

PROTOCOL

Advance Care Planning (ACP) and decision making at the end-of-life for children and young people (CYP) with life-limiting conditions (LLC) in the Paediatric Intensive Care Unit (PICU): A qualitative study of the experiences and perceptions of bereaved parents.

Version 1.2.1

	Chief Investigator	Co-Investigator	Researcher
Name	Dr Adrian Plunkett	Dr Sarah Mitchell	Miss Jenna Spry
Position	PIC Consultant	Honorary Research Fellow (BCH) and GP	PIC Clinical Research Nurse
Employer	Birmingham Children's Hospital Steelhouse Lane Birmingham B4 6NH	Birmingham Children's Hospital / Northfield Health Centre	Birmingham Children's Hospital
Contact Information	0121 333 9652 07714 237 481 Adrian.plunkett@bch.nhs.uk	07986183313 S.Mitchell6@nhs.net	0121 333 9684 Jenna.spry@bch.nhs.uk
	Co-investigator	Co-investigator & Qualitative methodological and user involvement advisor	
Name	Professor Jeremy Dale	Professor Jane Coad	
Position	Professor of Primary Care	Professor of Children and Family Nursing	
Employer	University of Warwick Medical School, Division of Health Sciences	Centre for Children and Families Applied Research, University of Coventry	
Contact Information	02476574880 Jeremy.dale@warwick.ac.uk	02476795966 Jane.coad@coventry.ac.uk	

Definitions

The study team: refers to the CI (Adrian Plunkett) and the Research Nurse (Jenna Spry)

The wider study team: refers to the whole team identified in the table above

The BCH Bereavement Team: refers to the Palliative Care Team, the PICU Family Liaison Team, the Chaplaincy and the bereavement team

Abstract

Background

The majority of child deaths in the UK occur in the context of a life limiting condition (LLC). The majority of these deaths occur in hospital, most commonly in the Paediatric Intensive Care Unit (PICU). Birmingham Children's Hospital (BCH) hosts the largest and busiest PICU in the UK; on average, approximately 70 children die in BCH PICU every year. Virtually all of these children have a LLC, yet virtually none have an Advance Care Plan (ACP) in place at the time of PICU admission.

Aim

To investigate the impact of end-of-life care decision-making on bereaved parents of children and young people (CYP) with LLC who die in PICU at BCH.

Design

Bereaved parents of CYP with life-limiting conditions will be identified and invited to participate in a qualitative semi-structured interview study. Thematic content analysis will be performed to explore the parents' experiences and perceptions about end of life decision making in the PICU.

Outcomes & Benefits

Improved understanding of parents' perceptions of end-of-life decision-making for children with LLC. This will add strength to the weak evidence base in this area; catalyse future research; and inform quality improvement of clinical management of this growing patient group.

Advance Care Planning (ACP) and decision making at the end-of-life for children and young people (CYP) with life-limiting conditions (LLC) in the Paediatric Intensive Care Unit (PICU): A qualitative study of the experiences and perceptions of bereaved parents.

Purpose of Proposed Investigation

The study aims to improve our understanding of parents' perceptions of end-of-life care decision-making and Advance Care Planning (ACP) for children and young people (CYP) with life-limiting conditions (LLC) in the Paediatric Intensive Care Unit (PICU). The research findings will help us to improve the future care of children with LLC, and pave the way for future research in this important area.

Background

The death of a child is one of the most complex and ethically challenging scenarios that exist in clinical medicine. With increasing numbers of CYP with LLC living in the community, and with those CYP living longer due to advances in medical technology, this scenario is increasingly important to consider. Recent epidemiological data suggest that around 49,000 CYP in the UK live with LLC, and the number is rising (1). Around 70% of children who die per year in England will have had LLC (2) (3). Deaths in this group are predictable to some extent, and therefore consideration of palliative care needs, care planning and referral to palliative care services is likely to be appropriate at some stage of the patient journey.

Currently, the majority of children who die, do so in hospital, frequently on PICU (4) (5) despite increasing evidence that the community is the preferred place of care. There is evidence that outcomes are better for families when preferences for care are enabled (6). Families have a wish for well-co-ordinated, continuous, holistic healthcare and an expectation that, as far as possible, this care should be provided at home (7).

Advance Care Planning (ACP) is a process of discussion between an individual and their care providers about their understanding of their illness and preferences for future care (8). It can help patients and families to achieve a sense of control around their treatment choices (9). ACP has been advocated to help parents plan for the unpredictable journey that is associated with caring for a CYP with a life-limiting condition (10). ACP is a core element of national adult and paediatric palliative care strategies (11), and has been described as a "standard of care" (12). However, although ACP can help to elicit patient and family choices, discussions around death are difficult and can be distressing for all involved, and may therefore not take place. Currently the evidence base for ACP, particularly in paediatrics, is scarce.

The West Midlands Paediatric Palliative Care Toolkit

In 2010, the Department of Health (DH) invested £30 million in to projects designed to work towards sustainable, nationally equitable services – the "30 Million Stars" projects (13). The West Midlands, via the West Midlands Paediatric Palliative Care Network (WMPPCN), was successful in obtaining over £5 million of that funding. One of the funded projects was the development of the WMPPCN Palliative Care Toolkit, which included a formal ACP document (14).

Epidemiological studies (15) suggest that there are over 5000 CYP living in the West Midlands with LLC who may benefit from ACP. Gathering evidence around the experiences of patients and families at this point in time provides an opportunity to compare the effects of having an ACP versus not. This study will particularly focus on the parents of CYP with LLC who have died in PICU.

National Perspective and Policy

The Chief Medical Officer's Annual Report 2012, "Our children deserve better: prevention pays", focussed on the importance of health in childhood, including early interventions and coordination of care for those with long term conditions (16). The delivery of integrated, holistic healthcare for patients with long-term LLCs is a priority area elsewhere in the NHS and for the UK Government (17). The Palliative Care Funding Review has advocated the provision of a system "which provides better outcomes for patients and better value for the NHS" (18).

