Study Protocol

Full Title:

End of Life Care for Infants, Children and Young People: a mixed methods evaluation of current practice in England and Wales

Short title:

End of Life Care for Infants, Children and Young People

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1.1	Lorna Fraser	27/11/2020	Structured interviews with clinical leads in WS1 added	

Summary of Research (abstract)

Research question

Do outcomes and experiences for infants, children and their families, and resources required, vary depending on the model of End of Life (EoL) care that they receive?

Background

4500 children die in England and Wales every year and although there are increasing numbers of paediatric palliative care and hospice services, these services vary in their professional configuration, services provided, funding sources and population served. There is little evidence on the models of care, quality of care, resourcing and outcomes of children and their families who use these services. Most of the recommendations in the NICE guidance for EoL care for children are based on low quality evidence.

Aims and objectives

Aim: To identify and investigate different models of providing EoL care for infants, children and young people, in terms of outcomes and experiences for children and parents, resource use and costs to families and the NHS.

Objectives:

1. To identify and describe current models of delivering EoL care to infants, children and young people in England and Wales
2. Identify barriers and facilitators to implementation of these EoL models
3. To assess inequalities in access or availability of these models of care
4. To explore whether the experiences and outcomes for child, parent or family vary dependent on the model of EoL care received
5. To compare the resource implications of the different models of EoL care for the NHS and families

Methods

This mixed methods study of three linked workstreams and cross-cutting health economics theme will examine three exemplar clinical settings which care for more than 50% of children that die each year: cancer services, paediatric intensive care units (PICU) and neonatal units (NNU).

WS1: A questionnaire survey of Service Leads, and structured interviews with Chairs of regional paediatric palliative care networks, will systematically capture data relevant to current practice to providing EoL care, develop a typology of models of EoL care and associated resource use and costs. These models will inform the sample selection in WS2 & 3 and the analyses in WS3.

WS2: A qualitative study will explore how these models are implemented and experienced, and identify the important outcomes and resources associated with EoL care. Focus groups with healthcare professionals will explore experiences of meeting EoL care needs and views on the factors affecting access to EoL care including identification of any inequalities and barriers in access. Interviews with bereaved parents will explore experiences of their child's treatment and care towards the end of their life and associated costs to them.

WS3: We will use routinely collected data to compare EoL outcomes (e.g. high intensity treatments, place of death) in children with cancer who have died and had different EoL models of care. Then prospective data collection in PICU and NNUs will collect data on 800 dying children including other outcomes (e.g. choices given to parents, symptoms). Data collection from the parents after death will provide additional information. We will assess whether outcomes vary according to the different models of EoL care and assess the associated resource use and costs.

Timelines for delivery

This 4 year study includes ongoing dissemination to key audiences (parents, service providers, commissioners) via knowledge exchange events, web-based platforms, social media and clinical/academic forums.

Anticipated impact and dissemination

The results will inform service delivery in order to utilise finite resources to maximise impact.

Background and Rationale

Although child mortality has decreased over the last few decades, around 4500 infants and children (0- 19 years) die in England and Wales every year [1, 2] and there has been a recent rise in infant mortality [3]. Approximately half of these deaths are from underlying life-limiting conditions and are therefore 'expected' [4].

Over the last 30 years, there are growing numbers of paediatric palliative care and hospice services in the UK that provide EoL care for children, but there is little evidence on the models of care, quality, resource implications and outcomes of children and families who use these services. We know that these services vary in their professional configuration, services provided, funding sources and population served [5]. Palliative care services for children and young people in the UK have developed locally with heavy reliance on individual clinician and third sector organisations e.g. children's hospices [6]. As a result, delivery of palliative care for children is 'inconsistent and incoherent' [7].

The recent NICE guideline (NG61) on the EoL care for infants, children and young people [8, 9] includes a comprehensive list of 143 recommendations. However, the quality of evidence on which most of these recommendations is made is low or very low. Furthermore, the NICE Guideline found only one relevant study in a review of health economics evidence [7], highlighting the poor current level of understanding about the costs of patient wellbeing and implications of current care.

Our systematic review of specialist paediatric palliative care for children with cancer[10] found that children who receive specialist input are cared for differently, with evidence of more advance care planning and less intensive care at the end of life. However, the conclusions which can be drawn are limited given the poor quality of the evidence, and reliance on North American studies which are not necessarily transferable to the UK context. In addition, few studies investigated families' views or assessed how inequalities in access may influence EoL care. A recent review of quality indicators to assess the impact of paediatric palliative care highlighted the breadth of indicators used, lack of consensus and limited input from the children's perspectives [11]. The need for more research on EoL care has been identified in research prioritization exercises [12, 13].

Previous research [7, 14, 15], including the NICE Guideline [8] has emphasised the challenges of conducting economic evaluation in EoL care, when conventional health maximisation is no longer the aim of the intervention. Increasingly economic evaluations of health care interventions are used to inform decisions on how best to allocate limited resources for optimal health gain. In EoL care a comprehensive view of the costs and benefits which are relevant to the decision problem extend beyond health to encompass broader cross-sector impacts spanning the statutory and non-statutory sector, as well as the private sphere with impact on the patient and their network of family and friends. In addition, dimensions of care beyond health are important since patient care is no longer primarily curative, nor with any likely extension in time lived. This makes economic evaluation of palliative care non-standard [7, 8, 14, 16, 17]. In practice, there has been almost no economic evaluation of EoL care in children [7, 8, 18]. Failure to consider the costs or benefits of the range of end of life care packages has contributed to the inconsistent and variable provision of care throughout the NHS, and internationally. Furthermore, at a time of extensive budgetary pressures the inability to define the benefits of a healthcare budget or argue for the value of additional funding puts the delivery of end of life care on the back foot, with increasing reliance on third sector support - a sector which itself is under substantial pressure [19].

Why is this research needed now?

Children's palliative care is a priority in the recent NHS long term plan which also includes a commitment to increase funding to children's hospices, one component of EoL care for children [20]. However, the recent NICE guidelines [8] noted there is little evidence on which to base service delivery and made recommendations for future research e.g. effectiveness of a home-based package of care compared to hospital or hospice care.

The evidence gap is clear from the low quality evidence informing the NICE guidance [8]. A search of the NIHR HS&DR portfolio identified no published or ongoing research projects on EoL care in people under 20, though findings from an evaluation of hospice at home models (HS&DR - 14/197/44) and a feasibility trial of managing clinical uncertainty (HTA - 15/10/17) could help inform our study. This also builds on existing work from the study team [10].

This application fits under three of the categories listed in the Commissioned call for EoL care: End of Life care for infants, children and young people; reducing health inequalities in EoL care; and the time and place of access to services.

The results from the study will be based on services which care for >50% of babies, children and young people who die in England and Wales. These results should be transferrable to all other paediatric services. NHS commissioners and policy makers require high quality research in order to develop their services and this research will inform robust guidance on further development of EoL care for this population.

This study will identify and investigate the different models of providing EoL for infants, children and young people with an aim to understand the impact of the models of care on the EoL experiences for this population, the costs for whom and where.

Aims and Objectives

Aim: This study will identify and compare different models of providing EoL care for infants, children and young people, in terms of outcomes and experiences for children and parents, resource use and costs to families.

