Parental experiences of end-of-life decision-making in the Paediatric Intensive Care Unit: A Qualitative Interview Study

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Parental experiences of end-of-life decision-making in the Paediatric Intensive Care Unit: A Qualitative Interview Study

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Abstract

Objectives

To provide an in-depth insight into the experience and perceptions of bereaved parents who have experienced end-of-life care decision-making for children with life-limiting conditions in Paediatric Intensive Care (PIC).

Design

An in-depth qualitative interview study with a sample of parents of children with life-limiting or life-threatening conditions who had died in PIC within the previous 12 months. A thematic analysis was conducted on the interview transcripts.

Setting

A PIC in a large National Health Service (NHS) tertiary children's hospital in the West Midlands, UK.

Participants

17 parents of 11 children who had died in the PIC.

Results

Five interrelated themes were identified:

1. Parents knowledge and previous experiences are relevant to end of life care decision-making
2. The importance of trusted relationships with healthcare professionals in end of life care decision-making
3. Variability in communication with healthcare professionals
4. End-of-life care decision-making and Advance Care Planning are complex processes
5. Experiencing the death of a child in the intensive care unit

Conclusions and Implications

The death of a child is an intensely emotional experience for all involved. This study adds to the limited evidence base related to parental experiences of end-of-life care decision-making and provides findings that have international relevance. Communication around end-of-life decision-making and ACP should be conducted by trusted healthcare professionals. The expertise and previous experience of parents is highly relevant and should be acknowledged. End-of-life care decision-making is a complex and nuanced process; the information needs and preferences of each family are individual and need to be understood by the professionals involved in their care. This may not always include an ability or willingness to discuss discharge from PIC, or an ACP. The findings should help to inform future practice and service design.

Article summary

Strengths and limitations of the study

- Improving end-of-life care and decision-making for children with life-limiting and life-threatening conditions in PIC is a pressing concern.
- This in-depth qualitative interview study provides insights into such decision- making from a parental perspective.
- The study was conducted with parents whose children had died from a range of different conditions.
- The qualitative nature of the study provides detailed, in-depth insights and an understanding of the parental experience of end-of-life care decision-making in PIC; however, recruitment was challenging and the number of participants is relatively small.
- The findings are relevant across a range of healthcare settings as the numbers of children with life-limiting and life-threatening conditions increases and more high profile cases received attention from the media.

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Competing interests

The authors have no competing interests.

Ethical Approval

Ethical approval was granted on 5.1.16 by the East Midlands Research Ethics Committee IRAS No: 184171 REC 15/EM/0539. Amendment 1.0.1 was approved on 31.5.16.

Keywords

Pediatrics

Critical Care

Decision Making

Palliative Care

Advance Care Planning

Introduction

Improvements in survival associated with advances in medical therapy have resulted in increasing numbers of children and young people (hereafter described as children) living with life-limiting and life-threatening conditions (LLC) (1, 2). Uncertainty is part of daily life for many of these children and their families, with a constant risk of sudden and unpredictable deterioration leading to the need for emergency medical care, admission to paediatric intensive care (PIC) and the possibility of dying. Over 50% of children who die in England have a pre-existing LLC (3, 4), and the most frequent place of death is PIC, commonly following the withdrawal of life-sustaining treatments (5, 6). The time spent on PIC before death is increasing (7), reflecting a trend towards longer attempts to sustain life. At times, parents and professionals may disagree about the indication for ongoing life sustaining treatments, as illustrated in several recent high-profile cases (8, 9, 10).

Advance Care Planning (ACP) is a process that may help patients and families achieve a sense of control around their treatment choices (11, 12), and is a core element of national palliative care strategies for both children and adults (13-15). While NICE (National Institute for Health and Care Excellence) has published Quality Standards for end-of-life care in infants, children and young people (14, 15), the current evidence base to inform policy and practice in end-of-life care decision-making for children, including ACP, is scarce (12, 16).

Aim of the study

This study was designed to provide in-depth insights into the experiences and perceptions of parents who have experience of end-of-life care decision-making for children with LLC who have died in PIC.

Patient and Public Involvement (PPI)

Two bereaved parents joined the study team as PPI advisors to ensure that the study design and outputs were relevant to their experience, one of whom has co-authored this paper. PPI was integral to the design of the study, including the wording of participant information sheets and interview schedule. Communication with PPI team members took place throughout the study via email (their expressed preference).

Methods

Study setting

The study setting was PIC at Birmingham Children’s Hospital, a large tertiary referral centre in the West Midlands, UK. The PIC unit has 31 beds and manages approximately 1400 admissions per year.

Recruitment

Participants were recruited purposively; all participants were parents of children with a pre-existing life-limiting condition who had died in PIC. Potential participants (those who were legally the parents or guardians of the child) were identified (by AP and JS) from the PIC mortality database retrospectively for a period of 12 months prior to study commencement and then prospectively over a 12 months period. Exclusion criteria were parents who were unwilling or unable to provide informed consent in English; bereaved parents of a child who had died from acute illness or trauma; and parents aged 16 years or less at the time of recruitment.

Retrospectively, 59 cases of death in PICU were identified as suitable for approach; 58 letters were posted, one was sent by email (as the parents were participating in an email discussion about bereavement follow-up). Of the 59 invitations, there were 11 responses. 8 were positive, and led to interviews. 2 declined to participate without further explanation. 1 letter was returned to sender.

Prospectively, 29 cases were identified and sent letters. The approach for the prospective patients was made at the time of invitation to bereavement follow-up, by letter within six weeks of the child's death.

Explicit decline was only stated in two retrospective replies. No specific reason was stated in any case. If there was no reply to the invitation, non-participation was assumed. The shortest time between bereavement and interview was 7 months (average time 11.8 months, median = 10 months).

Data collection

Interviews

A semi-structured interview schedule was devised to elicit in-depth details and reflections about end-of-life care decision-making in PIC, including experiences and perceptions of ACP (Table 1). Participants were offered a choice of face-to-face or telephone interview. Interviews were conducted with either parent, separately or together, according to their preference.

Table 1: Interview Schedule

Open questions	Prompts
Demographics and Introduction Check understanding of what the interview is about	Do you have any questions?
About child and family Please can you start by telling me about [child's name] and your family?	Were you aware that [child] was unwell before they were born? When was [child] 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 [child's] 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 [child] became poorly and went to hospital and PICU?	What was that like? / How did you feel? Did you have plans or discussions about [child's] 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 [child's] care at this time?
EOLC/PC Decisions, ACP and Planning on PICU When did you realise/understand that [child] was going to die?	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 [child] was on PICU did you / were you offered the chance to complete an Advanced Care Plan?	Did you know what it was? 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 might be the problems with an ACP?

Around Time of Death and Beyond Do you feel able to describe what happened when [child] died?	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

Data Analysis

All interviews were transcribed verbatim. Data was managed using NVivo software. Thematic analysis of transcripts and field notes was carried out using an inductive approach as described by Braun & Clarke (17). This began with familiarisation with the data, reading and re-reading the transcripts, and coding the complete dataset. Three members of the study team independently coded a selection of transcripts (JS, AP and SM). Coding was discussed and compared at intervals, to allow the iterative development of themes and to decrease lone researcher bias (18).

Findings

Study population

Recruitment began following ethical approval in January 2016. 17 parents of 11 children participated in a total of 11 interviews. The sample characteristics are outlined in Table 2.

Table 2: Sample characteristics

Family	Participants (mother / father)	Time since bereavement	Age of child	Child's diagnosis (Together for Short Lives category)(2)
1	M & F	1 year 6 months	3 years	**3**
2	M & F	9 months	6 months	**4**
3	M	5 months	5 months	**3**
4	M & F	8 months	11 years	**1**
5	M & F	1 year 7 months	2 years 11 months	**1**
6	M	1 year 7 months	9 months	**1**
7	M & F	10 months	18 years	1
8	M	1 year 1 month	5 months	**4**

9	M & F	7 months	16 years	4
10	M	1 year 11 months	2 years	**1**
11	M	10 months	1year 11 months	**4**

Themes

Five overarching, interrelated themes were identified:

1. Parents knowledge and previous experiences are relevant to end of life care decision-making
2. The importance of trusted relationships with healthcare professionals in end of life care decision-making
3. Variability in communication with healthcare professionals
4. End of life care decision-making and Advance Care Planning are complex processes
5. Experiencing the death of a child in the intensive care unit

1. Parents knowledge and previous experience must be taken into account in end of life care decisions-making

Parents had intimate knowledge of their child as a person and had significant expertise related to their child's condition. They frequently used medical slang and jargon during the interviews, such as *"her blood pressure was in her boots", "*[oxygen] *sats"* and *"oscillated again"*. Having seen how clinical measurements, blood test results and other investigations were used in medical decision making, they referred numerical measurements to provide some certainty:

> *"towards the end once we'd really got into the grips of treatment, you know, we understood our kids blood work, you know, literally like breathing and everything that's going on… we automatically look at blood work and know what's going on" (Mother 10)*

Almost all parents had experienced several serious episodes of dramatic deterioration before the terminal events culminating in the child's death, during which they had observed and gained detailed knowledge of high intensity, specialised treatments. These experiences shaped and influenced their hopes and expectations:

> *"… about a week before we kept saying [Child010] probably could do with going onto [intervention] but there was another child on [intervention] at that time and … so we were sort of waiting, umming and arring over the [intervention] … I remember [child's father] saying at the time 'but we told you days ago'" (Mother 11)*

Decisions about high intensity treatments could be influenced by a sense that there was "*nothing to lose*" when the alternative was that the child would almost certainly die:

> *"if we leave the child as she is, she's going to pass away anyway. But if we do the, if we put her on the machine there's a chance that she's going to have a problem – said well it's a no brainer, you know. What's the point? You know, if you're going to leave her, she's going to pass away anyway. You might as well on the machine, what have you got to lose?" (Father 9)*

Parents' expertise extended to the ability to recognise that their child was dying when that time came. Many of the parents described recognising that their child was dying before any open conversations had occurred with healthcare professionals about this. Frequently there was a tacit realisation, knowing "*in my heart*", or having a feeling that "*she just wasn't right*". For some, the realisation was associated with seeing their child's increasing dependence on life-sustaining intervention:

> *"Because there was really nothing and there was no improvement whatsoever … The machine got knocked and … it was only for a second, it just jumped in and his blood pressure crashed down low and his heart rate got down very low and, off literally just for a second" (Mother 4)*

For others, it was a realisation that their child's condition was deteriorating despite the treatment they were receiving:

> *"… you then don't necessarily need to be a doctor to understand that you're on a bit of a one-way street" (Father 1)*
>
> *"If I'm honest, the moment they ventilated her, I think deep down we knew she wasn't coming back that time. … I didn't want to say it out loud and I didn't want to admit it, but I think that we all knew" (Mother 10)*

2. The importance of trusted relationships with healthcare professionals in end-of-life care decision-making

Trusted relationships with healthcare professionals (HCPs) were highly valued by parents, and frequently referred to in the interviews. Continuity of care was a key factor underpinning the development of such relationships. Parents often identified, by name, the individuals who were particularly trusted. Such HCPs had advocated for the child or the parents at difficult times and offered extra support, such as providing a personal contact number or going into work on a day off to see the child and family:

> *"So he gave me his mobile number, so 'Just text me or something and I'll make sure that one of my team would come down and see you and then we'll make sure that she's getting the right sort of ….' That was really very kind of the [Doctor] to do that and that made life a lot easier for us, a lot easier for everybody" (Father 9)*

These individuals were particularly important at times when key decisions were being made.

