C, Baird K, et al. What do parents want to know when considering autopsy for their child with cancer? *J Pediatr Hematol Oncol* 2014; 36: 464–470.

Wolf J, Robert R, Sommerer A, et al. Impact of a pediatric palliative care program. *Pediatr Blood Cancer* 2010; 54: 279–283.