Objectives:

1. To identify and describe current models of delivering EoL care to infants, children and young people (0-18 years) in England and Wales.
2. Identify barriers and facilitators to implementation of these EoL models.
3. To assess inequalities in access or availability of these services.
4. To explore whether the outcomes and experiences for child, young person, parent or family vary dependent on the model of EoL care received.
5. To compare the resource implications of the different models of EoL care for the NHS and families.

Research Plan/Methods (including costs of each stage)

This mixed methods study consists of three linked workstreams (WS) with a cross-cutting health economics theme (see flowchart). Our focus are three clinical settings which together care for approximately 50% of the children who die in England and Wales each year.

1. Children's Cancer Services (~350 deaths per year)[21]
2. Paediatric Intensive Care Units (PICU) (~700 deaths per year)[22]
3. Neonatal Units (NNU) (i.e. Special Baby Care Units (SCBU), Local Neonatal Units (LNU), Neonatal Intensive Care Units (NICU)) (~1100 deaths per year)

Cross-cutting health economics theme (£165,061)

We will embed health economics within all elements of this research to ensure that there can be genuine progress in the ability of researchers, decision makers, the children and their families to contribute to an

understanding of how we can ensure the limited funding for EoL care can be used for greatest benefit for the children at the end of their lives. The details of how the theme will interact with each of the 3 core workstreams are detailed in each section below.

In addition to the stated contributions to each WS we will conduct an exploratory analysis of how the latest methodological research in health economics can be used to address some of the challenges faced in this area. This will primarily concern the incorporation of inequality of care considerations into economic evaluation [23, 24], and the comparison of costs that fall across different stakeholders [25].

As well as contributing to the final HS&DR report the health economists will contribute to all of the planned knowledge exchange activities to ensure the cross-cutting theme is responsive to the reality faced by decision makers and patients. We will also ensure that the research outcomes are relevant to the future updates of the NICE Guidance, especially the resource impact toolkit.

WS1: Identifying and describing models of service delivery for neonatal and paediatric end of life care (objective 1,2,3 & 5)(£209,060):

WS1 will describe current models of delivery of end of life care in children's and teenage & young adult (TYA) Principal Treatment Centres (PTCs), s paediatric intensive care units, and neonatal units in England and Wales. It will also provide a description of the resources and costs associated with each model. Core distinguishing characteristics of delivery models will be nature of involvement (or not) of:

- specialist paediatric palliative care clinicians (e.g. specialist trained consultants within clinical team vs separate, consultant-led paediatric palliative care team),
- community paediatric palliative care services (e.g. children's hospice, children's community nursing team),
- other professions identified by research and current guidance as being key elements of paediatric palliative care (e.g psychological services, bereavement support, social work, chaplaincy).

In addition, it will investigate factors (e.g. diagnostic group, hospital type (e.g. children's vs general); geographical location; other specialisms within the setting; the remit/operation of the local Managed Clinical Network) associated with the delivery model implemented. It will also describe current inadequacies or limitations in EoL care provision, both in terms of equity of access to services with paediatric palliative care involvement and extent to which a holistic approach to EoL care is being achieved.

Design: A survey will collected from each centre/unit. A structured interview will collect regional data on provision relevant to end of life care of babies, children and young people.

Sampling:

- Survey: Clinical leads of all child (n=16) and TYA (n=14) cancer PTC), PICUs n=25, and NNUs; n=182 in England and Wales
- Structured interview: Chairs of regional paediatric palliative care networks (n=13).

The research instrument:

The survey:

A questionnaire designed specifically for this study will systematically capture data on the organization and delivery of EoL care to infants, children and young people in PCTs, PICUs and NNUs . Survey content will be informed by: existing evidence on service-related factors/characteristics associated with end of life outcomes of children and parents, [10, 26, 27] the NICE guideline [8] (and associated quality standards [28]) for EoL

care of infants, children and young people, and in consultation with the study's Parent Advisory Panel and the Study Steering Committee. Data collected will include:

- organizational context (e.g. type of hospital, other medical specialisms within hospital)
- annual 'caseload' and number of deaths/year
- settings in which they are responsible for providing (i.e. coordinate, oversee or direct care) end of life care (e.g. inpatient, home, hospice)
- medical and other health/allied health specialisms represented in core clinical teams (in terms of full-time equivalent for each specialism).
- number and type of staff within core clinical team holding qualifications in paediatric palliative care, including date and nature of qualification
- other medical and healthcare specialisms (e.g. paediatric palliative care, clinical psychology, physiotherapy) not part of core clinical team but contracted to work in service, including role, level of expertise, full-time equivalents for each specialism, nature of that access (e.g. working hours vs 24/7, commissioning arrangements).
- end-of-life care delivery partners, both for joint/shared delivery of care (e.g. children's community nursing team, paediatric palliative care team, ambulance Trust, children's hospice, GP, Local Authority, local clinical network) and onward transfers (e.g. LNU to NICU; PICU to hospice).
- nature of each of above partnership (e.g. ad hoc vs specified in care pathways/protocols; commissioning arrangements) and clinical situations in which such partnerships are operationalized,
- nature of out of hours access to consultant-level (and other relevant) expertise available to clinical team, delivery partners and families
- existence of within-service policies/protocols regarding: advanced/end of life care planning, rapid transfers of care, instigation of palliative/end of life care, out of hours cover.
- key details of each protocol (e.g. lead clinician, core team members involved, other clinicians/practitioners involved, clinical 'triggers', notification-/information sharing arrangements)
- senior clinician's membership of local/regional children's palliative care network
- commissioning arrangements
- where relevant, approximate dates/timings of implementing aspects of end of life care will also be collected.

We will take care to achieve the correct balance between the "granulatory" of data collection and response burden/feasibility. Fixed response questions will be used where possible and early piloting work with specific questions or sections of the survey will utilise those members of the research team working in or representing target services. The final draft survey will be piloted with a clinical lead from each type of service to be surveyed. Cognitive interviewing techniques will be used to evaluate content, feasibility, respondent burden, and wording of question and response options. Where appropriate this will be an iterative process with revisions being made and evaluated in subsequent pilot interviews.

The survey will be created in and administered using Qualtrics survey software platform, this will facilitate use of routing to minimize respondent burden and support engagement. The survey will be set up to maximize accuracy of information provided (e.g. pre-specifying minimum and maximum values for requested numerical data).

<u>The structured interviews:</u>
<u>Interviewees will be asked to describe: all services involved in providing paediatric palliative care provision in their region (statutory and third sector); any regional or local service provision models, region-wide services, protocols, posts and funding; views on gaps in pathways/provision (for whole or sub-populations) within the region.</u>

Data collection: Desk-based research will identify clinical leads of PTCs, PICUs and NNUs, and chairs of regional paediatric palliative care networks.

Survey:

Around a month prior to distribution, clinical leads will receive a brief introductory email outlining the study and its objectives and notifying them about the survey. This will also serve to identify any changes to staff or anomalies/errors in contact details. Our clinical co-apps will champion this study with their professional organisations (see letters of support from PIC-SG and CCLG). Clinical leads will receive the invitation to take part in the survey via email, with the link to the survey embedded in the email. The Study Information Sheet will be attached. The first section of the survey will comprise the consent form. Respondents will be given a reasonable time period to complete the questionnaire with reminders (via email and, if required, telephone) used to maximize response rate. Where service leads are unable to provide information regarding service costs, we will contact relevant finance departments directly.