> *"He was very good at explaining things, he was and he would answer any questions … from my perspective when he was telling us 'I'm very sorry. There's nothing else that we can do.' And then it was believable, I didn't feel like we're being fobbed off, or anything like that" (Father 4)*

Parents recognised that these HCPs had their own emotions and stresses to cope with, and empathised with how this affected their work.

> *"the doctor that helped us at the end was lovely. … Couldn't have asked for a better doctor and it turns out that unfortunately five people died on [intensive care] that day. So he had a bad day" (Father 7)*

Relationships with HCPs were fragile, reflecting the anxiety and concern that parents lived with, and trust could be compromised easily. Parents described occasions when they had a feeling that there had been an unexpected complication in their child's care that was not being openly discussed, or when they were provided with conflicting advice:

> *"One would be happy to do something or they'd put him on something. And then another one would come in and say, 'Now take him off that…' So that was where we found it a bit hard -- it's different" (Mother 4)*

These occasions led to suspicion and fear. One mother described how her trust was compromised when she discovered that a meeting had been held about her child's care without her involvement; "*you have broke[n] my trust again completely*" and "*don't lie to me*" (Mother 9). It also happened when parents discovered that an aspect of their child's medical treatment was not being openly discussed with them:

> *"It's that trust relationship, you trust so openly because your consultants, doctors, registrars, nurses, these are like Gods and you're looking at them and thinking come on, I know this happened. It doesn't change what's happened, we know that [complication]'s part and parcel and this is a risk, we know." (Mother 10)*

Trust was also compromised if parents felt that they were not being listed to and so having to repeat their concerns to many members of staff. Staff were perceived to be overstretched and busy, but there were occasions, particularly as a child approached the end-of-life, that a change in staff left parents feeling abandoned:

> *"you're having to explain what [the child's condition] is, and it becomes "groundhog day" that you're doing it over and over again. You think actually -- I may not have the practical skills that the nurse has, but my knowledge of my child and my child's illness is far, it surpasses that. That's a bad position to be in … " (Mother 6)*

3. Variability in communication with healthcare professionals

Parents felt intensely vulnerable as their children approached the end of life, as described in the examples below:

> *"looking back, I realise just how, you know, mentally worn down with an overload of information I was because I remember them asking me to sign the consent form for the [treatment] and I was looking at the form and … my mind had just completely gone … I just couldn't physically remember how to sign my own name" (Mother 10)*

> *"It was such a busy few hours that morning. And the noise was incredible. And it probably wasn't any more noisy than normal, but it felt massively noisy. I just, probably because I was just exhausted and I'd had sleeping tablets and I think I was still recovering from them and I couldn't think straight about anything. … And then the next significant thing was they said 'we need to go and have a chat". (Mother 11)*

Parents described their experiences of receiving information both at the bedside, and during more formal meetings. Both verbal and non-verbal communication made a difference to parents. In terms of verbal communication, parents described how important it was for information to be presented in a clear and sometimes brutally honest fashion. It helped if this information was given by a trusted HCP, but that sometimes depended on the uncertainties of the shift roster:

> *"I always asked her from the beginning if I'm looking at mortality, then tell me. And a few nights before the end, she said, 'You're looking at it.' So yes, but that was more to do with kind of -- I don't know, good luck or whatever you want to put in that. It might not have been her, if it would have been someone else, I wouldn't have had that at all." (Mother 6)*

Clinical uncertainty was a common experience and was particularly confusing and difficult for parents when they were used to making decisions based on precise medical explanations and test results. In this situation, parents hoped for consensus amongst their HCPs:

> *"You couldn't help but feel -- not quite them and us, but was more -- it was case where you thought, 'Are they all on the same page?' Well, they probably were on the same page…. But if they're not, we've got a fight on our hands …" (Father 4)*

Meetings to discuss end-of-life care with the clinical team were challenging experiences for parents. They were frequently outnumbered by an "*overwhelming*" number of staff which they interpreted as an indication of the severity of the situation:

> *"[Child 4] was very popular with the staff in the hospital and so a lot of people took an interest in him. They just wanted to be there at that meeting and we very much appreciate them wanting to do that. But I think it was a bit disconcerting" (Father 4)*

Parents' described a realisation that their child was dying informed by actions rather than explicit communication, such as a move to particular area of PIC or being given priority to use the parent's bedroom:

> *"And we moved over to [letter] side; that was awful. Because there you're surrounded by a lot of other sick children and then you think, 'Well, that must mean [child]'s really sick as well'" (Mother 3)*

> *"And so when she [nurse] came over … I said 'oh I'm sorry, I've left all my stuff in the parents' bedroom, I'll get it out in a sec so you can use it' and she said 'oh no, no, nobody's using it'…. and she said 'nobody's need is greater than yours today' and I thought 'shit'. And just hearing that, I thought 'oh hang on, so you're telling me basically I've got the sickest child on the unit'. And it was a bit of a wake-up call" (Mother 11)*

One parent described a palpable shift in the manner in which HCPs spoke to her as her child was dying:

> *"my relationship with all the consultants on the unit shifted and they all of a sudden became very business-like and very, how I'd seen them with other parents but never with me. It had always been quite a chatty friendship almost and very comfortable with each other, chatting to people. And now they were very matter of fact, very focused and very negative. So Sunday morning we're all stood round the bed and each and every one of them are just looking at [Child] with this grave look on their face and they're clearly all upset, shocked and uncomfortable … So I found them quite difficult and quite brutal really even though I could see why they were doing it……. They were all of a sudden now, a bit like barriers were going up and they were stepping back from, like 'we can't be chatting, we can't be your friend now, we've got a job to do and we need to focus on this'". (Mother 11)*

4. End-of-life care decision-making and Advance Care Planning are complex processes

A range of contrasting emotions, including fear and hope, were described by parents particularly with respect to ACP and decision-making. Such decision-making may feel overwhelmingly difficult, particularly where they were being asked to make decisions about the withdrawal of life-sustaining treatments, and clear guidance of trusted clinicians could be critical. They experienced conflicting emotions related to a hope that their child would not suffer, and simultaneously not wanting their child to die. These emotions created a state of inner conflict and cognitive dissonance, reflected in stress and anxiety.

> *"that decision didn't come about easy. It didn't -- people think oh well, you chose to switch his life support off. Yeah, we did but we also chose to say, we had to choose to say goodbye to him, you know what I mean?" (Father 5)*

> *"I think the hard thing was that, you know, they were kind of, like, 'what do you think?', which is great that they wanted to ask our opinion but, at the time, we were like so overwhelmed. And I remember thinking, why are they asking us! No, I understand it's good to give parents that power but I was like, you know, 'I have no idea, they're the experts'" (Mother 8)*

> *"So whilst I know it comes under sort of end-of-life decision, it didn't feel like we were having to decide whether he'll live or not. It was more about agreeing that it was time to stop, which I think was the right way to do it. All the way through this, we've been led by the medical teams" (Father 4)*

Some parents expressed a desire to know what to expect when it came to their child dying, although they appreciated that this may not be something that other parents would want:

> *"...not knowing what death is and what it's going to look like... when you're seeing it for the first time, when you're kind of dealing with it, both as an experience of death but also as your baby …. I would like to have known that…sorry … Not everybody would ..." (Mother 6)*

Where parents felt they were missing crucial information, they sought it out from other parents:

> *"We did need to know what happened if we switched the machines off. And so I did ask another parent about what happened to their friend when at the hospital, because I wanted to imagine that scenario if it was going to happen …" (Mother 1)*

Not all of the parents were aware of advance care planning (ACP), and many had not experienced this for their child. There were opposing views, with some parents feeling that ACP "*would have been very useful*", and others that a plan which considered the child's death was not acceptable ("*never an option*"). Parents reported that the timing of conversations with respect to ACP was important, but could be particularly difficult where there was clinical uncertainty about the likely outcomes of a treatment or procedure:

> *"We knew that his life would be short … but we never planned for a negative, we always planned for positives. I don't think anybody told us the potential negative of that [intervention] and I don't think they knew the potential negatives that could happen because they weren't expecting that to happen" (Father 7)*

Parents described the need to be in a "place of acceptance" in order for ACP conversations to take place:

> *"I think you have to come to a place yourself to kind of accept what's going to happen. And until you've got to that acceptance, I think it can make people very aggressive. … And for me, I kind of accepted that was it at that point." (Mother 6)*

Parents whose child had had an ACP made practical suggestions related to the information and knowledge that are needed in order to make informed decisions. They felt that it would be easier to fully participate in ACP if there was opportunity to observe and understand the implications of particular interventions, such as ventilation:

> *"it'd be nice to have that little bit of a tick box 'Has the parent seen a ventilator?' I know they've made this decision but you know like when you have at the end, 'cause some just don't want anything, and that's fine and some have just the oxygen. Some want IVs, you know you've got that little paragraph at the bottom, so just underneath it you know, it would be nice 'Have they seen it?' and at least then you'll know. And maybe then that's the time when the consultant will say 'Would you like to see it?' you know just, we can do a little session for you where you can go in and have a look" (Mother 9)*

5. Experiencing the death of a child in the intensive care unit

Being in PIC and having more days of life than might have been possible in other clinical settings was highly valued:

> *"we had [doctor] worked several nights through with her, where he didn't leave her bedside for lines and things like that; of which in hindsight, she was going to go anyway. But by doing that, he gave me an extra couple of days of which, if we'd gone to a hospice you can't do things like that. She would have just gone. So for me, I wouldn't have wanted that" (Mother 6)*

The death of their child was an incredibly painful time for all of the parents. The need to preserve their role as parents, providing love and care for their child, was strongly apparent in their accounts. One parent described how much she valued being given the opportunity to hold her child:

> *"all you can see is that your child is just hooked up to everything possible and they made my day when they lifted her up once she was – because she was relatively flat with all the drugs and what not – and lifted her up so I could actually have a cuddle and put her on me. Oh, even now, I'm so grateful that they did that" (Mother 10)*

There was some comfort in making decisions that they considered to be "*best*" for their child.