The proposed study builds on previous research completed by the study team in Birmingham Children's Hospital PICU:

- Dr Plunkett - Epidemiological study of temporal trends in length of stay in children who died in PICU (19).
- Dr Mitchell – Qualitative study examining the end-of-life care decision-making process from the perspective of senior PICU medical and nursing staff (20).
- Miss Spry – Qualitative study exploring the experiences of PICU nurses, when caring for a child whose care changes from curative/treatment to palliative and end-of-life care (unpublished).
- Dr Mitchell and Dr Plunkett – Survey of UK PICUs regarding use of formal advanced care planning documents (21).

The research questions were generated following presentation of the results of Dr Mitchell's study (20) at the PICU research and audit meeting. It is also informed by a service user involvement event hosted by the WMPPCN, attended by parent champions and a young ambassador for Acorns.

Research Questions

1. What are the experiences and perceptions of bereaved parents in relation to ACP and end-of-life care decision making in PICU for CYP with life-limiting illness?
2. What are the facilitators and barriers to end-of-life care decision-making, including ACP for CYP with LLCs as perceived by bereaved parents?
3. What are the benefits and risks related to the ACP process as perceived by the parents of CYP with LLCs who have died on PICU?

Plan of investigation

The study will comprise the following four phases:

1. Review of the published evidence in this field
2. Data collection.
3. Data analysis.

4. Publication and dissemination.

Patient and Public Involvement (PPI) will be sought for as many aspects of the study as they wish to contribute to. For this study three bereaved families well known to the PICU team will be contacted about potential PPI work. (See separate PPI section on pages 10-11)

Phase 1 – literature review:

A comprehensive review of existing literature will be completed to address the question:

“What is the current, published evidence base describing parental experiences of end of life decision making and Advanced Care Planning for their children on PICU?”

Initially the Cochrane Review Library will be searched however it is anticipated that there will be no relevant reviews. Online databases Medline, Embase, and Cinahl will then be searched with search terms derived using the SPICE model:

Setting	Perspective	Intervention	Context	Evaluation
PICU	Parents	End of life care	CYP	Decision making

SPICE Model (Booth, 2006)

Specific search terms can be found in Appendix 1. Additional references will be located from reference list searches.

The study team (Adrian Plunkett and Jenna Spry) will review the titles, abstracts and then full text articles to identify relevant literature. The relevance and quality of the remaining articles will then be assessed using the CASP checklist for qualitative studies.

The results of this review will inform the design of the interview schedule.

Phase 2 – data collection (includes study design and methodology):

Study design

A qualitative design will be used to elicit details and reflections about what people did, how they thought and felt, including what influenced them and why, within a particular environment or situation (22); in this case the end-of-life-care of a CYP with a LLC in PICU. Such methods are appropriate for studying complex, emotional subjects such as end-of-life care, and have the benefits of allowing an in-depth insight into the needs of families, understanding their experiences, and providing a human dimension (23). The PICU at Birmingham Children’s Hospital is an extremely complex and emotionally charged environment to experience. It is a large mixed unit with 31 beds, seeing approximately 1400 admissions per year, from multiple specialities including cardiac surgery (40% of planned admissions), liver and small bowel transplant, oncology, trauma and burns, as well as general surgery and medicine. Approximately 70 CYP die on the PICU each year.

A sample size of the parents of 20 CYP who have died on PICU will be aimed for. For the purposes of the study, “parents” will be those who are legally the parents or guardians of the child or young person, whether biological or adopted. This will give a variable sample size with the maximum of 40 individuals if 2 parents for each child participate.

Data will be collected using one-to-one, in-depth, semi-structured interviews with parents. These are the preferred data collection method, since the confidential nature of the interview allows participants to freely disclose their experiences, thoughts and feelings relating to a subject, while the semi-structured approach allows some focus on the research questions (24). Another benefit is that interviews can be arranged at a time to suit participants. Data collection will therefore not be dependent on the organisation of focus groups. Other qualitative methodology, including observational studies and conversation analysis, would not be feasible in this particular context. Questionnaire studies are unlikely to provide the rich, contextual data that is expected from an interview study.

Previous studies involving interviews with bereaved relatives have demonstrated that the interview process can be a positive experience for participants (25) (26) (27) (28). The VOICES survey and associated research suggests that the views of bereaved relatives provide a valid method of evaluation of services. (29)

Sample

Purposive sampling involves deliberately selecting participants because they have the experience or characteristics that the researchers are looking to explore. Purposive sampling will be used for this study in order to reach bereaved parents who have experienced the end-of-life-care of their child on PICU in order that the sample is able to provide the data needed for the aim of the study. More random techniques for sampling would not benefit this study as it is important to interview those who meet these specific criteria.

Participants

Participants will be identified from mortality records in PICU. The study team will screen PICU deaths prospectively (from the time of study commencement), and retrospectively for a period of 12 months prior to study commencement, with the aid of an existing PICU database. Retrospective screening will allow extension of recruitment pool to facilitate adequate sample size.

For the purposes of this study, CYP will be defined as **aged 0 to 19 years**, inclusive (this is the age criterion for admission to BCH PICU, **including CYP who are undergoing transition to adult services**). Although Neonatal Intensive Care Units are not involved in this study, neonates who require PICU at BCH will be included. Recruitment will be supported by the BCH bereavement team.

LLC will be defined as “those for which there is no reasonable hope of cure and from which children or young people will die” (30). These can be further categorised into four groups, each with distinctive characteristics and illness trajectories:

- *Group 1:* life-threatening conditions where access to palliative care services is necessary alongside attempts at curative treatment and / or if treatment fails, such as cancer.
- *Group 2:* conditions such as Duchenne muscular dystrophy, where premature death is inevitable, but where there may be long periods where the child is well.
- *Group 3:* progressive conditions without curative treatment options, such as Batten disease.
- *Group 4:* irreversible but non-progressive conditions, with complex disabilities and healthcare needs which lead to increased likelihood of premature death, such as severe brain injury.