Structured interviews:

These will be administered via a telephone call. Invitations to take part in an interview will be distributed via email, with Study Information Sheet attached. They will audio-recorded, with consent will be audio-recorded at the start of the interview. After the interview, the recording will be used to populate a series of Excel spreadsheets, structured according to a priori topic areas.

Data Analyses:

We will: i) develop a typology of current approaches/service models of delivering EoL care, and the service characteristics/features of practice which distinguish (or are shared across) models; ii) provide a descriptive account of these models and occurrence/distribution within the three service types (cancer services, PICU, NNU) and geography; and iii) provide information on the resources required to deliver each model.

Univariate descriptive statistics will be used to describe categorical and numerical data including resources and costs. Based on preparatory work and existing published evidence, we anticipate up to six EoL care delivery models will be identified but are open to there being more or less. Within each model, services will vary in the degree to which they fully adhere to the model. In addition, it is likely that some services will report no or only very limited involvement of these professionals/services and therefore do not align to any of the identified models.

Cross-tabulation will be used to describe and compare delivery models with respect to organisational, patient and contextual characteristics, and to describe factors affecting access to end of life care adhering to these delivery characteristics. Descriptive accounts of provision in each region, and commentaries of differences between regions, and inequities of access within and between regions, will be derived from the Excel spreadsheets containing data from interviews with regional chairs.

We will explore whether provision of these services implies a cost burden on other public and third sector stakeholders. This will contribute to the planned summaries of the cost of the different types of service in addition to informing the structure of subsequent resource use explorations in WS2 and WS3.

An interim report of analysis of survey data will be discussed with the SSC, with this informing final definitions of the alternative models of providing end of life care, the final stages of analysis, categorisation of services to each delivery model, and selection of research sites for WS2.

Draft typologies will be created for each model, and drawing on the findings of WS1 and existing literature [11, 26, 27, 29-32] we will develop a logic model which represents how these models work (i.e. how the combined inputs and resources are expected to produce a set of outcomes such as improved end of life care). These draft outputs will be presented to family members, clinicians and other relevant stakeholders for discussion at the

first of three knowledge exchange workshops, with expert input provided at the workshop used to refine the typologies and logic model(s) for further exploration and refinement in WS2 and WS3.

Outputs:

- A description of the current situation regarding the provision of EoL care for children with cancer and those being cared for in PICUs, and NNUs. This will include identifying elements of EoL care most likely to be under-developed and whether diagnosis, care setting and geographical factors are associated with limitations in end-of-life care, or better developed provision.
- A typology of models of delivering EoL care, including a description of key service/delivery characteristics which distinguish delivery models and any commonalities in characteristics across two or more models.
- A draft logic model for end-of-life care for children
- Data collected and the service delivery typology will be used for the sampling for WS2 and 3.
- As well as text-based reports and summaries, infographic representations of the typology of models by sector (NNU, PICU, oncology) and their geographical distribution will be created.

These findings will be presented as part of the first knowledge exchange event in Month 13 and form an academic publication.

WS 2: Qualitative evaluation and exploration of the delivery, experience and impacts of different models of End of Life Care provision (objectives 1-5) (£280,455):

In this workstream, we will conduct a qualitative process evaluation [33, 34] to learn about how the models of care identified in WS1 are implemented in routine practice and experienced by those providing and receiving EoL care, and identify potential mechanisms of impact, outcomes and resources associated with EoL care to investigate in WS3. Focus groups with healthcare professionals will explore experiences of meeting EoL care needs and factors affecting access to EoL care including identification of any inequalities in access. Individual interviews with bereaved parents will explore their experiences of their child's treatment and care towards the end of their life, and perceived benefits and costs to them. This WS draws on MRC guidance on evaluating complex interventions[33, 35], which highlights the importance of exploring both the institutional and service-level structures that underpin models of care (as identified in WS1) but also the ways in which actors, in this case healthcare staff and families, interact with and respond to those structures, which is the focus of WS2.

Setting: Children's Cancer Services, Paediatric Intensive Care Units and Neonatal Units in English and Welsh NHS hospitals, purposively sampled to have representation of those services which best fit each of the models identified in WS1. For example, in the case that 6 models are identified in WS1, approx. 3 services per model will be sought for WS2 (total approx. 18), sampling by best exemplars of each model, size, geography and distance from key EoL care providers within the model.

Interviews with bereaved parents.

Sample: we will purposively recruit parents of children who have died to ensure a mix of families who have received different EoL care models (identified in WS1) and relevant clinical and demographic characteristics (e.g. child age, underlying diagnosis, expected / unexpected death, place of death, family composition, ethnicity, socioeconomic status). This diversity will be achieved using two strategies; 1) screening of children's records in advance of recruitment, and 2) monitoring of data collected to focus later recruitment on particular characteristics not reflected in the sample. To ensure we capture the range and diversity of experience among families and also explore how experience may differ by EoL care model, we aim to interview a minimum of 7parents per EoL care delivery model (e.g. where 6 models are identified, total n≈42 parents). Final sample size will be determined by the number of models identified in WS1 and through monitoring data saturation (i.e. we will stop recruiting when no new themes are emerging from parent interviews). We will not approach parents who are recently bereaved (within the first 3 months of their child's death) or longer than 3 years since the death of their child. This decision has been made with reference to

relevant research [36, 37], our own experience of conducting research with bereaved parents, and input from our parent advisors.

Recruitment: Clinical teams will identify eligible parents (mothers or fathers), and a member of the clinical team in the child's service will make first contact (by postal invitation and follow-up phone call). Interested parents will be asked to complete an expression of interest form and return it to the research team, who will contact them directly to explain the study and arrange an interview. Following input from bereaved parents we will also advertise the study via social media, and use posters and leaflets where appropriate (e.g. in packs sent to bereaved parents from their clinical service, displayed at bereavement sessions attended by parents).

Data collection: In-depth qualitative interviews using a narrative approach followed by semi-structured questions will be used [38, 39], This has the benefit of allowing parents to share their experiences without imposing a structure or order in the first part of the interview but ensuring that key topics are consistently covered with all participants. Following the advice of bereaved parent advisors on a study about the early days after the death of their child [40], parents will be offered the choice of a face-to-face or telephone interview, both of which have been found to offer potential to collect in-depth data on sensitive topics [41]. Face-to-face interviews will be conducted in families' homes or another suitable venue of their choosing (e.g.the University or participating NHS Trust). Written informed consent will be taken for face-to-face interviews, and audio-recorded consent (using the statements on the approved consent form) for telephone interviews.