> *"As much as it did hurt us to let him go, we were thinking what was best for him to be comfortable and not in pain" (Mother 2)*

Some gave poignant and traumatic accounts of witnessing a resuscitation as their child's terminal event:

> *"And then I think for me the last straw was when the last time they were doing the CPR, is when the guy came with the drill and he started to put a drill in her" (Father 9)*
>
> *"then at some point we knew they'd been working for ages, they were going at it and then I heard one of them crack a rib … we'd obviously heard that and we knew that there's no way her chest was coming back from that, she was struggling already and it's not their fault, it happens, you know, it is what it is" (Mother 10)*

Even in these traumatic circumstances, parents valued the reassurance provided in PIC that all possible treatment options had been explored for the child:

> *"There was nothing that they could do for him and they had made that clear – they'd tried everything. They even tried things they thought weren't going to work" (Mother 2)*
>
> *"By this time I knew a lot of what was goes on in the unit and I knew that [intervention] wasn't a good place to be. I knew that I hadn't seen many kids come off [intervention] and go home but, at the same time, I thought well it's worth a try and I also knew if they didn't think it was worth doing, they wouldn't have done it. It's expensive. And I know you can't put a price on life, can you, but I knew that they wouldn't have tried that unless there was a chance" (Mother 11)*

Discussion

Summary

Until now, there has been little evidence about how parents of children with life-limiting conditions experience PIC as their child approaches end-of-life. This study has identified a number of important themes that affected their experience. Firstly, parents felt that their expert knowledge of their child and the child's condition was not always sufficiently acknowledged or valued, particularly as death was approached. They were often aware that their child may be dying before this was openly acknowledged by their HCPs. Trusted relationships with HCPs were critical to their experience of end-of-life care and making decisions for their child. Every situation was unique and the communication needs of each family highly individual. The idea of an ACP was received positively by some parents, but was completely unacceptable, even in principal, to others.

Strengths and Limitations of the study

The study was conducted with parents whose children had died from a diverse range of life-limiting conditions. However, the number of participants is relatively small, and they were all recruited through the same PICU which may limit the generalisability of the findings. Recruiting to research regarding end-of-life care in children is recognised as being challenging (19). The study's findings are based on retrospective accounts that may have been re-framed over time. We did not capture the experiences and perceptions of families who are currently in the process of making end-of-life care decisions for their children on PIC, or the views of any children or young people regarding their own end-of-life care decision-making or opinions regarding ACP.

While data saturation was reached around the key themes reported here, with interviews conducted later in the study period adding no new insights, it is likely that the parents who felt unable to participate may have had views, experiences and perceptions that were different. There were a number of emerging themes in our data analysis which we are not reported here, including the experience of end-of-life care meetings, the care of siblings, spiritual needs and bereavement care; all are worthy of further research.

Comparison with existing literature

There is a lack of empirical research examining end-of-life care decision-making in PIC, perhaps because admission to PICU does not tend to be explicitly for end-of-life care (20). Our findings contrast with previous research that suggested that HCPs are aware that a child may be dying before this is recognised by their parents (21). However, there are case studies and parental narratives which suggest an earlier recognition amongst parents (22, 23) in the same way as described by some of our parents. It seems that a situation of "mutual pretence" may be reached, where there is unspoken understanding of the situation (24).

Previous studies have described influences on parental decision-making as the child's diagnosis, prognosis and the extent of their pain, discomfort or suffering (21, 25). Our findings indicate that clinical uncertainty, unpredictable outcomes of treatments and the intense emotional burden that can exist for both parents and HCPs add complexity of end-of-life care decision-making in PIC. As medical treatments advance, and information about such treatments becomes ever more available, these complexities are becoming increasingly prominent. In this context, for parents who may already be finding it difficult to understand that their child's condition is incurable (26), conversations about end-of-life care may represent a significant change from previously cure-focussed management plans. Parents do not always need to fully acknowledge their child's situation in order to place emphasis on the relief of suffering (21); a situation which potentially provides the opportunity for conversations about ACP or referral to specialist paediatric palliative care services.

Parents value affirmation in their decision-making from a HCP who is known and trusted and who has witnessed the magnitude of their child's illness (27). Trusted relationships with HCPs were critical to the experience of the parents in this study as they tried to make decisions which were "best" for their child. Continuity of care, and a visible commitment to understanding and addressing the child's and family's end-of-life care needs, was an important factor in achieving such relationships. In keeping with previous studies, there were times when changes in staff or the manner in which staff communicated, could cause parents to feel confused and abandoned (28).

Parents wanted to know that all possible treatments had been tried, and valued the extra days of life provided by the delivery of high intensity treatments in PIC. However, with advances in technology, there is an increasing need for preparing families in children for the time when high intensity treatments may become futile and potentially harmful towards the end of a child's life (29).

Recommendations

Wider recognition of the complex factors that relate to end-of-life care decision-making in PIC, and an organisational commitment to providing a clinical environment in which continuity can be provided to families, could both assist with the implementation of policy guidance related to end-of-life care decision-making.

An important area for research is further investigation into child and family perspectives of ACP, and the impact of earlier integration of palliative care into a child's care. Specialist paediatric palliative care services (SPPC) are inconsistently funded and provided both in the UK and internationally (30); ACP and end-of-life care decision-making therefore depends on the principles of palliative care being practiced amongst the wider workforce. A recent study suggested that children who received SPPC were five times less likely to receive high intensity treatments at the end of their lives (31). Another study examined the potential impact of routine referral to SPPC when a child was commenced on extracorporeal life support in PIC (32). This approach to the introduction of SPPC could be further explored.

There is more work to be done to understand how end-of-life care can be effectively achieved in PIC for individual children and families. Future research into the views of children regarding their own end-of-life care decision-making and ACP, as well as research to further understand the experiences of families who are currently in the process of making end-of-life care decisions for their children would be of value.

Conclusion

Learning from the experiences and perceptions of families is imperative in order to improving practice. This study highlights the need for recognition of parental expertise and experience of PIC, and the critical importance of a trusted relationship between families and their HCPs. Parents are highly vulnerable and may be exhausted, confused and uncertain at the times they are asked to take part in end-of-life care decision-making. The needs of each family in terms of information-giving and involvement in decision-making are highly individual. The parents in this study expressed a preference for end-of-life care discussions to be conducted by a trusted HCP in small meetings. ACP is not well understood by parents, and appears to be more helpful for some than others.

Author Contributions

The study was conceptualised by SM, AP, JC and JD, informed and guided by Patient and Public Involvement including advice from ER. JS conducted the interviews. AP managed the conduct of the study. SM, JS and AP drafted the article with contributions from JC and JD. ER acted as PPI co-author and reviewed the article to ensure relevance to the family situation. JC and JD reviewed the article for intellectual content and edited the final version. All authors reviewed, edited and agreed this version.

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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

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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, it's 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.

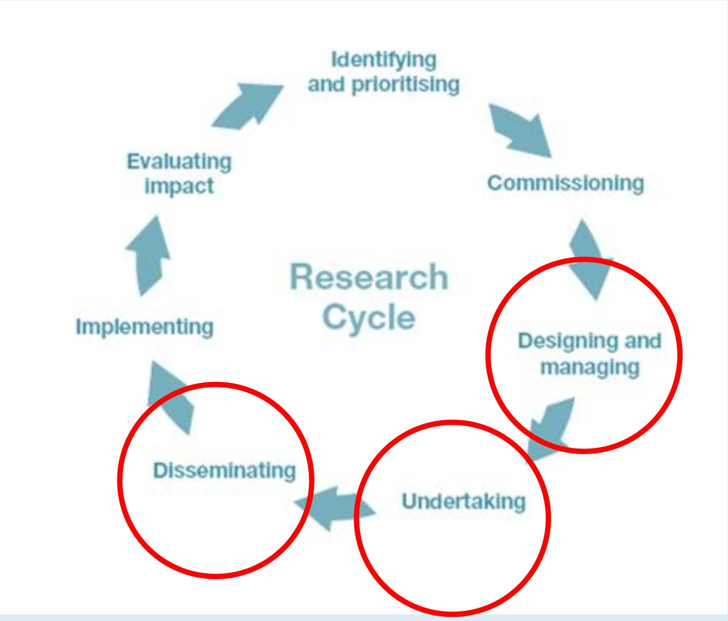
Aim

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.

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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	1) Stop the interview 2) Allow time for the participant to regroup and offer support 3) Assess further with following questions: a) How are you feeling right now? b) What thoughts are you having? c) Do you feel able to continue with your day? Decide if they are experiencing **acute emotional distress beyond what would be normally expected in an interview about a sensitive topic.**		
Display behaviours suggesting they are too stressed (crying uncontrollably, struggling to speak clearly)	1. Stop the interview 2. Allow time for the participant to regroup and offer support 3. Assess further with following questions: a) How are you feeling right now? b) What thoughts are you having? c) Do you feel able to continue with your day? Decide if they are experiencing **acute emotional distress beyond what would be normally expected in an interview about a sensitive topic.**		

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.

Adapted by J. Spry from Drauker, Martsolf & Poole (2009)

Reporting checklist for qualitative study.

Based on the SRQR guidelines.

Instructions to authors

Complete this checklist by entering the page numbers from your manuscript where readers will find each of the items listed below.

Your article may not currently address all the items on the checklist. Please modify your text to include the missing information. If you are certain that an item does not apply, please write "n/a" and provide a short explanation.

Upload your completed checklist as an extra file when you submit to a journal.

In your methods section, say that you used the SRQR reporting guidelines, and cite them as:

O'Brien BC, Harris IB, Beckman TJ, Reed DA, Cook DA. Standards for reporting qualitative research: a synthesis of recommendations. Acad Med. 2014;89(9):1245-1251.