Where there is uncertainty about which of these categories a child would fall in to, consensus will be sought from the wider study team to guide suitability for inclusion. For the purposes of the study, “parents” will be those who are legally the parents or guardians of the child or young person, whether biological or adopted.

Inclusion Criteria

- Bereaved parents of CYP who had a LLC as defined by Together for Short Lives in PICU during the study period or 12 months previously

Exclusion Criteria

- Parents who are unwilling or unable to provide valid, informed consent.
- Bereaved parents of CYP who have died from acute illness or trauma.
- Parents aged 16 years or less at the time of recruitment

It is important to include parents for whom English is not the first language; however the use of interpreters in qualitative studies is not straightforward. Should the need arise, the feasibility of using interpreters within the financial constraints of the project will be reviewed, and the BCH interpreter service will be approached for support with provision of an interpreter.

Recruitment and consent

Prospectively identified bereaved parents will be invited to participate in the study at the time of invitation to bereavement follow-up, or at the PICU bereavement meeting. This is a routine PICU follow-up bereavement meeting, and typically occurs 6-12 weeks after the death of the child at BCH. The bereavement meeting has been chosen as a suitable time for potential recruitment of parents due to the likelihood that a good clinical relationship has already been established, and because parents have already agreed to travel back to BCH for the meeting.

Retrospectively identified bereaved parents will be invited to receive information about the study during on-going bereavement follow-up and contact with BCH (via the BCH Bereavement Team). Parents who indicate interest in the study at this stage will be contacted by the study team and formally invited to participate.

Parents will receive a letter of introduction, a participant information sheet, and a detailed consent form. **After written information has been delivered, a member of the research team will contact each family once by telephone or email, to give the opportunity to discuss the study further.** It will be made clear that participation is entirely voluntary, and participants may withdraw consent at any time. Parents will be offered the opportunity to provide consent at any time. If they wish to withdraw consent, all data relating to the interview, including recordings and transcripts, will be destroyed and not included in the study.

The study team will aim to create a sample representing the breadth of LLCs, ages and ethnicities seen in the PICU, however even with purposive sampling this may not be achievable with a sample number anticipated for this study, which will greatly depend upon who responds to the invitation to participate.

Sample size

Around 4-10 deaths occur in Birmingham Children’s Hospital PIC per month, therefore a sample size of the parents of 20 CYP (i.e. up to 40 parents in 20 interviews) will be aimed for in the study period. Attempts will be made to engage both parents where possible. Interviews will be conducted with either both parents together or separately as individuals according to parental preference. In order to maximise recruitment, telephone interviews will be offered to those unable, or would prefer not to, to attend a face to face interview. This study is limited by the time, resources and funding available, therefore in reporting the findings, it will be transparent about the limitations this posed for recruitment, sample size and potential data saturation.

The ideal sample size for a qualitative study of this nature is one which is sufficient to allow data saturation. This occurs when the interviews are no longer providing any new information or insights in responses (22). Data saturation is a complex concept with different meanings assigned to it. The concept originates from within 'Grounded Theory' which provides clear guidance and definition, but outside of this methodology, its use and meaning varies greatly. When researching a topic such as parental end of life experiences, it would be difficult to know that no new information would be shared in a future interview.

“...to the extent that each life is unique, no data are ever truly saturated: There are always new things to explore.”

(34)

Qualitative studies are often confined by funding, resources and time, and this probably impacts on sample size more often than data saturation.

Interview Plan

Setting

If parents choose to participate, they will be offered the opportunity to take part in the interview at a time of their choice. If this is on the same day as the bereavement meeting at BCH, arrangements will be made to accommodate this. Otherwise, a future date will be arranged during the study period at their convenience. The location of the interview will either be at BCH or at the parents' home, depending on their preference. One or both parents will be interviewed, depending on preference. Attempts will be made to engage both parents where possible with the offer of telephone interviews to facilitate this.

Procedure

Interviews will be digitally audio-recorded, and field notes made. The interview will not be directive, and there will be no time constraints other than those of the participants. The topic guide (Appendix 2) has been developed by the study team in conjunction with the PPI families. The topic guide will be developed iteratively throughout the study, with changes made to reflect any important emergent themes from initial analysis. The interview will start with asking parents to talk about their child, their illness and death in whatever way they feel able. Further questions will specifically ask about their experience of health care and other support, and, where appropriate, ACP. The interview will be conducted using a blended approach of passive (listening) and more active interview techniques as appropriate.

Demographic data will be collected from the parents at each interview, including their age, other children, and marital status (See Appendix 3). This information will be used to add context to the family situation during analysis and presentation of themes.

Conversations with bereaved parents will be emotive, and may cause distress. The interview will be informal and conducted in a conversational manner, allowing participants to set the pace. Should participants become in any way distressed during the interview process, they will be offered the chance to pause or stop the interview. Adequate time will be allowed for the participant to recover and debrief.

A distress protocol (Appendix 4) adapted for this study from a published tool (31), will be used by the researcher during the interview process, if any of the participants display any signs of increased stress or emotional distress.

Should participants raise any cause for concern during or at the end of the interview, such as suicidal ideation, arrangements will be made, with the participant's knowledge, to contact their GP and an

appointment made for follow-up as soon as possible. A follow-up telephone call will also be made by the researcher. The research nurse, who will be conducting the interviews, has many years of experience as a PICU nurse and has conducted qualitative semi-structures interviews regarding end of life care in PICU for a previous study. It is hoped that this previous experience will reassure participants and foster an environment of trust and of a shared knowledge of the PICU; encouraging detailed conversations.