Interviews will be based around a topic guide, informed by WS1 findings, consultation with key stakeholders and existing research that has explored end of life care provision with families[31, 42-47], and piloted with at least two parent advisors. After introductory questions, the narrative section of the interview will involve inviting parents to tell their story of their child's end of life and period immediately following. The second substantive section of the interview will use in-depth, semi-structured questions to explore the following topics: experiences of their child's treatment and care towards the end of their life, desires (including changes in desires) for EoL fulfilled, the role of different services involved in their child's care and the perceived impacts of these experiences for their child and wider family, identification of unmet needs for care towards the end of their child's life, and the demands and impacts on household finances during end of life and subsequently (e.g. lost working hours, lost employment, out of pocket expenditures associated with/during the end of life phase and funeral costs, and statutory benefits received or removed during this period. Interviews will be designed to last 60 minutes, to allow sufficient exploration of parental experience whilst still covering key topics and minimising participant burden. However, because of the narrative approach it is expected that some interviews may take longer. Interviews will be conducted by an appropriately qualified and experienced researchers under the supervision of the experienced workstream lead.

Focus groups with professionals

Sample: We will aim to undertake a minimum of 3 staff focus groups per EoL care model, which will allow us to explore differences between and within models in terms of factors affecting access and uptake, and service provision. Each focus group will include 6-9 staff working in a cancer service (n=2-3), PICU (n=2-3) or NICU (n=2-3) (e.g. where 6 models are identified, total staff n≈108-162). Each focus group will include staff in different roles (e.g. physicians, nurses, allied health professionals). The final sampling strategy will be informed by the number and configuration of models identified in WS1.

Recruitment: The local Principal Investigator at each site will identify suitable staff participants, circulate recruitment materials and coordinate arrangements for a focus group. Interested staff will be asked to contact the research team directly, who will liaise with them to organise a focus group that everyone can attend.

Data collection: Focus groups will last approximately 60-80 minutes and located on Trust premises to facilitate attendance. A focus group schedule, informed by WS1 findings, stakeholder consultation and existing research[48], will be used to structure discussions, and provide an opportunity for all staff to contribute individually but also to generate discussion amongst participants. Likely topics are their experiences of providing EoL care and involving other EoL care providers, the perceived advantages and challenges to

working within their particular model of care provision, and views on solutions to these challenges or ways care could be improved. The focus groups will also explore perceived impacts of EoL care for children and families, and include a discussion of the proportion of staff time spend on the delivery of EoL care, the role of staff in its delivery, and how this fits within the broader package of care. We will also explore their views on workforce configuration (types of staff involved in provision of the care, their training requirements and the cost of this, and time spent on this activity) to provide the service, including potential inputs beyond the health care sector. These data will inform the exploration of outcomes and resource use implications in WS3.
Focus groups will be facilitated by the WS lead and an appropriately qualified researcher with informed consent taken at the start.
Interviews and focus groups will be audio-recorded using an encrypted digital audio recorder and transcribed (intelligent verbatim) for analysis by an external transcription company with experience of transcribing data collected for health research (and who is GDPR compliant).
Researchers involved in data collection will also keep field notes throughout the data collection process, commenting on important non-verbal data and interesting observations to either follow-up in subsequent focus groups, or to explore during the analysis process.
Data Analyses:
Interview and focus group data will be analysed using the process of thematic analysis, the purpose of which is to draw out key themes that "capture something important about the data in relation to the research question, and represent some level of patterned response or meaning within the data set"[49]. Where relevant, parent and professionals' data will be analysed together; however, staff and parent data will be compared during the development of themes to identify similarities, differences and, disagreements. The analysis will focus primarily on exploring in more detail how the EoL care models identified in WS1 are implemented in routine practice, identifying barriers and facilitators to providing quality and equitable care for children requiring EoL care, understanding how EoL care impacts on families and the resources associated with caring for their child during the end of life phase (which will inform selection of outcomes and costs to investigate in WS3), and exploring the information needs of families and staff to support decision-making processes about EoL care provision. Other key themes that offer additional insights and contexts that help to explain these findings will also be included.
An appropriately qualified researcher (i.e. someone with qualitative research training and experience, likely to be the researcher conducting fieldwork for WS1 and WS2) will be the primary analyst for this WS (analyst 1), working closely with the WS lead (JT, analyst 2) and parent co-investigator (GW, analyst 3), who will be involved in the analysis of data to provide a different interpretation from their experience as a parent, and receive appropriate training and support for the role. Having three analysts working together will help to ensure rigour, and in particular dependability of findings [50]. Analyst 1 will also keep a reflective journal during the data collection and analysis period to record thoughts and reflections on the process and how it may be shaping the responses of participants and interpretation of data. The wider research team and parent advisory group will be utilised at key points during the analysis to help identify key themes that represent the data, and interpret the meaning of these. This will help to ensure credibility and authenticity of study findings.
The following five steps of thematic analysis will be applied as follows:
1) familiarising with the data: analyst 1 will read and re-read all the transcripts, starting with parent transcripts and then moving on to the staff focus group data to explore how similar they are in content. During this process, the researcher will make notes of interesting concepts and ideas (referred to as 'codes' from herein) that relate to the research objectives. Analyst 2 and 3 will read a proportion of transcripts, (selected by analyst 1 to represent some of the diversity in experience), and also note down commonly occurring codes. Working together, the analysts will discuss the selection, labelling and meaning of codes to inform step 2, and decide whether to generate separate codebooks for parent and staff data.
2) generating initial codes: analyst 1 will continue to generate codes that represent the data and discuss these regularly with analyst 2 and 3 before applying the agreed codes systematically across the dataset, the

purpose of which is to organise the data into meaningful analytical categories. For step 2 the data will be managed and coded in NVivo software.
3) searching for themes: using the coded data, the analysts will work together to identify themes that represent the data and explore relationships between codes and themes, e.g. identifying how groups of codes may be combined to generate a theme, and how these relate to the different models of end of life care. Analytical tools such as mind mapping and brainstorming will be used during this step, with constant reference to the raw coded data to ensure the meaning is retained. Similarities, differences and disagreements between the parent and staff data will also be explored during this step, and a thematic map will be produced to illustrate the links between themes and codes.
4-5) reviewing and defining themes: during these steps the analysts will work with the wider research team and parent advisory group to review and refine the themes and the thematic map. Each final theme will be defined and described using quotations to illustrate meaning and relationships between themes, and findings will be incorporated into the model typologies and logic model developed in WS1, e.g. adding details about implementation, causal mechanisms, outcomes.
WS2 outputs will be presented at a second knowledge exchange workshop for discussion, again with input from those attending used to refine the model typologies and logic model, and plans for WS3.
Outputs:

- A model-based output using the typologies developed in WS1 and integrating WS2 findings to build on further in WS3
- A refined logic model for EoL care for children
- Understanding of factors affecting access to and uptake of end of life care generally to address across models
- The results will inform the prospective data collection in WS3 and be presented as part of the second knowledge exchange event in month 34 and presented in a peer review publication.