		Reporting Item	Page Number
	#1	Concise description of the nature and topic of the study identifying the study as qualitative or indicating the approach (e.g. ethnography, grounded theory) or data collection methods (e.g. interview, focus group) is recommended	1
	#2	Summary of the key elements of the study using the abstract format of the intended publication; typically	2

		includes background, purpose, methods, results and conclusions	
Problem formulation	#3	Description and signifcance of the problem / phenomenon studied: review of relevant theory and empirical work; problem statement	4
Purpose or research question	#4	Purpose of the study and specific objectives or questions	4
Qualitative approach and research paradigm	#5	Qualitative approach (e.g. ethnography, grounded theory, case study, phenomenolgy, narrative research) and guiding theory if appropriate; identifying the research paradigm (e.g. postpositivist, constructivist / interpretivist) is also recommended; rationale. The rationale should briefly discuss the justification for choosing that theory, approach, method or technique rather than other options available; the assumptions and limitations implicit in those choices and how those choices influence study conclusions and transferability. As appropriate the rationale for several items might be discussed together.	5
Researcher characteristics and reflexivity	#6	Researchers' characteristics that may influence the research, including personal attributes, qualifications / experience, relationship with participants, assumptions and / or presuppositions; potential or actual interaction between researchers' characteristics and the research	5

		questions, approach, methods, results and / or transferability	
Context	#7	Setting / site and salient contextual factors; rationale	5
Sampling strategy	#8	How and why research participants, documents, or events were selected; criteria for deciding when no further sampling was necessary (e.g. sampling saturation); rationale	5
Ethical issues pertaining to human subjects	#9	Documentation of approval by an appropriate ethics review board and participant consent, or explanation for lack thereof; other confidentiality and data security issues	3
Data collection methods	#10	Types of data collected; details of data collection procedures including (as appropriate) start and stop dates of data collection and analysis, iterative process, triangulation of sources / methods, and modification of procedures in response to evolving study findings; rationale	4
Data collection instruments and technologies	#11	Description of instruments (e.g. interview guides, questionnaires) and devices (e.g. audio recorders) used for data collection; if / how the instruments(s) changed over the course of the study	4
Units of study	#12	Number and relevant characteristics of participants, documents, or events included in the study; level of participation (could be reported in results)	5

Data processing	#13	Methods for processing data prior to and during analysis, including transcription, data entry, data management and security, verification of data integrity, data coding, and anonymisation / deidentification of excerpts	4
Data analysis	#14	Process by which inferences, themes, etc. were identified and developed, including the researchers involved in data analysis; usually references a specific paradigm or approach; rationale	4
Techniques to enhance trustworthiness	#15	Techniques to enhance trustworthiness and credibility of data analysis (e.g. member checking, audit trail, triangulation); rationale	5
Syntheses and interpretation	#16	Main findings (e.g. interpretations, inferences, and themes); might include development of a theory or model, or integration with prior research or theory	5-12
Links to empirical data	#17	Evidence (e.g. quotes, field notes, text excerpts, photographs) to substantiate analytic findings	5-12
Intergration with prior work, implications, transferability and contribution(s) to the field	#18	Short summary of main findings; explanation of how findings and conclusions connect to, support, elaborate on, or challenge conclusions of earlier scholarship; discussion of scope of application / generalizability; identification of unique contributions(s) to scholarship in a discipline or field	13

Limitations	#19	Trustworthiness and limitations of findings	`13
Conflicts of interest	#20	Potential sources of influence of perceived influence on study conduct and conclusions; how these were managed	3
Funding	#21	Sources of funding and other support; role of funders in data collection, interpretation and reporting	3

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Parental experiences of end of life decision-making for children with life-limiting conditions in the Paediatric Intensive Care Unit: A Qualitative Interview Study

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Parental experiences of end of life decision-making for children with life-limiting conditions in the Paediatric Intensive Care Unit: A Qualitative Interview Study

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Abstract

Objectives

To provide an in-depth insight into the experience and perceptions of bereaved parents who have experienced end of life care decision-making for children with life-limiting or life-threatening conditions in the Paediatric Intensive Care Unit (PICU).

Design

An in-depth qualitative interview study with a sample of parents of children with life-limiting or life-threatening conditions who had died in PICU within the previous 12 months. A thematic analysis was conducted on the interview transcripts.

Setting

A PICU in a large National Health Service (NHS) tertiary children's hospital in the West Midlands, UK.

Participants

17 parents of 11 children who had died in the PICU.

Results

Five interconnected themes were identified related to end of life care decision making:

1. Parents have significant knowledge and experiences that influence the decision-making process.
2. Trusted relationships with healthcare professionals are key to supporting parents making end of life decisions.
3. Verbal and non-verbal communication with healthcare professionals impacts on the family experience.
4. Engaging with end of life care decision making can be emotionally overwhelming, but becomes possible if parents reach a "place of acceptance".
5. Families perceive benefits to receiving end of life care for their child in a PICU.

Conclusions and Implications

The death of a child is an intensely emotional experience for all involved. This study adds to the limited evidence base related to parental experiences of end of life care decision-making and provides findings that have international relevance, particularly related to place of care and introduction of end of life care discussions. The expertise and previous experience of parents is highly relevant and should be acknowledged. End of life care decision-making is a complex and nuanced process; the information needs and preferences of each family are individual and need to be understood by the professionals involved in their care.

Article summary

Strengths and limitations of the study

- Improving end of life care decision-making for children with life-limiting and life-threatening conditions in PICU is a pressing concern.
- This in-depth qualitative interview study provides insights into such decision-making from a parental perspective.
- The study was conducted with parents whose children had died from a range of different conditions.
- The qualitative nature of the study provides detailed, in-depth insights and an understanding of the parental experience of end of life care decision-making in PICU; however, recruitment was challenging and the number of participants is relatively small.
- The findings are relevant across a range of healthcare settings as the numbers of children with life-limiting and life-threatening conditions increases and more high profile cases received attention from the media.

Funding statement

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Competing interests

The authors have no competing interests.

Ethical Approval

Ethical approval was granted on 5.1.16 by the East Midlands Research Ethics Committee IRAS No: 184171 REC 15/EM/0539. Amendment 1.0.1 was approved on 31.5.16.

Keywords

Pediatrics / Paediatrics

Critical Care

Decision Making

Palliative Care

Advance Care Planning

Data Availability Statement

The research protocol is available as a supplementary file (supplementary file 1)

The data that support the findings of this study are not publicly available due to their containing information that could compromise the privacy of the research participant.

Introduction

Improvements in survival associated with advances in medical therapy have resulted in increasing numbers of children and young people (hereafter described as children) living with life-limiting and life-threatening conditions (1, 2). Uncertainty is part of daily life for many of these children and their families, with a constant risk of sudden and unpredictable deterioration leading to the need for emergency medical care, admission to the paediatric intensive care unit (PICU) and the possibility of dying. Over 50% of children who die in England have a pre-existing life-limiting condition (3, 4), and the most frequent place of death is PICU, commonly following the withdrawal of life-sustaining treatments (5, 6). The time spent on PICU before death is increasing (7), reflecting a trend towards longer attempts to sustain life. At times, parents and professionals may disagree about the indication for ongoing life sustaining treatments, as illustrated in several recent high-profile cases (8, 9).

End of life care decision-making for children is complex. Furthermore, the provision of specialist paediatric palliative care services is currently inconsistent (10). Advance Care Planning (ACP) is advocated as a process that may help patients and families achieve a sense of control around their treatment choices towards the end of their child's life (11, 12). ACP is a core element of national palliative care strategies for both children and adults (13-15), and is included in the NICE (National Institute for Health and Care Excellence) Quality Standards for end of life care in infants, children and young people (14, 15). However, the current evidence base to inform policy and practice in end of life care decision-making for children, including ACP, is scarce (12, 16).

Aim of the study

This study was designed to provide in-depth insights into the experiences and perceptions of parents who had experienced end of life care decision-making for their children with life-limiting or life-threatening conditions in PICU.

Patient and Public Involvement (PPI)

Two bereaved parents joined the study team as PPI advisors to ensure that the study design and outputs were relevant to their experience, one of whom has co-authored this paper. PPI was integral to the design of the study, including the wording of participant information sheets and interview schedule. Communication with PPI team members took place throughout the study via email (their expressed preference).

Methods

The study protocol, which outlines the ethical issues raised by the study and the plans made to address these, is provided as Supplementary File 1.

Study setting

The study setting was PICU at Birmingham Children's Hospital, a large tertiary referral centre in the West Midlands, UK. The PICU has 31 beds and manages approximately 1400 admissions per year. At the time of the study, the hospital palliative care team comprised two specialist nurses and a bereavement team. PICU was supported by a family liaison team. There was no standardised referral process to the palliative care team.

Recruitment

Participants were recruited purposively; all participants were parents of children with a pre-existing life-limiting condition who had died in PICU. Potential participants (those who were legally the parents or guardians of the child) were identified (by AP and JS) from the PICU mortality database retrospectively for a period of 12 months prior to study commencement and then prospectively over a 12 month period. Exclusion criteria were parents who were unwilling or unable to provide informed consent in English; bereaved parents of a child who had died from acute illness or trauma; and parents aged 16 years or less at the time of recruitment.

Retrospectively, 59 cases of death in PICU were identified as suitable for approach; 58 letters were posted, one was sent by email (as the parents were participating in an email discussion about bereavement follow-up). There were 11 responses. Eight were positive, and led to interviews. Two declined to participate without further explanation. One letter was returned to sender.

Prospectively, 29 cases were identified. The prospective approach was made at the time of invitation to bereavement follow-up, by letter, within six weeks of the child's death. Five positive responses were received which led to interviews.

Explicit decline was only stated in two retrospective replies. No specific reason was stated in either case. If there was no reply to the invitation, non-participation was assumed. The shortest time (for both retrospective and prospective approaches) between bereavement and interview was 7 months (average time 11.8 months, median = 10 months).

Data collection

Interviews

A semi-structured interview schedule was devised to elicit in-depth details and reflections about end of life care decision-making in PICU, including experiences and perceptions of ACP (Table 1). This was piloted with the PPI parents. Participants were offered a choice of face-to-face or telephone interview. Interviews were conducted with either parent, separately or together, according to their preference.

Table 1: Interview Schedule

Open questions	Prompts
Demographics and Introduction Check understanding of what the interview is about	Do you have any questions?
About child and family Please can you start by telling me about [child's name] and your family?	Were you aware that [child] was unwell before they were born? When was [child] 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 [child's] 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 [child] became poorly and went to hospital and PICU?	What was that like? / How did you feel? Did you have plans or discussions about [child's] 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 [child's] care at this time?
EOLC/PC Decisions, ACP and Planning on PICU When did you realise/understand that [child] was going to die?	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 [child] was on PICU did you / were you offered the	Did you know what it was? What was the process like? / How did it feel?

chance to complete an Advanced Care Plan?	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 might be the problems with an ACP?
Around Time of Death and Beyond Do you feel able to describe what happened when [child] died?	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

Data Analysis

All interviews were transcribed verbatim. Data was managed using NVivo software. Thematic analysis of transcripts and field notes was carried out using an inductive approach as described by Braun & Clarke (17). This began with familiarisation with the data, reading and re-reading the transcripts, and coding the complete dataset. Three members of the study team independently coded a selection of transcripts (JS, AP and SM). Coding was discussed and compared at regular intervals, to allow the iterative development of themes and to decrease lone researcher bias. The developing themes were reviewed and discussed further with JC and JD. A formal framework was deliberately not applied; the focus of the analysis was firstly on the subjective experiences of the participants, and then on the key interactions with HCPs in relation to end of life care decision making (18).