Materials

Digital audio equipment will be used to record interviews, unless consent is withheld for this. In this situation, detailed notes will be made during the interview instead. Agreeing to the recording of the interview will not be a condition of consent.

Audio recordings will be transcribed verbatim by professional transcription services governed by the Data Protection Act. Each participant will have a study number assigned and the transcripts will be anonymised using pre-determined codes or alternatives provided by the research team. Professional transcription services have existing confidentiality and storage agreements, with processes in place to ensure typists are aware that they might be exposed to distressing material, and ways of managing issues should they arise. Participants will be offered the opportunity to review the transcript of their data. Any feedback or removal of data will be discussed with the study team and if they still require it to be removed, it will be. Digital recordings will be destroyed following data analysis.

Phase 3: Data Analysis

Data analysis will commence alongside data collection where possible, and will inform the iterative development of the interview schedule.

Thematic analysis of transcripts and field notes (32) will be carried out using an inductive approach. The analysis process will be guided by the 6 phases recommended by Braun & Clarke (33):

1. Familiarisation with the data
2. Coding the data (complete coding will be carried out to identify any data of relevance to the research study questions and aims)
3. Searching for themes
4. Reviewing themes
5. Defining and naming themes
6. Finalising analysis and writing the report

This is anticipated to be a manual process, however use of qualitative data handling computer packages, such as NVivo will be considered.

Verification

Verification of the study data will be enhanced by peer review of interview transcripts (24), which will be carried out by the wider study team (AP, SM, JD and JC). Team members will each review and independently code a selection of transcripts. Coding will then be discussed and compared, allowing further development of themes. This method decreases lone researcher bias. The PPI families will also be asked to review the themes (during phases 4 & 5 as described above) to check whether the themes and coding reflects their own experiences as well as the experiences that have been shared. This opportunity will also be offered to the participants themselves.

Phase 4: Publication and Dissemination

The results of the study will be presented for submission to relevant national and international, peer-reviewed journals, such as Archives of Disease in Childhood, Pediatric Critical Care Medicine

and the Journal of Medical Ethics. Presentations will be delivered locally, and abstracts prepared for submission to national and international conferences (e.g. RCPCH scientific meeting and meetings of the Paediatric Intensive Care Society and European Society of Paediatric and Neonatal Intensive Care Medicine). The PPI group will guide the study team on how best to feedback to the study participants. This written report and letter of thanks will be sent to all who participate. It is hoped that this continued involvement in the study enable parents to see that the information they shared has been used with care and sensitivity.

It is anticipated that completion of this study will lead to further research in this emerging field, such as detailed investigation of the effect of multiculturalism and religion in end-of-life care for CYP; the involvement of CYP with capacity in their own end-of-life care planning discussions; investigation in to the impact on healthcare professionals of end-of-life care for CYP, including the effects of moral distress, and how this is managed; and further studies to investigate the impact of bereavement, including long term morbidity for parents, for example by way of a longitudinal qualitative study.

Patient and Public Involvement

Introduction

The involvement of patients and the public in research is extremely important, and is strongly recommended by the NIHR (2014) (National Institute for Health Research) and INVOLVE (2012). INVOLVE defines public involvement in research as research being carried out 'with' or 'by' members of the public rather than 'to', 'about' or 'for' them.

The term 'public' can refer to service users, parents, organisation or charity representatives, potential patients and carers (Involve, 2012). Reasons cited for its importance within health and social care research include:

- Ensures the research is, and remains relevant
- Helps to identify new areas for research
- Improves research quality
- Includes different perspectives (Involve, 2012 & NIHR, 2014)

The study team share these views and acknowledge the important input PPI families could provide the study.

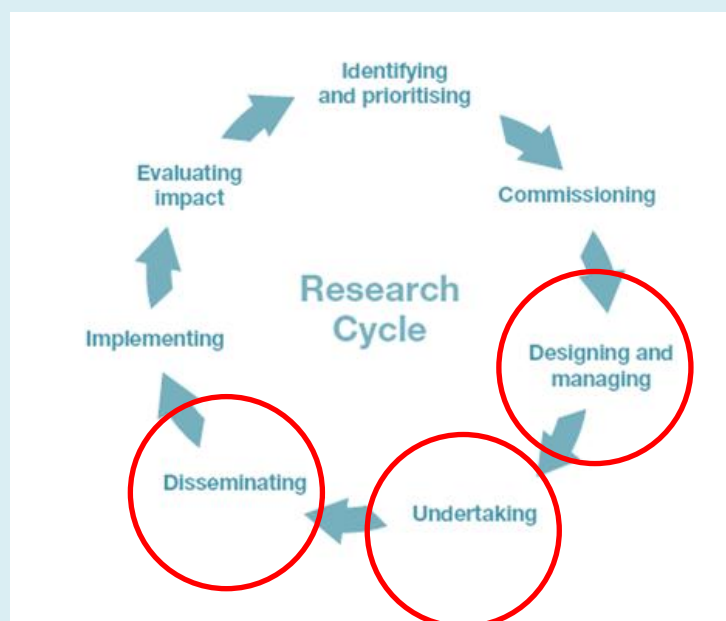
Aim

The aim was to recruit the parents of 3 patients who have died on PICU in the last 2 years to form a PPI or study advisory group. The 3 patients' parents were identified by the study team as families who had differing experiences of PICU and decision making in regards to palliative and end of life care. These parents have a good level of understanding of spoken and written English which would be important in the roles for the advisory group.

Recruitment

The identified parents were contacted via their primary contact within the hospital – either their child's named PICU consultant or the family liaison team and bereavement team members. After initial contact was made they were sent a leaflet inviting them to be involved and contact details for the study team. Ethical approval and written consent are not required for PPI work, however we asked for verbal consent to take part. At this point 2 families have been involved.

PPI work



These are the three areas of the research cycle described by INVOLVE (2012) in which we hope to involve parents.