WS 3: Quantitative evaluation and exploration of the impacts of different models of End of Life care provision (objective 2-5)(£490,015):
The models of care identified in WS1 will be compared in terms of child and parent outcomes. Retrospective and prospective data collection will be used across the three clinical settings, maximising the use of routinely collected data sources and existing IT platforms where possible:
Part 1: Children in Cancer Principal Treatment Centres:
Using routinely collected data sources we will assess whether the use of high intensity treatments in children who have died from cancer varies depending on the model of EoL care that their service delivered.
Design: Retrospective secondary analysis will exploit the linked population level datasets available for children and TYAs with cancer.
Sampling: All children and TYAs with cancer in England who have died from 2012-2018 (n approx. 2750)
Setting: All NHS cancer treatment centres in England allocated an EoL care models using WS1 outputs.
Data sources: University of Leeds (PICANet) and Public Health England (national cancer registry data (NCRAS), hospital episodes data (inpatient, outpatient and A & E)(HES), Systemic Anti-Cancer Therapy Data set (SACT), Radiotherapy Data Set (RTDS), ONS death certificate data). The data sources held by PHE are already linked on an individual level, The PICANet data will be linked by PHE using deterministic data linkage techniques using name, NHS number, date of birth, sex, postcode and date of death.
Pseudonymised data will be securely transferred to the University of York for data analyses.
Approvals required: NHS REC, CAG, HRA, Public Health England ODR, HQIP (PICANet).
Data Analyses:
Once linkage has been undertaken an assessment of data quality and completeness will be undertaken for all the key clinical and demographic variables of interest (Table 1).
An assessment of missing data will be undertaken once the data are linked and multiple imputation using

chained equations will be used where appropriate [51]. If imputed datasets are used then a sensitivity analyses comparing complete case analyses with the imputed analyses will be undertaken.

Derivation of Key variables:

Some of the key demographic variables will be obtained by combining different data sources e.g. ethnic group, deprivation score. In this situation if any conflict between data sources occurs we will assign the most commonly recorded ethnic group (census 2011 categories) assuming that is not 'unknown'.

Descriptive Statistics:

Appropriate summary statistics, e.g. frequencies and proportions for categorical variables and mean (with standard deviation) or median (with interquartile range) for continuous variables will be produced for all the key variables to describe any variation.

Primary outcome: any one of the following high intensity treatments: intravenous chemotherapy < 14 days from death (yes/no); more than one emergency department visit (yes/no); and more than one hospitalization or intensive care unit admission < 30 days from death (yes/no) [52].

Secondary outcomes: mechanical ventilation < 14 days from death, place of death (hospital, home, hospice).

Analyses will evaluate and compare outcomes used in different EoL care models (identified in WS1) using appropriate regression models. Each analysis will account for the multiple confounding factors in this population (age, underlying diagnoses, comorbidities, outpatient attendance, socioeconomic status (Index of multiple deprivation [53] identified using causal inference methods [54, 55].

The health economic analyses will embed the estimation of the resource use of each package of care into the regression analyses of the retrospective data. The findings of these regressions will be used to inform a full costings analysis by combining with estimates of the unit costs of each resource use element and the findings of WS1 and WS2. This analysis will explore the variation in the cost of the EoL care models through extensive sensitivity and scenario analyses.

Outputs:

These findings will be presented as part of the second knowledge exchange event in month 34 and published in an academic paper.

Table 1 Key Variables and Source Dataset

NCRAS (Primary dataset):	Treatment Data (SACT/RDTS)	Intensive care data (PICANet data)	Hospital admission data (HES)	Outpatient data (HES)	A & E data (HES)	Death registration data (ONS)
Cancer diagnoses Age Sex Date of Diagnoses	Chemotherapy and dates Radiotherapy and dates	Age Sex Ethnicity Deprivation score Planned or unplanned admission Date & time of admission Source of admission Date of discharge Destination on discharge Date of death (if occurred) Primary reason for admission Comorbidities Paediatric Index of Mortality score and variables used to derive this Daily intervention data (e.g. mechanical ventilation, inotropic support, renal replacement therapy)	Age Sex Ethnicity Deprivation score Diagnoses (ICD10 codes) Procedures (OPCS codes) Date of admission Source of admission Specialty of admission Emergency or planned admission Date of discharge Discharge destination Date of death (if occurred)	Age Sex Ethnicity Deprivation score Date of appointment Specialty of appointment	Age Sex Ethnicity Deprivation score Date and time of attendance Diagnoses/reason for attendance Outcome Treatment	Date of death Cause(s) of death Place of death

Part 2: PICU and NNU

Design: prospective longitudinal data collection which will enable us to explore additional individual level outcomes beyond those in Part 1.

Sampling: Units will be purposively sampled to include best exemplars of each model, size, geography and distance from key EoL care providers within the model.

Setting: Paediatric Intensive Care Units and Neonatal Units in England and Wales

Data Collection:

Data will be collected prospectively, using a deferred model of consent whereby the clinical team will record standardised information on outcomes prior to death, with parents being approached after the child dies to consent to the study. Previous studies have shown how difficult it is to obtain consent in the PICU setting [56]. There is also debate over whether true informed consent can be obtained from parents at times of very high levels of anxiety [57] and in the intensive care setting [58, 59]. There is evidence that deferred consent is acceptable to parents in the emergency/PICU setting [60, 61]. The research team would not receive any data until parents had consented to inclusion in this study.

Data will be collected for 1200 children (to yield 800 deaths) who either die in the units or who are transferred home or to hospice prior to death, over an **18 month** period.

a. PICUs: Children will be identified by PICU staff (in approx. 10-12 PICUs) when they are at high risk of death e.g. starting to discuss do not attempt resuscitation (DNA CPR) (we will need to recruit ~600 children to capture ~400 deaths).

b. NNUs: Infants will be identified by NNU staff (in approx. 40-50 NNUs) at the point that they are identified at risk of death e.g. high clinical risk index for babies (CRIB) score [62], severe hypoxic- ischemic encephalopathy [63], extreme prematurity (23/24wks) or starting to discuss DNA CPR (we will need to recruit ~600 babies to capture ~ 400 deaths).

If there are six models of care to explore and compare then with a sample size of 800 we would have 80% power to detect differences of the magnitude of 0.44 (i.e. effect size) on the primary outcome quality of death scale [64]. If there are fewer models of care then smaller differences could be detected whilst retaining 80% power, e.g. 0.34 with 4 models of care.

Utilising current clinical IT platforms (BadgerNet - NNU and PICANet [22] – PICU) we will collect information on quality indicators of care [28] and outcomes up to and including death. These outcomes will be informed by WS1 and WS2 but will likely include symptoms, choices offered to parents about place of care, involvement of SPPC team, place of death, presence of an advanced care plan and bereavement support offered. These data will be collected prospectively by the clinical team with additional data collection from the parents (via postal or telephone questionnaire) approximately 3 -6 months after the child's death. These additional parent reported data will include a quality of death scale [64] to assess EoL care and one of the tools for economic evaluation (ICE-CAP-CPM[65], PICU-QODD-20 [65, 66] or the children's palliative care outcome scale (cPOS)) and to explore the resource use and cost implications beyond secondary care, including primary care, hospice care, and parental out of pocket costs and loss of employment. We will also assess parent outcomes using EQ-5D-5L [67]. We will work with our Parent Advisory Group to determine the most appropriate tool to use to assess EoL care.

Data analyses: Clinical and demographic data of the infants and children who have died and their parents will be summarised in a table using descriptive statistics. Continuous measures will be reported as means and standard deviations or medians and interquartile ranges (as appropriate) and categorical data will be reported as counts and percentages. The flow of participants through the study will be presented in a diagram detailing reasons for withdrawal where data are available. One key outcome of interest will be the quality of death scale to assess end of life care. This will be analysed using multiple linear regression with the quality of death scale as the outcome and model of EOL care as the independent variable of interest adjusting for the multiple confounding factors in this population (age, underlying diagnoses, comorbities).