Findings

Study population

Recruitment began following ethical approval in January 2016. 17 parents of 11 children participated in a total of 11 interviews. The sample characteristics are outlined in Table 2. Together for Short Lives categories are outlined in Table 3.

Table 2: Sample characteristics

Family	Participants (mother / father)	Time since bereavement	Age of child	Child's diagnosis / Together for Short Lives category (2)
1	M & F	1 year 6 months	3 years	3
2	M & F	9 months	6 months	4
3	M	5 months	5 months	3
4	M & F	8 months	11 years	1
5	M & F	1 year 7 months	2 years 11 months	1
6	M	1 year 7 months	9 months	1
7	M & F	10 months	18 years	1
8	M	1 year 1 month	5 months	4
9	M & F	7 months	16 years	4
10	M	1 year 11 months	2 years	1
11	M	10 months	1year 11 months	4

Table 3: Together for Short Lives Categories

Category	Description
1 Life-threatening conditions for which curative treatment may be feasible but can fail	Access to palliative care services may be necessary when treatment fails or during an acute crisis, irrespective of the duration of threat to life. On reaching long-term remission or following successful curative treatment there is no longer a need for palliative care services. Examples: cancer, irreversible organ failures of heart, liver, kidney.
2 Conditions where premature death is inevitable	There may be long periods of intensive treatment aimed at prolonging life and allowing participation in normal activities. Examples: cystic fibrosis, Duchenne muscular dystrophy.
3 Progressive conditions without curative treatment options	Treatment is exclusively palliative and may commonly extend over many years. Examples: batten disease, mucopolysaccharidoses.

4 Irreversible but non-progressive conditions causing severe disability, leading to susceptibility to impaired health	Children can have complex health care needs, a high risk of an unpredictable life-threatening event or episode, health complications and an increased likelihood of premature death. Examples: severe cerebral palsy, multiple disabilities, such as following brain or spinal cord injury.

Themes

Five overarching, interrelated themes were identified:

1. Parents have significant knowledge and experiences that influence the decision-making process
2. Trusted relationships with healthcare professionals are key to supporting parents making end of life decisions vital
3. Verbal and non-verbal communication with healthcare professionals impacts on the family experience
4. Engaging with end of life care decision making can be emotionally overwhelming, but becomes possible if parents reach a "place of acceptance"
5. Families perceive benefits to receiving end of life care for their child in a PICU

1. Parents have significant knowledge and experiences that influence the decision-making process: *"I may not have the practical skills that the nurse has, but my knowledge of my child and my child's illness far surpasses that"*

Parents had intimate knowledge of their child as a person and significant expertise related to their child's condition. They frequently used medical slang and jargon during the interviews, such as *"her blood pressure was in her boots"*, "[oxygen] *sats"* and *"oscillated again"*. Having seen how clinical measurements, blood test results and other investigations were used in medical decision-making, they referred to numerical measurements to provide some certainty:

> *"towards the end once we'd really got into the grips of treatment, you know, we understood our kids blood work, you know, literally like breathing and everything that's going on… we automatically look at blood work and know what's going on" (Mother 10)*

Almost all parents had experienced several serious episodes of dramatic deterioration in their child's condition, during which they had gained detailed knowledge of high intensity, specialised treatments. These experiences shaped and influenced their hopes and expectations for the medical management that would be provided for their child:

> *"… about a week before we kept saying [Child010] probably could do with going onto [intervention] but there was another child on [intervention] at that time and … so we were sort of waiting, umming and arring over the [intervention] … I remember [child's father] saying at the time 'but we told you days ago'" (Mother 11)*

Parental decisions related to their child receiving high intensity treatments could also be influenced by a sense that there was "*nothing to lose*", when the alternative was that their child would almost certainly die:

> *"if we leave the child as she is, she's going to pass away anyway. But if we do the, if we put her on the machine there's a chance that she's going to have a problem – said well it's a no brainer, you know. What's the point? You know, if you're going to leave her, she's going to pass away anyway. You might as well on the machine, what have you got to lose?" (Father 9)*

Clinical uncertainty was a common experience and was particularly confusing and difficult for parents when they were used to making decisions based on precise medical explanations and test results. In this situation, parents hoped for consensus amongst their HCPs:

> *"You couldn't help but feel -- not quite them and us, but was more -- it was case where you thought, 'Are they all on the same page?' Well, they probably were on the same page…. But if they're not, we've got a fight on our hands …" (Father 4)*

Parents' expertise extended to the ability to recognise that their child was dying when that time came. Many of the parents described recognising that their child was dying before any open conversations had occurred with healthcare professionals about this. Frequently there was a tacit realisation, knowing "*in my heart*", or having a feeling that "*she just wasn't right*". For some, the realisation was associated with seeing their child's increasing dependence on life-sustaining interventions:

> *"Because there was really nothing and there was no improvement whatsoever …   The machine got knocked and … it was only for a second, it just jumped in and his blood pressure crashed down low and his heart rate got down very low and, off literally just for a second" (Mother 4)*

For others, it was a realisation that their child's condition was deteriorating despite the treatments that they were receiving:

> *"… you then don't necessarily need to be a doctor to understand that you're on a bit of a one-way street" (Father 1)*

> *"If I'm honest, the moment they ventilated her, I think deep down we knew she wasn't coming back that time. … I didn't want to say it out loud and I didn't want to admit it, but I think that we all knew" (Mother 10)*

2. Trusted relationships with healthcare professionals are key to supporting parents making end of life decisions: *"I've got to admit they went a little bit above and beyond their duties"*

Trusted relationships with healthcare professionals (HCPs) were highly valued. Continuity of care was a key factor underpinning the development of such relationships. Parents often identified, by name, the individuals who they particularly trusted. These were often HCPs from PICU but were also members of other teams involved in their child's care. Such HCPs had usually advocated for the child or the parents at difficult times and offered extra support, such as providing a personal contact number or going into work on a day off to see the child and family:

> *"So he gave me his mobile number, so 'Just text me or something and I'll make sure that one of my team would come down and see you and then we'll make sure that she's getting the right sort of ….' That was really very kind of the [Doctor] to do that and that made life a lot easier for us, a lot easier for everybody" (Father 9)*

These individuals were particularly important at times when key decisions were being made.

> *"He was very good at explaining things, he was and he would answer any questions … from my perspective when he was telling us 'I'm very sorry. There's nothing else that we can do.' And then it was believable, I didn't feel like we're being fobbed off, or anything like that" (Father 4)*

Parents recognised that these HCPs had their own emotions and stresses to cope with, and empathised with how this affected their work.

> *"the doctor that helped us at the end was lovely. … Couldn't have asked for a better doctor and it turns out that unfortunately five people died on [intensive care] that day. So he had a bad day" (Father 7)*

Relationships with HCPs were fragile and trust was easily compromised. Parents described occasions when they were provided with conflicting advice were described as difficult:

> *"One would be happy to do something or they'd put him on something. And then another one would come in and say, 'Now take him off that…' So that was where we found it a bit hard -- it's different" (Mother 4)*
>
> *"it's a great idea to have one consultant that will oversee because there's so many doctors in-out, in-out, in-out, you know, and obviously everybody's got different opinions as to how things should be done. I think for [Child010] she would have really benefited from having one person that had one say" (Mother 10)*

One mother described how her trust was compromised when she discovered that a meeting had been held about her child's care without her involvement; "*you have broke[n] my trust again completely*" and "*don't lie to me*" (Mother 9). It also happened when parents discovered that an aspect of their child's medical treatment was not being openly discussed with them:

> *"It's that trust relationship, you trust so openly because your consultants, doctors, registrars, nurses, these are like Gods and you're looking at them and thinking come on, I know this happened. It doesn't change what's happened, we know that [complication]'s part and parcel and this is a risk, we know." (Mother 10)*

Trust was also compromised if parents felt that they were not being listened to or felt that they had to repeat their concerns over and over to many members of staff:

> *"you're having to explain what [the child's condition] is, and it becomes "groundhog day" that you're doing it over and over again"(mother 6).*

3. Verbal and non-verbal communication with healthcare professionals impacts on the family experience: "*somebody needs to sit with you and explain why you can't have this or why you can't have that*"

Parents described their experiences of receiving information both at the bedside, and during more formal meetings. Both verbal and non-verbal communication made a difference to parents. In terms of verbal communication, parents described how important it was for information to be presented in a clear and sometimes brutally honest fashion. It helped if this information was given by a trusted HCP, but that sometimes depended on the uncertainties of the shift roster:

> *"I always asked her from the beginning if I'm looking at mortality, then tell me. And a few nights before the end, she said, 'You're looking at it.' So yes, but that was more to do with kind of -- I don't know, good luck or whatever you want to put in that. It might not have been her, if it would have been someone else, I wouldn't have had that at all." (Mother 6)*

Meetings to discuss end of life care with the clinical team were challenging experiences for parents. They were frequently outnumbered by an "*overwhelming*" number of staff which they interpreted as an indication of the severity of the situation:

> *"[Child 4] was very popular with the staff in the hospital and so a lot of people took an interest in him. They just wanted to be there at that meeting and we very much appreciate them wanting to do that. But I think it was a bit disconcerting" (Father 4)*

Parents' described a realisation that their child was dying informed by actions rather than explicit communication, such as a move to particular area of PIC or being given priority to use the parent's bedroom:

> *"And we moved over to [letter] side; that was awful. Because there you're surrounded by a lot of other sick children and then you think, 'Well, that must mean [child]'s really sick as well'" (Mother 3)*

> *"And so when she [nurse] came over … I said 'oh I'm sorry, I've left all my stuff in the parents' bedroom, I'll get it out in a sec so you can use it' and she said 'oh no, no, nobody's using it'…. and she said 'nobody's need is greater than yours today' and I thought 'shit'. And just hearing that, I thought 'oh hang on, so you're telling me basically I've got the sickest child on the unit'. And it was a bit of a wake-up call" (Mother 11)*

One parent described a palpable shift in the manner in which HCPs spoke to her as her child was dying:

> *"my relationship with all the consultants on the unit shifted and they all of a sudden became very business-like and very, how I'd seen them with other parents but never with me. It had always been quite a chatty friendship almost and very comfortable with each other, chatting to people. And now they were very matter of fact, very focused and very negative. So Sunday morning we're all stood round the bed and each and every one of them are just looking at [Child] with this grave look on their face and they're clearly all upset, shocked and uncomfortable … So I found them quite difficult and quite brutal really even though I could see why they were doing it……. They were all of a sudden now, a bit like barriers were going up and they were stepping back from, like 'we can't be chatting, we can't be your friend now, we've got a job to do and we need to focus on this'". (Mother 11)*