Firstly, we asked parents to assist with the **design** of several integral aspects of the research study. This included assistance in designing and writing information leaflets that will be given to bereaved parents when inviting them to take part in the study interviews and the interview topic guide. Their experience will also be extremely valuable when considering how and when we should offer further support to the parents taking part.

Secondly, there will be the opportunity for the parent advisors to assist with **undertaking** the verification of the emergent themes.

Lastly, we would like the parents involved in the design of the research study to help us decide how we **share the information** we have gathered and the results of the study, particularly in regards to feeding back to the participants.

The parents approached to take part in the PPI work for the study are able to decide how much and with what aspects of the work they would like to be involved in. They can join and leave the process whenever they wish. So far the families have preferred to communicate via email but face to face meetings will also be possible.

Training

No specific training is planned; however the study team will be available for advice and to signpost to sources of information which might be useful for the families. The wider study team have experience of working with PPI groups for large research studies and will be able to advise about any support or training needs that may be identified throughout the process.

Support

Support will be available from the hospital bereavement team, PICU family liaison team and chaplaincy department, or the families' usual source of support.

The future – involvement after the study ends

Future involvement will be decided by the parents themselves. They will have experience of PPI work and research in sensitive areas such as death, bereavement and care in PICU, which will be a valuable resource for researchers wishing to run research in these areas in the future. We would hope that the group will be interested in this and providing continued support and friendship for one another.

Summary of Ethical Issues

Identification of potential participants

The study team will screen PICU deaths prospectively (from the time of study commencement), and retrospectively for a period of 12 months prior to study commencement, with the aid of an existing PICU database. Both the CI and research nurse are part of the clinical team who already have access to this database and there is therefore no need to share any patient or parent identifying information with anyone else.

Initial contact and provision of information

Prospectively identified bereaved parents will be invited to participate in the study at the time of invitation to bereavement follow-up, or at the PICU bereavement meeting. This is a routine PICU follow-up bereavement meeting, and typically occurs 6-12 weeks after the death of the child at BCH. The bereavement meeting has been chosen as a suitable time for potential recruitment of parents due to the likelihood that a good clinical relationship has already been established, and because parents have already agreed to travel back to BCH for the meeting. They will also have access to support from the BCH Bereavement Team and to ask questions and seek clarification from the study team. It is anticipated that invitation to take part in a study at this stage will not create any additional distress.

Retrospectively identified bereaved parents will be invited to receive information about the study during on-going bereavement follow-up and contact with BCH (via The BCH Bereavement Team). Parents who indicate interest in the study at this stage will be contacted by the study team and formally invited to participate.

Parents who indicate their interest at this initial stage will receive a letter of introduction, a participant information sheet, and a detailed consent form. The opportunity to discuss the study further will be offered. It will be made clear that participation is entirely voluntary, and participants may withdraw consent at any time. Parents will be offered the opportunity to provide consent at any time. If they wish to withdraw consent, all data relating to the interview, including recordings and transcripts, will be destroyed and not included in the study.

The study team are mindful that the receipt of information from PICU about their child who died may be upsetting for the parents and every effort will be made to ensure that information is not sent at the time of important dates such as the child's birthday or the anniversary of their death.

Interview scheduling

If parents choose to participate, they will be offered the opportunity to take part in the interview at a time of their choice. If this is on the same day as the bereavement meeting at BCH, arrangements will be made to accommodate this. Otherwise, a future date will be arranged during the study period at their convenience. The location of the interview will either be at BCH or at the parents' home, depending on their preference. One or both parents will be interviewed, depending on preference. Attempts will be made to engage both parents where possible with the offer of telephone interviews to facilitate this. These choices are important to offer the parent as it enables them to have some control over the location, timing and privacy of the interview; hopefully ensuring that they are not inconvenienced too much by participating and choose and time and setting in which they will feel most comfortable.

Where the location is the family home, the research nurse will be travelling there alone. The hospital has a detailed Lone Worker Policy which will be followed. This includes an independent person having access to the diary of where and when each visit is, and receiving a contact phone call to inform when a visit is finished. The CI will have access to this information as he will already know the identity of the participants from the identification process.

Interview process

Participants: The research nurse, who will be conducting the interviews, has many years of experience as a PICU nurse and has conducted qualitative semi-structures interviews regarding end of life care in PICU for a previous study. It is hoped that this previous experience will reassure participants and foster an environment of trust and of a shared knowledge of the PICU; encouraging detailed conversations.

Minimal demographic data will be collected from the parents at each interview, including their age, other children, and marital status.

Interviews will be digitally audio-recorded, and field notes made. The interview will not be directive, and there will be no time constraints other than those of the participants. The interview topic guide has been developed by the study team in conjunction with the PPI families. The PPI families' involvement in this aspect of the study is critical to optimise the questions and language used. They have been through similar experiences to the families who are being interviewed and will have 'insider knowledge' about how questions sound and whether they have the potential to offend or cause undue distress.

Conversations with bereaved parents will be emotive, and may cause distress. The interview will be informal and conducted in a conversational manner, allowing participants to set the pace. Should participants become in any way distressed during the interview process, they will be offered the chance to pause or stop the interview. Adequate time will be allowed for the participant to recover and debrief. A distress protocol adapted for this study from a published tool (31), will be used by the researcher during the interview process, if any of the participants display any signs of increased stress or emotional distress. Should participants raise any cause for concern during or at the end of the interview, such as suicidal ideation, arrangements will be made, with the participant's knowledge, to contact their GP and an appointment made for follow-up as soon as possible. A follow-up telephone call will also be made by the researcher. All participants will have access to support within the hospital from the BCH bereavement team. If participants opt for a telephone interview, the researcher's ability to see visual cues of emotional upset is absent. They will therefore need to be mindful of this and listen carefully to auditory cues and responses. The same actions would be taken as for the face to face interviews.