Model assumptions will be checked and if they are in doubt the data will be transformed prior to analysis or alternative non-parametric analysis methods will be used. The difference between the different models of care in the mean quality of death scale and corresponding 95% confidence interval (CI) will also be presented. Other outcomes of interest will be analysed using an analogous approach as to that outlined above for the quality of death scale.

To determine the cost of the PICU and NNU we will use the same method as for WS3 part 1, where possible, in order to ensure consistency of findings across the centres of care. Through discussion with our Parent advisory panel and the results of WS1 and 2 we will determine which of the outcome measures will be the most informative to decision makers and reflective of child and parent experiences. The chosen outcome will be used to inform a summary of the non-cost benefits associated with each care package for use in a cost-consequence analysis. We will assess whether outcomes for children and families vary according to the different models of EoL care and assess the associated costs. While the development of the available outcome tools is not expected to be sufficient to inform a full cost-effectiveness analysis, their incorporation into research such as this represents an important first step and will be used within a cost-consequence analysis.

Bringing together the findings from WS1, WS2 and WS3, the model typologies and logic model will be refined further and presented at the final knowledge exchange workshop for discussion and to inform development of policy and practice implications. Input will feed into the final study outputs and report. Outputs:

- The results of WS3 will be integrated with the findings of WS1 and 2 into a final report with key recommendations for future development of EoL care for infants, children and young people.
- The final logic model for end of life care for children
- The final model typologies
- Recommendations for future routine data collection
- We will also produce key summaries for parents, commissioners and clinicians
- An accessible animation with study findings.
- Estimates of the cost of EoL care in a paediatric population produced in this work will be submitted to the PSSRU Unit Cost of Health and Social Care Volume

Dissemination, Outputs and anticipated Impact (£36,000)

What do you intend to produce from your research

We will undertake an ongoing process of communication throughout the study to ensure that progress and interim results are made available to key decision makers and influencers throughout the study (see project timeline below). This has been accounted for in the resourcing of this study so we will have a dedicated team communications lead as well as using our departmental and the University of York press office and media team where appropriate e.g., when a key output from the study is launched.

We have identified the following local, national and international *priority audiences* for this study:

1. Parents
2. Clinicians
3. Healthcare managers and commissioners
4. Clinical membership bodies

The *key influencers* are:

1. Royal College of Paediatrics and Child Health
2. Paediatric Intensive Care Society
3. British Association of Perinatal Medicine
4. Association for Paediatric Palliative Medicine
5. Together for Short Lives

The *power brokers* and *decision makers* for this clinical area are NHS England and NICE (both part of our study steering committee).

The communication channels described in the following section will be used to update progress on the study and to disseminate the research outputs which will all be available to download from the study website:

1. Logic model of EoL care for children
2. Infographic representation of the typology of models of EoL care for children
3. Research briefing for clinicians, setting out key findings and implications for practice and training+ animation
4. Recommendations on future routine data collection
5. Summary for commissioners provided to each ICS/STP.
6. Summary of findings for parents for distribution via parent facing organisations e.g. Together for Short Lives.
7. Estimates of the cost of EoL care in a paediatric population produced in this work will be submitted to the PSSRU Unit Cost of Health and Social Care Volume
8. The wider clinical and academic audiences will be reached via conference presentations and academic articles.
9. Final report for the HS and DR journal.
10. Minimum of six journal papers (open-access).

All the study outputs will be available via the study website and via links from other websites. As well as email alerts to the key stakeholders a copy of the research briefing will be sent to the clinical leads in all the paediatric oncology centres, PICUs and NNUs in the UK.

How will you inform and engage patients, NHS and the wider population about your work

This study has received support from key organisations includes NHS England where the Chair of the Paediatric Medicine CRG and their general palliative care team have agreed to be part of the SSC. The clinical advisor to NICE for the EoL care guidelines has also agreed to be on the SSC. The Childhood cancer and leukaemia group and the Paediatric Intensive Care Society have provided letters of support and will be part of the SSC and the knowledge exchange workshops.

We appreciate that one method of communication will be unlikely to reach all of the audiences, identified above, therefore we will use the following communications channels:

- Three knowledge exchange workshops (month 13, 34 and 48 of the study). The first two will involve interim results of the study and the final one with the overall results.
- Website blogs including those from PPI members
- Email newsletters
- Conference presentations
- Academic publications (x6)
- Twitter feed
- Podcasting

These communication channels will be used to update progress on the study and to disseminate the research outputs which will all be available to download from the study website

We will work with professional (Royal College of Paediatrics and Child Health, the Association of Paediatric Palliative Medicine, Paediatric Intensive Care Society, CCLG, BAPM) and third sector (Together for Short Lives, Council for Disabled Children) to disseminate study outputs. Email alerts to highlight the key outputs from the study will be coordinated through these professional networks and third sector organisations which the applicants are linked into. The applicants have worked effectively with all these organisations in the past.

The Parent Advisory Panel and our Parent co-applicant will also assist with dissemination through their parent and family networks.

The key professional organisations which currently produce clinical guidelines on this topic (NICE, APPM)

are proposed members of our study steering committee and we will work with them to ensure effective dissemination to their members.

The wider clinical and academic audiences will be reached via presentations at the Royal College of Paediatrics and Child Health national conference and the World Congress on Paediatric Palliative Care. Six papers will be submitted for publication in peer review journal e.g. Palliative Medicine and Pediatrics. The final report will be published in the NIHR HS &DR Journal.

How will your outputs enter our health and care system or society as a whole?

The protocol for this study will be published so professionals and the public will be aware of the study. Through the mechanisms described in the section above, the multidisciplinary professionals and parents/carers will be informed of the ongoing progress, interim and final results of our study. Ensuring that the results of our study are incorporated into updated versions of clinical guidelines and policy statements is important. This will be achieved through directly informing the key professional organisations, including NHS England and NICE and also publishing the findings in peer-review journals so that any future literature searches will include the results of this study.

What further funding or support will be required if this research is successful (e,g, from NIHR, other government departments , charity or industry?)

The results of this study will inform future development of EoL care for children. The results may indicate that further investment and resources, including training key professional groups, may be required from the Department of Health/ Higher Education England or NHS England.

What are the possible barriers for further research, development, adoption and implementation?

Palliative/End of life care services for children and young people in the UK have developed locally with heavy reliance on individual clinician and third sector organisations e.g. children's hospices. There may be some resistance to change within organisations but having the key professional organisations engaged with this study throughout should enable more effective implementation of these study findings.

What do you think the impact of your research will be and for whom?

Results from this study will identify and compare the models of EoL care for infants, children and young people and the associated resource implications and child and parent outcomes. Estimates of the cost of EoL care in a paediatric population produced in this work will be submitted to the PSSRU Unit Cost of Health and Social Care Volume as their current estimates only consider adult care.

New knowledge about inequalities in access will be identified. These results will feed into the revised NICE guidelines for EoL care for this population and shape delivery of EoL care in order to utilise finite resources to maximise impact.