4. Engaging with advance care planning can be emotionally overwhelming, but becomes possible if parents reach a "place of acceptance"

Parents experienced wide-ranging, intense emotions towards the end of their child's life which impacted on their ability to take part in end of life care decision-making. They described a range of conflicting emotions related to a hope that their child would not suffer, and a simultaneous fear that their child was going to die. These emotions created stress, anxiety, a state of inner conflict and cognitive dissonance. Parents felt intensely vulnerable as their children approached the end of life. They described feeling *"mentally worn down*" and "*not being able to think straight*", as below:

> *"looking back, I realise just how, you know, mentally worn down with an overload of information I was because I remember them asking me to sign the consent form for the [treatment] and I was looking at the form and … my mind had just completely gone … I just couldn't physically remember how to sign my own name" (Mother 10)*

> *"It was such a busy few hours that morning. And the noise was incredible. And it probably wasn't any more noisy than normal, but it felt massively noisy. I just, probably because I was just exhausted and I'd had sleeping tablets and I think I was still recovering from them and I couldn't think straight about anything. … And then the next significant thing was they said 'we need to go and have a chat". (Mother 11)*

In these contexts, end of life care decision-making could feel overwhelmingly difficult for parents, particularly when they were being asked to make decisions about the withdrawal of life-sustaining treatments:

> *"I think the hard thing was that, you know, they were kind of, like, 'what do you think?', which is great that they wanted to ask our opinion but, at the time, we were like so overwhelmed. And I remember thinking, why are they asking us! No, I understand it's good to give parents that power but I was like, you know, 'I have no idea, they're the experts'" (Mother 8)*

Clear guidance and the support of trusted clinicians was critical. The manner in which they were engaged in the decision-making process was important, for example feeling that they have made a choice to "*say goodbye*" rather than having to make a choice to withdraw life-sustaining treatments.

> *"that decision didn't come about easy. It didn't -- people think oh well, you chose to switch his life support off. Yeah, we did but we also chose to say, we had to choose to say goodbye to him, you know what I mean?" (Father 5)*

> *"So whilst I know it comes under sort of end of life decision, it didn't feel like we were having to decide whether he'll live or not. It was more about agreeing that it was time to stop, which I think was the right way to do it. All the way through this, we've been led by the medical teams" (Father 4)*

There was some comfort in making decisions that they considered to be "*best*" for their child.

> *"As much as it did hurt us to let him go, we were thinking what was best for him to be comfortable and not in pain" (Mother 2)*

The need to preserve their role as parents, providing love and care for their child, was strongly apparent in their accounts. One parent described how much she valued being given the opportunity to hold her child:

> *"all you can see is that your child is just hooked up to everything possible and they made my day when they lifted her up once she was – because she was relatively flat with all the drugs and what not – and lifted her up so I could actually have a cuddle and put her on me. Oh, even now, I'm so grateful that they did that" (Mother 10)*

Some parents expressed a desire to know what to expect when it came to their child dying, although they appreciated that this may not be something that other parents would want:

> *"...not knowing what death is and what it's going to look like... when you're seeing it for the first time, when you're kind of dealing with it, both as an experience of death but also as your baby …. I would like to have known that…sorry … Not everybody would …" (Mother 6)*

Where parents felt they were missing crucial information, they sought it out from other parents:

> *"We did need to know what happened if we switched the machines off. And so I did ask another parent about what happened to their friend when at the hospital, because I wanted to imagine that scenario if it was going to happen …" (Mother 1)*

Not all of the parents were aware of advance care planning (ACP), and many had not experienced this for their child. There were opposing views, with some parents feeling that ACP "*would have been very useful*", and others that a plan which considered the child's death was not acceptable; "*never an option*". Parents reported that the timing of conversations with respect to ACP was important, but could be particularly difficult where there was uncertainty about the likely outcome of a treatment or procedure, such as surgery or a new medical intervention:

> *"We knew that his life would be short … but we never planned for a negative, we always planned for positives. I don't think anybody told us the potential negative of that [intervention] and I don't think they knew the potential negatives that could happen because they weren't expecting that to happen" (Father 7)*

Parents described the need to be in a "place of acceptance" in order for ACP conversations to take place:

> *"I think you have to come to a place yourself to kind of accept what's going to happen. And until you've got to that acceptance, I think it can make people very aggressive. … And for me, I kind of accepted that was it at that point." (Mother 6)*

Parents who had made a formal ACP for their child made practical suggestions related to the information and knowledge that should be considered for parents in order for them to make an informed decision about whether to create an ACP. They suggested that it was necessary to observe and understand the implications of particular interventions, such as ventilation, before considering this in an ACP:

> *"it'd be nice to have that little bit of a tick box 'Has the parent seen a ventilator?' I know they've made this decision but you know like when you have at the end, 'cause some just don't want anything, and that's fine and some have just the oxygen. Some want IVs, you know you've got that little paragraph at the bottom, so just underneath it you know, it would be nice 'Have they seen it?' and at least then you'll know. And maybe then that's the time when the consultant will say 'Would you like to see it?' you know just, we can do a little session for you where you can go in and have a look" (Mother 9)*

5. Families perceive benefits to receiving end of life care for their child in a PIC: *"the support that we received on [PICU] was just amazing"*

The parents all provided poignant accounts of their child's death. Some were shocking and traumatic, for example when they witnessed resuscitation as their child's terminal event:

> *"And then I think for me the last straw was when the last time they were doing the [cardio-pulmonary resuscitation] CPR, is when the guy came with the drill and he started to put a drill in her" (Father 9)*
>
> *"then at some point we knew they'd been working for ages, they were going at it and then I heard one of them crack a rib … we'd obviously heard that and we knew that there's no way her chest was coming back from that, she was struggling already and it's not their fault, it happens, you know, it is what it is" (Mother 10)*

However, even in these traumatic circumstances, parents valued the reassurance provided in PICU that all possible treatment options had been explored for their child:

> *"There was nothing that they could do for him and they had made that clear – they'd tried everything. They even tried things they thought weren't going to work" (Mother 2)*
>
> *"By this time I knew a lot of what was goes on in the unit and I knew that [intervention] wasn't a good place to be. I knew that I hadn't seen many kids come off [intervention] and go home but, at the same time, I thought well it's worth a try and I also knew if they didn't think it was worth doing, they wouldn't have done it. It's expensive. And I know you can't put a price on life, can you, but I knew that they wouldn't have tried that unless there was a chance" (Mother 11)*

Being in PICU and having more days of life than might have been possible in other clinical settings was also highly valued:

> *"we had [doctor] worked several nights through with her, where he didn't leave her bedside for lines and things like that; of which in hindsight, she was going to go anyway. But by doing that, he gave me an extra couple of days of which, if we'd gone to a hospice you can't do things like that. She would have just gone. So for me, I wouldn't have wanted that" (Mother 6)*

Discussion

Summary

This study provides important insights into the experiences of end of life care decision-making of parents of children with life-limiting and life-threatening conditions who die in a PICU. The study has identified important themes that affected their experience. The parents who participated in this study had expert knowledge of their child and their child's condition, and wanted this to be taken into account in medical decision-making. Parents were often aware that their child may be dying before this was openly acknowledged by their HCPs. Trusted relationships with HCPs were critical to their experience of end of life care decision-making, as were both verbal and non-verbal communication. Every situation was unique; making decisions about care at the end of a child's life was described as "overwhelming" by some. The parents in this study expressed a preference for end of life care discussions to be conducted by a trusted HCP in small meetings.

Parents described the need to have come to a "place of acceptance" in order to be able to take part in end of life care decisions. The idea of an ACP was received positively by some parents, but was completely unacceptable, even in principal, to others. The end of life was traumatic for parents to witness if associated with attempts at resuscitations and invasive medical procedures, however the continuity of care provided in PICU, reassurance that all possible treatment options had been tried for their child, and the extra hours of life that could be provided were all perceived as important benefits by parents.

Strengths and Limitations of the study

Recruiting to research about end of life care in children is known to be challenging (19). The study was conducted with parents whose children had died from a diverse range of life-limiting conditions. However, the number of participants is relatively small, and they were all recruited through the same PICU which may limit the generalisability of the findings. While data saturation was reached around the key themes reported here, it is likely that the parents who felt unable to participate may have had views, experiences and perceptions that were different. There were several emerging themes in our data analysis which are not reported here, including the experience of end of life care meetings, the care of siblings, spiritual needs and bereavement care; all are worthy of further research. Furthermore, the study's findings are based on retrospective accounts that may have been re-framed over time. We did not capture the experiences and perceptions of families who are currently in the process of making end of life care decisions for their children, or the views of any children or young people regarding their own end of life care decision-making.

Comparison with existing literature

There is a lack of empirical research examining end of life care decision-making in PICU, perhaps because admission to PICU does not tend to be explicitly for end of life care (20). Our findings contrast with previous research that suggested an awareness amongst HCPs that a child may be dying before this is recognised by their parents (21). In keeping with our finding that parents often had an unspoken knowledge that their child was dying, there are published case studies and parental narratives which also suggest an earlier recognition amongst parents (22, 23). A situation of "mutual pretence" may be reached between parents and HCPs, where this knowledge is unspoken; this situation of mutual pretence could potentially be acknowledged more openly in order to start discussions about end of life care (24).

Previous studies have described influences on parental decision-making as the child's diagnosis, prognosis and the extent of their pain, discomfort or suffering (21, 25). Our findings indicate that clinical uncertainty, unpredictable outcomes of treatments, a sense of loss of control, and the intense emotional burden that can exist for both parents and HCPs add complexity of end of life care decision-making in PICU. As medical treatments advance, and information about such treatments becomes more available particularly in media reports, these clinical and ethical complexities are becoming ever more prominent. In this context, for parents who may already be finding it difficult to understand that their child's condition is incurable (26), conversations about end of life care may represent a significant change from previously cure-focussed management plans. Previous research has suggested that parents do not always need to fully acknowledge their child's situation in order to place emphasis on the relief of suffering (21). It may therefore be possible to reach a situation earlier on in a child's illness which provides the opportunity for conversations about end of life care, ACP or referral to specialist paediatric palliative care services, through skilful acknowledgement of uncertainty and the conflicting emotions that parents may be experiencing, including fear and hope.

Parents value affirmation in their decision-making from a HCP who is known and trusted and who has witnessed the magnitude of their child's illness (27). Trusted relationships with HCPs were critical to the experience of the parents in this study as they tried to make decisions which were "best" for their child. Continuity of care, and a visible commitment to understanding and addressing the end of life care needs of the child and their family, were important in achieving such relationships. In keeping with previous studies, there were times when conflicting advice brought about through changes in staff could cause parents to feel confused and abandoned (28). Provision of a working environment that allows for continuity of care warrants attention, as does care for the workforce as they face these emotionally charged, ethically challenging situations with children and their families.