Previous research studies where bereaved parents have been interviewed have found that participants do not report any harm or regrets about taking part in the study, with most reporting some kind of benefit for themselves. (25) (26)

Researcher: As previously mentioned the research nurse will follow the guidance laid out in the Lone Worker Policy to maximise their safety when visiting participants' homes. Arrangements have also been made with the PICU Staff Support Practitioner for regular meetings and debriefing sessions for the research nurse. This is important as the emotive information shared has the potential to impact on the research nurse's own health and well-being. Regular meetings will allow for close supervision of this.

Data Storage

The identity of potential and consented participants will only be known by the study team. Minimal identifiable information collected by the study team will be kept on in password protected document on a secure NHS trust computer drive, accessible by the study team only.

Copies of consent forms will be locked in the PICU research team office which is located on a locked corridor with limited access. Audio recordings will also be kept securely in this office until the end of the analysis phase, after which they will be destroyed. Transcripts and analysis documentation will be made anonymous.

Transcription

Professional transcription services will be used for the transcription of the audio recordings. These services have existing confidentiality and storage agreements, with processes in place to ensure

typists are aware that they might be exposed to distressing material, and ways of managing issues should they arise.

Feedback

All participants will be offered the opportunity to read their own transcript and to review the themes which emerge from the analysis. The PPI families will also be offered the opportunity to verify the themes. A written report and letter of thanks will be sent to all who participate. It is hoped that this continued involvement in the study enable parents to see that the information they shared has been used with care and sensitivity.

*“How people die remains in the
memory of those who live on”
Dame Cicely Saunders (founder of
the modern hospice movement) (8)*

Paediatric Intensive Care is increasingly successful, in terms of achieving its primary goal of reducing preventable deaths: the crude mortality rate of children in British PICUs is falling year on year. But behind this success story is a relentless rise in the prevalence of LLC in British children. Thus, while more lives are saved, a higher proportion of survivors go into the community with disabilities and LLC. One consequence of this phenomenon is that nature and modality of death in the PICU is changing. It is less common for children to die suddenly, from acute illness; and more common for children to have prolonged, drawn-out deaths, resultant from their underlying chronic disease. Most of these children die as a result of withdrawal of life support agreed with the parents, but this agreement is rarely in place at the time of PICU admission, despite the acknowledgement of the LLC and the knowledge of the natural history of the disease. BCH is the biggest and busiest PICU in the UK, in terms of patient throughput, and is therefore an ideal environment to study the effects of child death on the parents. BCH is also the source of the WMPPCN Advance Care Plan, rendering it all the more suitable for this study.

The proposed study would be able to give a very important opportunity for parents' of bereaved children to share their stories and perceptions with the potential to inform the care of future children with LLCs and their families. This study, in addition to Dr. Mitchell's, Dr. Plunkett's and Miss Spry's previous work, would help cement a reputation for BCH as a national leader in this growing area.

References

1. *Rising national prevalence of life-limiting conditions in children in England.* **Fraser LK, Miller M, Hain R, Norman P, Aldridge J, McKinney PA, Parslow RC.** 4, 2012, *Pediatrics*, Vol. 129, pp. e923 - e929.
2. **Confidential Enquiry into Maternal and Child Health.** *Why Children Die: A pilot study (2006).* London : CEMACH, 2008.
3. **Office for National Statistics.** *Mortality Statistics: deaths registered in 2007.* 2007.
4. *Characteristics of deaths occurring in hospitalised children: changing trends.* **Ramnarayan P, Craig F, Petros A and Pierce C.** 5, 2007, *Journal of Medical Ethics*, Vol. 33, pp. 255-60.
5. *How children die in hospital.* **McCallum DE, Byrne P, Bruera E.** 6, 2000, *Journal of Pain and Symptom Management*, Vol. 20, pp. 417-23.
6. *The Dying Child.* **D, Black.** 1998, *British Medical Journal*, Vol. 316, pp. 1376-78.
7. **Coad J, Brown E, Owens C, Ashley N, May K, Kaur J.** *The Big Study Strand 2 Report: Qualitative Analysis of families' met and unmet needs. The Big Study Final Report.* s.l. : Together for Short Lives, 2012.
8. **Department of Health.** *End of Life Care Strategy. Promoting high quality care for all adults at the end of life.* London : Department of Health, 2008.
9. *A New Model of Advanced Care planning: Observations from people with HIV.* **Martin DK, Thiel EC, Singer PA.** 1999, *Archives of Internal Medicine.*, Vol. 159, pp. 86-92.
10. *Advanced care planning in children with life-limiting conditions - the Wishes Document.* **Fraser J, Harris N, Berringer AJ, Prescott H, Finlay F.** 2, 2010, *Archives of Diseases in Childhood.*, Vol. 95, pp. 79-82.
11. **Department of Health.** *Better Care: Better Lives.* London : Department of Health, 2008.
12. **K, Thomas.** *The Gold Standards Framework. A Programme for Community Palliative Care.* s.l. : Department of Health, 2005.
13. **Together for Short Lives.** s.l. : Department of Health, 2011.
14. **West Midlands Paediatric Palliative Care Network.** *Advanced Care Plan for a Child or Young Person West Midlands Paediatric Palliative Care Toolkit.* 2011.
15. **Fraser LK, Parslow RC, McKinney PA, Miller M, Aldridge JM, Hain R.** *Life-limiting and life-threatening conditions in children and young people in the United Kingdom; national and regional prevalence in relation to socioeconomic status and ethnicity.* Leeds : Division of Epidemiology, University of Leeds, 2011.
16. **SC, Davies.** *Our Children Deserve better: Prevention pays.* London : Department of Health, 2012.