This will ensure that there can be genuine progress in the ability of researchers, decision makers, the children and their families to contribute to an understanding of how we can ensure the limited funding for EoL care can be used for greatest benefit for the children at the end of their lives. Facilitate understanding of what budget is needed to offer different forms of care, and importantly the role of inequality. It will also allow us to understand the likely benefits of additional funding in EoL care in terms of patients outcomes for the first time, facilitating a clear indication to budget setters.

Project/research timetable

A detailed timeline, with key milestones and deliverables is shown below:

Abbreviations PAP Parent Advisory Panel, SMT study management team, SSC Study Steering Committee, HRA Health Research Authority

YEAR 1	Jan-21	Feb-21	Mar-21	Apr-21	May-21	Jun-21	Jul-21	Aug-21	Sep-21	Oct-21	Nov-21	Dec-21
WS1												
HRA Approvals												
Recruitment Grade 5 post												
Recruitment Grade 6 post												
Survey Development/Piloting												
Data Collection												
Data Analyses												
Manuscript writing												
WS2												
HRA Approvals												
Recruitment Grade 5 post												
Study Site setup												
WS3												
HRA & CAG Approvals												
Data access application to PHE and HQIP												
Part 1 Data management/cleaning												
Study Site setup												
Communication/Dissemination Activities												
Blog												
Email updates												
Twitter updates												
Governance												
SMT meeting												
PAP meeting												
SSC meeting												
Milestones												
HRA approval obtained												
Adequate number of surveys returned												
HQIP & PHE application submitted												
Analyses WS 1 complete												
HQIP & PHE approval obtained												
Progress Report												

YEAR 2	Jan-22	Feb-22	Mar-22	Apr-22	May-22	Jun-22	Jul-22	Aug-22	Sep-22	Oct-22	Nov-22	Dec-22
WS2												
Site setup												
Recruitment Parents												
Interview Parents												
Recruitment Professionals												
Focus Groups												
Transcription and Analyses												
WS3												
Part 1 Data analyses												
Manuscript writing												
Study Site setup												
Communication/Dissemination Activities												
Blog												
Email updates												
Twitter updates												
Conference Presentation (WS1)												
First knowledge exchange event												
Governance/Reporting												
SMT meeting												
PAP meeting												
SSC meeting												
Milestones												
WS1 manuscript submitted												
Recruitment Taget Parents hit												
Recruitment Target Professionals hit												
WS2 data collection completed												
Progress Report												

YEAR 3	Jan-23	Feb-23	Mar-23	Apr-23	May-23	Jun-23	Jul-23	Aug-23	Sep-23	Oct-23	Nov-23	Dec-23
WS2												
Analyses												
Manuscript writing												
WS3												
Part 2 Recruitment												
Communication/Dissemination Activities												
Blog												
Email updates												
Twitter updates												
Conference Presentation (WS2)												
second knowledge exchange event												
Governance/Reporting												
SMT meeting												
PAP meeting												
SSC meeting												
Milestones												
WS2 Analyses complete												
WS2 Manuscript submitted												
WS3 Manuscript Part 1 submitted												
Rercuitment Target hit WS3												
Progress Report												

YEAR 4	Jan-24	Feb-24	Mar-24	Apr-24	May-24	Jun-24	Jul-24	Aug-24	Sep-24	Oct-24	Nov-24	Dec-24
WS3												
Part 2 Recruitment												
Part 2 data collection												
Part 2 data analyses												
Resource implications analyses												
Communication/Dissemination Activities												
Blog												
Email updates												
Twitter updates												
Final Knowledge exchange/Dissemination Event												
Milestones												
WS3 Part 2 Recruitment Target Hit												
All analyses complete												
Progress Report												
Final Report												
Final Manuscript submission												

Project management

Dr Lorna Fraser will lead this study and will retain overall responsibility for the delivery of this study.

The *Study Management Team* (all applicants) will meet every two months, and applicants from outside York will attend via skype or teleconference. BB will oversee WS1, supervise the WS1 researcher and assist with supervision in WS2. JT will lead WS2 and supervise the researcher. LF will lead WS 3 and supervise the researcher and project manager. HW will lead the health economic theme and CH will oversee the statistical analyses. JT and GW will co-lead the PPI for the study.

The *Parent Advisory Panel* will be established for this study**.** It will be a panel of parents and carers and will meet a minimum of twice per year, at timings which fit with the key points in the study which require input from this panel (see PPI section for details on the role of this panel).

A *Study Steering Committee* will be established with an independent chair and representation from paediatricians, palliative care specialists, commissioners, NHS England, NICE, parents/carers (to represent the Parent Advisory Panel members), appropriate national charities (Together for Short Lives), and professional bodies (Royal College of Paediatrics and Child Health, Association for Paediatric Palliative Medicine). This panel will meet once a year to assess progress of the study against the defined milestones and deliverables and provide advice and expertise to the Study Management Team.

Ethics/Regulatory Approvals

Approval will be obtained from the Health Research Authority which will include research ethics committee approval. Approval from the Confidentiality Advice Group of the HRA will be required for the processing of identifiable data without consent for part 1 of WS3.

This study involves data collection from parents of children who have died which does raise several ethical issues.

i. *Informed consent:* The first approach to the parents will be via their own clinical team. The parent will be given adequate time to ask questions and read information about the study before they consent to participation. It will be made clear on the information leaflets that there is no obligation to participate in this study.

ii. Participating in this study may raise issues or concerns not previously perceived or articulated, and therefore generate needs for support. Therefore at the end of the interview, the researcher will ask if the interviewee would like to receive a follow-up contact (via telephone call, text of email) three or four days after the interview. This will accommodate interviewees wanting to share further reflections and also provides an additional opportunity to articulate the need for support. All participants will be informed in the information leaflets that they can withdraw from the study at any point time.

iii. All members of the research team involved in direct data collection will be required to have extensive experience of doing qualitative research on sensitive topics. Staff will be trained in managing distress and the articulation of concerns.

iv. Supervision (individual and group) of researchers involved in direct data collection and data analysis will pay attention to potential impacts on researchers. The University of York has a comprehensive staff well-being service which, if appropriate, staff will be encouraged to access.

Confidentiality All data generated by this study will be anonymised and securely stored in the Department of Health Sciences at the University of York. Personal data will be stored separately from the other study data in a restricted folder which will be password protected and only accessible by members of the research team. This will include scanned copies of the consent forms. This study will comply with the new General Data Protection principles and the Research governance framework for Health and Social Care Research. All information from this study will be kept confidential.

Patient and Public Involvement (£17,940)

Please describe how patients and the public have been involved in developing this proposal

Design of the research: Our research prioritization exercise for the car of children with life-limiting conditions which involved parents and young people, service delivery models were one key topic highlighted for future research.

This proposal has been discussed with two groups of parents. Childhood Cancer conference in June 2018 (~ 80 parents, carers, professional and voluntary sector representatives). All groups voted that the child's quality of life, parents understanding, symptom control and family quality of life were the most important outcomes.