Parents were reassured by the knowledge that all possible treatment options had been explored for their child, and valued the extra days of life provided by the delivery of high intensity treatments in PICU. Given the rising numbers of children with life-limiting and life-threatening conditions and the complexity of their needs, there is a need to consider how this care and associated reassurances can be offered to children in environments other than PICU, including high dependency units, and children's hospices. There is a careful and highly individual balance to be found for each family between the knowledge that everything possible has been tried for their child, alongside preparation for the time when high intensity treatments may become futile and potentially harmful towards the end of a child's life (29).

Recommendations

Wider recognition of the complex factors that relate to end of life care decision-making in PICU, and an organisational commitment to providing a clinical environment in which continuity can be provided to families, could both assist with the implementation of policy guidance related to end of life care decision-making.

An important area for research is further investigation into the child and family perspectives of ACP, and the impact of earlier integration of palliative care into a child's care. A recent study suggested that children who received SPPC were five times less likely to receive high intensity treatments at the end of their lives (30). Another study examined the potential impact of routine referral to SPPC when a child was commenced on extracorporeal life support in PICU (31). This approach to the introduction of SPPC could be further explored. However, SPPC services are inconsistently funded and provided both in the UK and internationally (10); ACP and end of life care decision-making therefore depends on the principles of palliative care being practiced amongst the wider workforce.

There is more work to be done to understand how end of life care can be effectively achieved in PICU for individual children and families. Future research into the views of children regarding their own end of life care decision-making and ACP, as well as research to further understand the experiences of families who are currently in the process of making end of life care decisions for their children would be of value.

Conclusion

Learning from the experiences and perceptions of families should inform improved policy and practice. This study highlights the need for recognition of parental expertise and experience, and the critical importance of a trusted relationship between families and their HCPs, which can often be established through repeated admissions or prolonged stays in PICU. Whilst trust is vital to the relationships between families and HCPs, it is also fragile, and can be easily lost. Parents are highly vulnerable and may be exhausted, confused and uncertain at the times when they are asked to engage with end of life care decision-making. Earlier acknowledgement of clinical uncertainty and the conflicting emotions that parents may be experiencing could help in earlier discussions about end of life care and introduction to specialist paediatric palliative care services, where available.

The needs of each family and their readiness for involvement in decision-making is highly individual. ACP is not well understood by parents, and appears to be more helpful for some than others. End of life care of a child on PICU provides potential benefits from a family perspective, and there is a need to consider how the care and reassurances they perceive can be provided in other environments.

There is also a pressing need for greater understanding of the child's experience, which should be the focus of further research.

Author Contributions

The study was conceptualised by SM, AP, JC and JD, informed and guided by Patient and Public Involvement, with specific guidance and advice provided by EH. JS conducted the interviews. AP managed the conduct of the study. SM, JS and AP drafted the article with contributions from JC and JD. EH acted as PPI co-author and reviewed the article to ensure relevance to the family situation. JC and JD reviewed the article for intellectual content and edited the final version. All authors reviewed, edited and agreed this version.

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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

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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, it's 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.

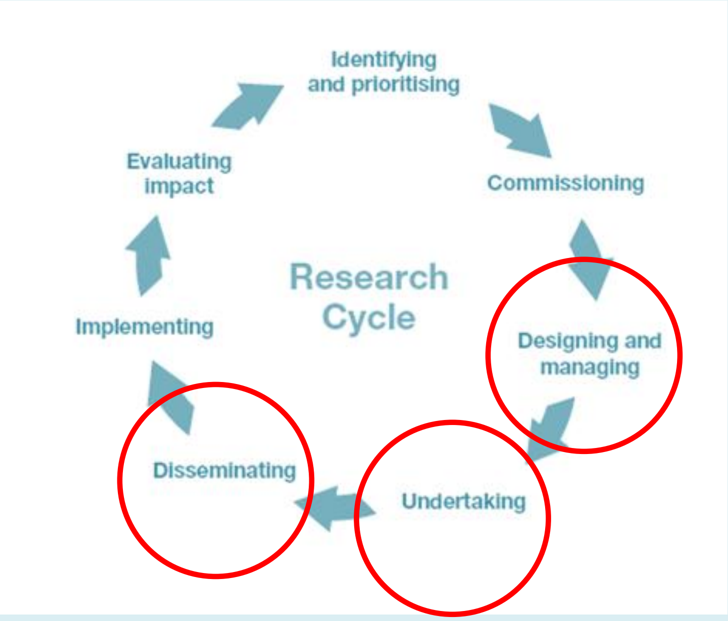
Aim

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.

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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	1) Stop the interview 2) Allow time for the participant to regroup and offer support 3) Assess further with following questions: a) How are you feeling right now? b) What thoughts are you having? c) Do you feel able to continue with your day? Decide if they are experiencing **acute emotional distress beyond what would be normally expected in an interview about a sensitive topic.**		
Display behaviours suggesting they are too stressed (crying uncontrollably, struggling to speak clearly)	1. Stop the interview 2. Allow time for the participant to regroup and offer support 3. Assess further with following questions: a) How are you feeling right now? b) What thoughts are you having? c) Do you feel able to continue with your day? Decide if they are experiencing **acute emotional distress beyond what would be normally expected in an interview about a sensitive topic.**		

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.

Adapted by J. Spry from Drauker, Martsolf & Poole (2009)

Reporting checklist for qualitative study.

Based on the SRQR guidelines.

Instructions to authors

Complete this checklist by entering the page numbers from your manuscript where readers will find each of the items listed below.

Your article may not currently address all the items on the checklist. Please modify your text to include the missing information. If you are certain that an item does not apply, please write "n/a" and provide a short explanation.

Upload your completed checklist as an extra file when you submit to a journal.

In your methods section, say that you used the SRQR reporting guidelines, and cite them as:

O'Brien BC, Harris IB, Beckman TJ, Reed DA, Cook DA. Standards for reporting qualitative research: a synthesis of recommendations. Acad Med. 2014;89(9):1245-1251.

		Reporting Item	Page Number
	#1	Concise description of the nature and topic of the study identifying the study as qualitative or indicating the approach (e.g. ethnography, grounded theory) or data collection methods (e.g. interview, focus group) is recommended	1
	#2	Summary of the key elements of the study using the abstract format of the intended publication; typically	2

		includes background, purpose, methods, results and conclusions	
Problem formulation	#3	Description and signifcance of the problem / phenomenon studied: review of relevant theory and empirical work; problem statement	4
Purpose or research question	#4	Purpose of the study and specific objectives or questions	4
Qualitative approach and research paradigm	#5	Qualitative approach (e.g. ethnography, grounded theory, case study, phenomenolgy, narrative research) and guiding theory if appropriate; identifying the research paradigm (e.g. postpositivist, constructivist / interpretivist) is also recommended; rationale. The rationale should briefly discuss the justification for choosing that theory, approach, method or technique rather than other options available; the assumptions and limitations implicit in those choices and how those choices influence study conclusions and transferability. As appropriate the rationale for several items might be discussed together.	5
Researcher characteristics and reflexivity	#6	Researchers' characteristics that may influence the research, including personal attributes, qualifications / experience, relationship with participants, assumptions and / or presuppositions; potential or actual interaction between researchers' characteristics and the research	5

		questions, approach, methods, results and / or transferability	
Context	#7	Setting / site and salient contextual factors; rationale	5
Sampling strategy	#8	How and why research participants, documents, or events were selected; criteria for deciding when no further sampling was necessary (e.g. sampling saturation); rationale	5
Ethical issues pertaining to human subjects	#9	Documentation of approval by an appropriate ethics review board and participant consent, or explanation for lack thereof; other confidentiality and data security issues	3
Data collection methods	#10	Types of data collected; details of data collection procedures including (as appropriate) start and stop dates of data collection and analysis, iterative process, triangulation of sources / methods, and modification of procedures in response to evolving study findings; rationale	4
Data collection instruments and technologies	#11	Description of instruments (e.g. interview guides, questionnaires) and devices (e.g. audio recorders) used for data collection; if / how the instruments(s) changed over the course of the study	4
Units of study	#12	Number and relevant characteristics of participants, documents, or events included in the study; level of participation (could be reported in results)	5

Data processing	#13	Methods for processing data prior to and during analysis, including transcription, data entry, data management and security, verification of data integrity, data coding, and anonymisation / deidentification of excerpts	4
Data analysis	#14	Process by which inferences, themes, etc. were identified and developed, including the researchers involved in data analysis; usually references a specific paradigm or approach; rationale	4
Techniques to enhance trustworthiness	#15	Techniques to enhance trustworthiness and credibility of data analysis (e.g. member checking, audit trail, triangulation); rationale	5
Syntheses and interpretation	#16	Main findings (e.g. interpretations, inferences, and themes); might include development of a theory or model, or integration with prior research or theory	5-12
Links to empirical data	#17	Evidence (e.g. quotes, field notes, text excerpts, photographs) to substantiate analytic findings	5-12
Intergration with prior work, implications, transferability and contribution(s) to the field	#18	Short summary of main findings; explanation of how findings and conclusions connect to, support, elaborate on, or challenge conclusions of earlier scholarship; discussion of scope of application / generalizability; identification of unique contributions(s) to scholarship in a discipline or field	13

Limitations	#19	Trustworthiness and limitations of findings	`13
Conflicts of interest	#20	Potential sources of influence of perceived influence on study conduct and conclusions; how these were managed	3
Funding	#21	Sources of funding and other support; role of funders in data collection, interpretation and reporting	3

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Parental experiences of end of life decision-making for children with life-limiting conditions in the Paediatric Intensive Care Unit: A Qualitative Interview Study

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Parental experiences of end of life decision-making for children with life-limiting conditions in the Paediatric Intensive Care Unit: A Qualitative Interview Study

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Abstract

Objectives

To provide an in-depth insight into the experience and perceptions of bereaved parents who have experienced end of life care decision-making for children with life-limiting or life-threatening conditions in the Paediatric Intensive Care Unit (PICU).

Design

An in-depth qualitative interview study with a sample of parents of children with life-limiting or life-threatening conditions who had died in PICU within the previous 12 months. A thematic analysis was conducted on the interview transcripts.

Setting

A PICU in a large National Health Service (NHS) tertiary children's hospital in the West Midlands, UK.

Participants

17 parents of 11 children who had died in the PICU.

Results

Five interconnected themes were identified related to end of life care decision making:

1. Parents have significant knowledge and experiences that influence the decision-making process.
2. Trusted relationships with healthcare professionals are key to supporting parents making end of life decisions.
3. Verbal and non-verbal communication with healthcare professionals impacts on the family experience.
4. Engaging with end of life care decision making can be emotionally overwhelming, but becomes possible if parents reach a "place of acceptance".
5. Families perceive benefits to receiving end of life care for their child in a PICU.