17. **Goodwin N, Sonola L, Thiel V, Kodner DL.** *Co-ordinated care for people with complex chronic conditions: Key lessons and markers for success.* London : The King's Fund, 2013.
18. **Hughes-Hallett T, Craft A, Davies C, Mackay I, Neilsson T.** *Palliative Care Funding Review. Funding the Right Care and Support for Everyone.* 2011.
19. *Is it taking longer to die in Paediatric Intensive Care?* **Plunkett A, Parslow RC and Lima S.** 4, 2014, *Pediatric Critical Care Medicine*, Vol. 15, pp. Suppl - 16-17.
20. *Advance Care Planning in palliative care: A qualitative investigation into the perspective of Paediatric Intensive Care Unit staff.* **J, Mitchell S and Dale.** 2015, *Palliative Medicine*, pp. 1-9.
21. *Use of formal advance care planning documents: a national survey of UK Paediatric Intensive Care Units.* **Mitchell S, Plunkett A and Dale J.** 2014, *Archives of Diseases in Childhood*, Vol. 99, pp. 327-330.
22. **J, Mason.** *Qualitative researching.* London : SAGE Publishing, 2002.
23. *Achieving a good death for all.* **Ellershaw J, Dewar S, Murphy D.** 2010, *British Medical Journal*, Vol. 341, pp. 656-58.
24. *Methods of data collection in qualitative research: interviews and focus groups.* **Gill P, Stewart K, Treasure E, Chadwick B.** 6, 2008, *British Dental Journal*, Vol. 204, pp. 291-5.
25. *Bereaved parents' experience of research participation.* **K, Dyregrov.** 2004, *Social Science & Medicine*, Vol. 58, pp. 391-400.
26. *Research with bereaved parents: a question of how not why.* **Hynson JL, Aroni R, Bauld C, Sawyer SM.** 2006, *Palliative Medicine*, Vol. 20.
27. *Bereaved relatives' views about participating in cancer research.* **Koffman J, Higginson IJ, Hall S, Riley J, McCrone P, Gomes B.** 4, 2012, *Palliative Medicine*, Vol. 26, pp. 379-383.
28. *Asking next-of-kin of recently deceased cancer patients to take part in research: 11 of 20 surveyed found it a positive experience.* **Foster TL, Hendricks-Ferguson VL.** 2, 2013, *Evidence-based Nursing*, Vol. 16, pp. 64-65.
29. **Office for national Statistics.** *National Bereavement Survey (VOICES), 2011.* London : The Office of National Statistics, 2012.
30. **Together for Short lives.** Definitions. [Online]
http://www.togetherforshortlives.org.uk/professionals/childrens_palliative_care_essentials/definitions.
31. *Developing distress protocols for research on sensitive topics.* **Draucker CB, Martsof DS, Poole C.** 5, 2009, *Archives of Psychiatric Nursing*, Vol. 23, pp. 343-50.
32. **D, Lacey A. and Luff.** *Qualitative Research Analysis.* Yorkshire & the Humber : The NIHR RDS for the East Midlands , 2007.

33. **V, Braun V and Clarke.** *Successful Qualitative Research: a practical guide for beginners.* London : SAGE, 2006.

34. *“Researcher Saturation”: The Impact of Data Triangulation and Intensive-Research Practices on the Researcher and Qualitative Research Process.* **Wray N, Markovic M, Manderson L.** 10, 2007, *Qualitative Health Research*, Vol. 17, pp. 1392-1402.

APPENDIX 1

Spice Model (Booth, 2006)

Setting	Perspective	Intervention	Context	Evaluation
PICU	Parents	End of life care	CYP	Decision making

Critical care Critical illness Critically ill Critically ill patient ICU Intensive care Intensive care neonatal Intensive care unit Intensive care units, neonatal Intensive care units, pediatric Neonatal intensive care unit(s) NICU Paediatric intensive care Paediatric intensive care unit(s) Pediatric critical care nursing Pediatric intensive care Pediatric intensive care unit(s) PIC PICU	Caregiver(s) Carer(s) Families Family Father(s) Guardian(s) Mother(s) Parent(s) Parental Parental attitudes Parental consent Parental role(s) Parenting Professional family relations	Attitude to death Bereavement End of life End of life care Life limiting illness Life limiting illnesses Life support care Life sustaining Life sustaining treatment Palliative care Palliative medicine Palliative therapy Terminal care Terminal disease Terminal illness Terminal illnesses Terminally ill Terminally ill patient(s) Treatment withdrawal	Adolescent(s) Child(s) Childhood Children(s) Hospitals, pediatric Infancy Infant(s) Neonatal Neonate(s) Paediatric(s) Pediatric care Pediatric hospital Pediatric(s)	ACP(s) Advance care discussion(s) Advance care plan(s) Advance care planning Advance directives Communication Consumer participation Decision making Family conference(s) Interpersonal communication Living will Parallel planning Patient care Patient care planning
--	--	--	--	---

APPENDIX 2

Interview Topic Guide

This interview topic guide is designed to illustrate the topics which may be covered in the semi-structured interviews with bereaved parents.

Each section gives example questions and ideas for wording and prompts to be used.

It is not designed to be followed in a prescriptive manner with all questions being asked.

Each interview will be conducted in a conversational manner, with direction being controlled by the interviewee.

The timing of questions will be judged by the interviewer, dependent upon what is being discussed and the overall wellbeing of the interviewee.

Demographics and Introduction

(Would be useful to already have some information from the medical notes prior to interview)

Reminder of what the interview is about

Reassure about pausing/stopping etc.

Answer any questions

About child and family

- Please can you start by telling me about [child's name] and your family?
- Were you aware that [] was unwell before they were born?
- When was [] diagnosed? (where were they at this time – home, hospital, PICU)
- What was that like for you and your family?
- At that point did you know that [] life would be limited?
- What plans or decisions were made at this point about their care?
- Who was involved?
- What were your wishes and fears at this time?

Child becoming ill and time on PICU

- Can you tell me about when [] became poorly and came in to hospital and PICU?
- What was that like? / How did you feel?
- Did you have plans or discussions about [] admission to PICU?
- How many times did they come to PICU? / How long were they on PICU?
- What were your wishes and fears at this time?