Martin House Research Centre Family Advisory Board – (7 parents). The parents were clear that research about end of life care was important. They had mixed views on the feasibility of collecting research data around the time of death and were keen that a flexible approach was used. They said that support should be signposted when completing the questionnaire and families should be offered choice about mode of completion. This group has also advised on timings e.g. they felt that 3 months after a child has died is an appropriate time to contact a parent about involvement in research. They have also advised that we use social media when recruiting parents into research studies so that they are given the choice rather than others gatekeeping on their behalf.

They have given us clear guidance on how and when to recruit bereaved parents into research studies. Our Parent co-app, Gabriella Walker, has reviewed our Plain English summary.

Please describe the ways in which patients and the public will be actively involved in the proposed research, including any training and support provided

Our PPI plans have been designed with reference to the NIHR INVOLVE National Standards for Public Involvement, which are referred to throughout to demonstrate our commitment to these standards.

The PPI workstream will be led by Dr Jo Taylor who has extensive experience with PPI and is the lead for PPI within the Martin House Research Centre. We have parent co-applicant, Gabriella Walker, whose young daughter died from a degenerative neurological condition in Feb 2019, and who will work closely with Jo to implement our PPI plans for the study. Gabriella will also attend management team meetings in her role as co-applicant, work alongside our other PPI representatives throughout the study (see below), be involved in the data analyses in WS2, and assist with dissemination of the study findings.

To ensure effective public involvement during the project, we will establish and work in partnership (Standard 2) with a parent advisory panel, who will work with the study team to:

1. guide the development of study materials, methods for recruitment and data collection (WS2)
2. review the three outcome measures proposed for inclusion in WS3 (e.g. CHU-9D (Steven) and the

POS measures POS-E, IPOS, C-POS[1] (Downing)) to check with parents as to which ones include the dimensions of most relevance to them. Parents' engagement with these outcomes and perceived value is key to determining the benefit of the packages of care.

3. contribute to data analysis (WS2) and assist with interpretation and integration of findings (all WS)
4. provide input on the design of knowledge exchange workshops
5. help to co-produce the study outputs, particularly the family facing outputs, and provide ideas for meeting the communication objectives for the study, e.g. identify routes for dissemination, sense-checking public facing outputs (Standard 4)
6. where appropriate, assist with communication and dissemination, e.g. presenting study findings at events.

The parent advisory group will be established at the start of the project and include 3-4 parents as members, recruited to achieve some diversity in experience. Members will be recruited through the research team's extensive networks, e.g. the MHRC and PICS SG, and new members will be recruited if people withdraw their involvement during the study (Standard 1).

The group will meet between 2 and 4 times each year at the University of York, depending on the involvement that is required at different stages in the study. Other planned activities where closer involvement / input is required will be undertaken by 1-2 members, with appropriate training where required (Standard 3). The WS1/2 research fellow will work closely with the group, keeping them updated on progress of the project between meetings, helping to identify and meet training needs of members, and ensuring regular feedback about how their input impacts on the study.

Two parents will also be a member of the study steering committee (Standard 6).

A PPI log, guided by the Public Involvement Impact Assessment Framework [68], will record planned and unplanned involvement, including details about who is involved and how, and how these activities impact on the study (Standard 5).

Project/research expertise

This strong, multidisciplinary team of academics and clinicians has extensive experience in the methodology and topic area of this study. The academics have strong track record of completing studies on time and within budget and several of the research team have worked together successfully on previous studies.

Dr Lorna Fraser will coordinate this study and lead WS2. Lorna is a Senior Lecturer with a background in clinical paediatrics and is the Director of the Martin House Research Centre (www.york.ac.uk/mhrc) which undertakes research in children with life-limiting conditions and medical complexity. She has expertise in cohort studies and extensive experience of accessing routinely collected healthcare data, including HES and ONS data and is a member of the Department of Health Sciences data governance committee and the HRA Confidentiality Advisory Group. She currently holds an NIHR Career Development Fellowship award.

Prof Bryony Beresford (WS 1 lead and contributing to WS2 supervision). Applied social scientist and health/care services researcher. Two relevant areas of expertise: i) has led HS-DR funded studies which have mapped /evaluated models of service delivery in different health/social care sectors, and including integrated working; ii)has extensive experience of research concerning children with LLC, and currently PI of an in-depth qualitative study of parents' experiences of the early days of bereavement.

Dr Johanna Taylor will lead WS 2 and the PPI is a mixed methods applied health researcher with experience of undertaking studies in children with LLC and their families.

Prof Catherine Hewitt, is a Senior Statistician and will oversee the statistical analyses in WS1 and 3.

Prof Jane Noyes is a Professor in Health and Social Services research and has expertise in children's palliative care research, paediatric intensive care, health economics and evidence synthesis.

Helen Weatherly will lead the health economic theme. She has considerable experience in economic evaluation applied to complex health and care interventions, including in children and young people.

Sebastian Hinde has expertise in economic evaluation applied to complex health and care interventions and analysis of observational datasets and will analyse the economic data.
Gabriella Walker is the parent of a child who died in Feb 2019.
Prof Sam Oddie is a research active clinical neonatologist who provides clinical leadership to the national neonatal audit and will provide input into WS1 and WS3.
Dr Richard Feltbower is a paediatric epidemiologist with expertise in childhood cancer and PI for the national PICU clinical audit programme (PICANet). He will assist with WS3.
Dr Bob Phillips is a clinical academic in paediatric oncology who will provide oncology input to the study.
Dr Richard Hain is a consultant in Paediatric Palliative medicine who will provide palliative medicine expertise.
Dr Chakrapani Vasudevan is a neonatal consultant who leads the Bradford neonatal palliative care program.
Dr Gayathri Subramanian is a Paediatric Intensive Care consultant with a special interest in Eol for children.

Success criteria and barriers to proposed work

The success criteria are based on achieving the key milestones and deliverables on time and within the budget of this study:

- Recruitment of research staff
- Receiving HRA approvals
- Achieving an adequate response rate for WS1
- Recruiting the required sample of healthcare professionals for WS2
- Recruiting the required sample of parents for WS2
- Completing the analyses for WS1
- Recruiting the required number of parents for WS3
- Obtaining the approvals for the routine data linkages in WS3
- Completing the analyses for WS2
- Completing the analyses for WS3
- Producing the final report, guide for professionals and summary for parents

There are several potential barriers to this proposed study:

- *Delays in recruiting the research staff.* This can be mitigated by our ability to access appropriately trained research staff from within the Martin House Research Centre team and the wider staff in the Department of Health Sciences at the University of York.
- *Low response rate to survey (WS1).* The team include key clinical specialists from oncology, PICU and NNU. We will work closely with the clinical co-apps and key professional organisations e.g PICS SG, BPAM and the CCLG to raise awareness of the study. Recruitment and reminder strategies proven to be effective by similar types of research will be implemented.
- *Failure to recruit the number of participants (WS2).* The team have experience in recruiting children and families, as well as other vulnerable populations, to research studies.
- *Failure to recruit the number of participants (WS3).* The team have experience in recruiting children and families, as well as other vulnerable populations, to research studies.
- *Delays in accessing routine data.* The PI for this study has experience of accessing routinely collected data and minimising the risk of delays in obtaining these. We have demonstrated in the project description section that we have the appropriate governance and security in place to hold, process and analyses these data. The application for these data will be submitted in the first month of this study.

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