Decision Making and planning on PICU: General

- When [] was being cared for on PICU, can you tell me about your experiences of decision making and planning for their care in the future?
- What decisions? / Who initiated? / Discussion? / Who made final decision? / Were you involved? How? / Timing / Feelings
- Plans made? You? / Medical team(s) / Together? / formal or informal? / Timing / Feelings
- Did you have any wishes or fears about decisions being made or planning about [] care at this time?

EOLC/PC Decisions, ACP and Planning on PICU

- When did you realise/understand that [] was going to die? **Prompts:** medics told you, you saw a difference in [] condition, event occurred, planning, support from others – who?
- Did you make plans for their end of life care?
- At this point did you have any idea about what ‘end of life care’ might mean or what it might look like?
- What were your wishes and fears at this time?
- Was a decision made to limit treatment / withdraw active treatment e.g. taking the tube out?
- How was this decision made?

ACP specific

- When [] was on PICU did you / were you offered the chance to complete an Advanced Care Plan (‘purple pages’)?
- Did you know what it was?
- **Prompts:** When? Who? How? Helpful? Problems?
- What was the process like? / How did it feel?
- Did it get reviewed at any time?
- What do you think was the most useful thing about the ACP? Were there any problems with using one?
- What were your hopes and fears at this time?
- **If not:**
- Do you know what an ACP is?
- Do you think you would have liked to have been offered the opportunity to complete one?
- Do you think you would have used it?
- What do you think would be the most useful thing about an ACP document?
- What do you think the problems would be with using one?

Around Time of Death and Beyond

- Do you feel able to describe what happened when [] died? **Prompts:** Decision, planned, ALTE leading to death, where, when, who?
- What was most important to you at this time?
- Who was there? Had this been planned previously? What decisions about who was present did you make?
- Who helped you or supported you?
- Did time / planning / discussions influence the decisions made? How?
- Are there any plans / decisions that you would make differently?
- Is there any advice that you would give other parents facing a similar situation in the future?

Taking part in the Interview

- What has it been like to be interviewed today?
- Has the interview influenced your thoughts in any way?
- What do you think are the risks / benefits of taking part?
- Do you think that research should continue in this area?
- How do you think the information you have shared today should be used?
- If you were offered this opportunity again, would you take part?
- Would you like to be contacted about future research by:
 - PICU
 - Bereavement team
 - Chaplains
- How: phone / letter / email

End Info about support services

APPENDIX 3

Demographic Data Collection

Please circle your answer

Gender Male/Female

Postcode:

Age

- (1) 16-19
- (2) 20-29
- (3) 30-39
- (4) 40-49
- (5) 50-59
- (6) 60-69
- (7) 70 and over

Nationality

- (1) UK, British
- (2) Irish Republic
- (3) India
- (4) Pakistan
- (5) Poland
- (6) Other (Please specify)

Religion

- (1) No religion
- (2) Christian (Church of England, Catholic, Protestant and all other Christian denominations)
- (3) Buddhist
- (4) Hindu
- (5) Jewish
- (6) Muslim
- (7) Sikh
- (8) Any other religion

Marital status

- (1) Single (never married)
- (2) Married and living with your husband/wife
- (3) A civil partner in a legally-recognised Civil Partnership
- (4) Married and separated from your husband/wife
- (5) Divorced
- (6) Widowed

Ethnicity

- (1) White
- (2) Mixed / Multiple ethnic groups
- (3) Asian / Asian British
- (4) Black / African / Caribbean / Black British
- (5) Chinese
- (6) Arab
- (7) Other ethnic group

Education

- (1) Post-graduate – Master’s Degree or PhD
- (2) Degree level
- (3) Diploma in Higher Education
- (4) A-levels or equivalent
- (5) GCSEs or equivalent
- (6) Other (Please specify)

Employment

- (1) Full time - employed
- (2) Part time - employed
- (3) Self-employed FT
- (4) Self-employed PT
- (5) Unemployed
- (6) Other (Please specify)

APPENDIX 4

Interview Distress Protocol

This protocol is for the use of the interviewer if during the interview process the participant should display any signs of increased stress or emotional distress.

Signs of distress	Actions to take	Participant response	Outcome
Verbalised they are getting stressed or emotionally distressed by the interview	<ol style="list-style-type: none">1) Stop the interview2) Allow time for the participant to regroup and offer support3) Assess further with following questions:<ol style="list-style-type: none">a) How are you feeling right now?b) What thoughts are you having?c) Do you feel able to continue with your day? <p>Decide if they are experiencing acute emotional distress beyond what would be normally expected in an interview about a sensitive topic.</p>		
Display behaviours suggesting they are too stressed (crying uncontrollably, struggling to speak clearly)	<ol style="list-style-type: none">1. Stop the interview2. Allow time for the participant to regroup and offer support3. Assess further with following questions:<ol style="list-style-type: none">a) How are you feeling right now?b) What thoughts are you having?c) Do you feel able to continue with your day? <p>Decide if they are experiencing acute emotional distress beyond what would be normally expected in an interview about a sensitive topic.</p>		

Actions:

- If the participant is displaying an emotional response that is thought to be of an expected level in an interview about a sensitive topic, offer support and the opportunity to either stop the interview, have time to regroup, and continue
- If a participant is experiencing **acute emotional distress beyond what would be normally expected in an interview about a sensitive topic, but is not in imminent danger:** encourage the participant to contact their usual source of support. With the participant's permission, contact the PICU Family Liaison Team/BCH Bereavement team/Chaplaincy staff to request some additional support.
- If the participant indicates that **they may harm themselves or others**, call for assistance and either arrange for them to be seen by the on-site clinical psychology team, or for a friend or relative to accompany them to an ED. Contact their GP and an appointment made for follow-up as soon as possible.