Conclusions and Implications

The death of a child is an intensely emotional experience for all involved. This study adds to the limited evidence base related to parental experiences of end of life care decision-making and provides findings that have international relevance, particularly related to place of care and introduction of end of life care discussions. The expertise and previous experience of parents is highly relevant and should be acknowledged. End of life care decision-making is a complex and nuanced process; the information needs and preferences of each family are individual and need to be understood by the professionals involved in their care.

Article summary

Strengths and limitations of the study

- Improving end of life care decision-making for children with life-limiting and life-threatening conditions in PICU is a pressing concern.
- This in-depth qualitative interview study provides insights into such decision-making from a parental perspective.
- The study was conducted with parents whose children had died from a range of different conditions.
- The qualitative nature of the study provides detailed, in-depth insights and an understanding of the parental experience of end of life care decision-making in PICU; however, recruitment was challenging and the number of participants is relatively small.
- The findings are relevant across a range of healthcare settings as the numbers of children with life-limiting and life-threatening conditions increases and more high profile cases received attention from the media.

Funding statement

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Competing interests

The authors have no competing interests.

Ethical Approval

Ethical approval was granted on 5.1.16 by the East Midlands Research Ethics Committee IRAS No: 184171 REC 15/EM/0539. Amendment 1.0.1 was approved on 31.5.16.

Keywords

Pediatrics / Paediatrics

Critical Care

Decision Making

Palliative Care

Advance Care Planning

Data Availability Statement

The research protocol is available as a supplementary file (supplementary file 1)

The data that support the findings of this study are not publicly available due to their containing information that could compromise the privacy of the research participant.

Introduction

Improvements in survival associated with advances in medical therapy have resulted in increasing numbers of children and young people (hereafter described as children) living with life-limiting and life-threatening conditions (1, 2). Uncertainty is part of daily life for many of these children and their families, with a constant risk of sudden and unpredictable deterioration leading to the need for emergency medical care, admission to the paediatric intensive care unit (PICU) and the possibility of dying. Over 50% of children who die in England have a pre-existing life-limiting condition (3, 4), and the most frequent place of death is PICU, commonly following the withdrawal of life-sustaining treatments (5, 6). The time spent on PICU before death is increasing (7), reflecting a trend towards longer attempts to sustain life. At times, parents and professionals may disagree about the indication for ongoing life sustaining treatments, as illustrated in several recent high-profile cases (8, 9).

End of life care decision-making for children is complex. Furthermore, the provision of specialist paediatric palliative care services is currently inconsistent (10). Advance Care Planning (ACP) is advocated as a process that may help patients and families achieve a sense of control around their treatment choices towards the end of their child's life (11, 12). ACP is a core element of national palliative care strategies for both children and adults (13-15), and is included in the NICE (National Institute for Health and Care Excellence) Quality Standards for end of life care in infants, children and young people (14, 15). However, the current evidence base to inform policy and practice in end of life care decision-making for children, including ACP, is scarce (12, 16).

Aim of the study

This study was designed to provide in-depth insights into the experiences and perceptions of parents who had experienced end of life care decision-making for their children with life-limiting or life-threatening conditions in PICU.

Patient and Public Involvement (PPI)

Two bereaved parents joined the study team as PPI advisors to ensure that the study design and outputs were relevant to their experience, one of whom has co-authored this paper. PPI was integral to the design of the study, including the wording of participant information sheets and interview schedule. Communication with PPI team members took place throughout the study via email (their expressed preference).

Methods

The study protocol, which outlines the ethical issues raised by the study and the plans made to address these, is provided as Supplementary File 1.

Study setting

The study setting was PICU at Birmingham Children's Hospital, a large tertiary referral centre in the West Midlands, UK. The PICU has 31 beds and manages approximately 1400 admissions per year. At the time of the study, the hospital palliative care team comprised two specialist nurses and a bereavement team. PICU was supported by a family liaison team. There was no standardised referral process to the palliative care team.

Recruitment

Participants were recruited purposively; all participants were bereaved parents of children with a pre-existing life-limiting condition who had died in PICU. Potential participants (those who were legally the parents or guardians of the child) were identified (by AP and JS) from the PICU mortality database retrospectively for a period of 12 months prior to study commencement and then prospectively over a 12 month period. Exclusion criteria were parents who were unwilling or unable to provide informed consent in English; bereaved parents of a child who had died from acute illness or trauma; and parents aged 16 years or less at the time of recruitment.

Retrospectively, 59 cases of death in PICU were identified as suitable for approach; 58 letters were posted, one was sent by email (as the parents were participating in an email discussion about bereavement follow-up). There were 11 responses. Eight were positive, and led to interviews. Two declined to participate without further explanation. One letter was returned to sender.

Over the 12 month study period, 29 deaths were identified. The approach to the parents of these children was made at the time of invitation to bereavement follow-up, by letter, within six weeks of the child's death. Five positive responses were received which led to interviews.

Explicit decline was only stated in two retrospective replies. No specific reason was stated in either case. If there was no reply to the invitation, non-participation was assumed. The shortest time (for both retrospective and prospective approaches) between bereavement and interview was 7 months (average time 11.8 months, median = 10 months).

Data collection

Interviews

A semi-structured interview schedule was devised to elicit in-depth details and reflections about end of life care decision-making in PICU, including experiences and perceptions of ACP (Table 1). This was piloted with the PPI parents. Participants were offered a choice of face-to-face or telephone interview. Interviews were conducted with either parent, separately or together, according to their preference. A distress protocol was developed for the interviews (see Supplementary file 1: Research protocol) and all participants had access to support from the hospital bereavement team.

Table 1: Interview Schedule

Open questions	Prompts
Demographics and Introduction Check understanding of what the interview is about	Do you have any questions?
About child and family Please can you start by telling me about [child's name] and your family?	Were you aware that [child] was unwell before they were born? When was [child] 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 [child's] 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 [child] became poorly and went to hospital and PICU?	What was that like? / How did you feel? Did you have plans or discussions about [child's] 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 [child's] care at this time?
EOLC/PC Decisions, ACP and Planning on PICU When did you realise/understand that [child] was going to die?	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	Did you know what it was?

When [child] was on PICU did you / were you offered the chance to complete an Advanced Care Plan?	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 might be the problems with an ACP?
Around Time of Death and Beyond Do you feel able to describe what happened when [child] died?	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

Data Analysis

All interviews were transcribed verbatim. Data was managed using NVivo software. Thematic analysis of transcripts and field notes was carried out using an inductive approach as described by Braun & Clarke (17). This began with familiarisation with the data, reading and re-reading the transcripts, and coding the complete dataset. Three members of the study team independently coded a selection of transcripts (JS, AP and SM). Coding was discussed and compared at regular intervals, to allow the iterative development of themes and to decrease lone researcher bias. The developing themes were reviewed and discussed further with JC and JD. A formal framework was deliberately not applied; the focus of the analysis was firstly on the subjective experiences of the participants, and then on the key interactions with HCPs in relation to end of life care decision making (18).

Findings

Study population

Recruitment began following ethical approval in January 2016. 17 parents of 11 children participated in a total of 11 interviews. The sample characteristics are outlined in Table 2. Together for Short Lives categories are outlined in Table 3.

Table 2: Sample characteristics

Family	Participants (mother / father)	Time since bereavement	Age of child	Child's diagnosis / Together for Short Lives category (2)
1	M & F	1 year 6 months	3 years	3
2	M & F	9 months	6 months	4
3	M	5 months	5 months	3
4	M & F	8 months	11 years	1
5	M & F	1 year 7 months	2 years 11 months	1
6	M	1 year 7 months	9 months	1
7	M & F	10 months	18 years	1
8	M	1 year 1 month	5 months	4
9	M & F	7 months	16 years	4
10	M	1 year 11 months	2 years	1
11	M	10 months	1year 11 months	4

Table 3: Together for Short Lives Categories

Category	Description
1 Life-threatening conditions for which curative treatment may be feasible but can fail	Access to palliative care services may be necessary when treatment fails or during an acute crisis, irrespective of the duration of threat to life. On reaching long-term remission or following successful curative treatment there is no longer a need for palliative care services. Examples: cancer, irreversible organ failures of heart, liver, kidney.
2 Conditions where premature death is inevitable	There may be long periods of intensive treatment aimed at prolonging life and allowing participation in normal activities. Examples: cystic fibrosis, Duchenne muscular dystrophy.
3 Progressive conditions without curative treatment options	Treatment is exclusively palliative and may commonly extend over many years. Examples: batten disease, mucopolysaccharidoses.

4 Irreversible but non-progressive conditions causing severe disability, leading to susceptibility to impaired health	Children can have complex health care needs, a high risk of an unpredictable life-threatening event or episode, health complications and an increased likelihood of premature death. Examples: severe cerebral palsy, multiple disabilities, such as following brain or spinal cord injury.

Themes

Five overarching, interrelated themes were identified:

1. Parents have significant knowledge and experiences that influence the decision-making process
2. Trusted relationships with healthcare professionals are key to supporting parents making end of life decisions vital
3. Verbal and non-verbal communication with healthcare professionals impacts on the family experience
4. Engaging with end of life care decision making can be emotionally overwhelming, but becomes possible if parents reach a "place of acceptance"
5. Families perceive benefits to receiving end of life care for their child in a PICU

1. Parents have significant knowledge and experiences that influence the decision-making process: *"I may not have the practical skills that the nurse has, but my knowledge of my child and my child's illness far surpasses that"*

Parents had intimate knowledge of their child as a person and significant expertise related to their child's condition. They frequently used medical slang and jargon during the interviews, such as *"her blood pressure was in her boots"*, "[oxygen] *sats"* and *"oscillated again"*. Having seen how clinical measurements, blood test results and other investigations were used in medical decision-making, they referred to numerical measurements to provide some certainty:

> *"towards the end once we'd really got into the grips of treatment, you know, we understood our kids blood work, you know, literally like breathing and everything that's going on… we automatically look at blood work and know what's going on" (Mother 10)*

Almost all parents had experienced several serious episodes of dramatic deterioration in their child's condition, during which they had gained detailed knowledge of high intensity, specialised treatments. These experiences shaped and influenced their hopes and expectations for the medical management that would be provided for their child:

> *"… about a week before we kept saying [Child010] probably could do with going onto [intervention] but there was another child on [intervention] at that time and … so we were sort of waiting, umming and arring over the [intervention] … I remember [child's father] saying at the time 'but we told you days ago'" (Mother 11)*

Parental decisions related to their child receiving high intensity treatments could also be influenced by a sense that there was "*nothing to lose*", when the alternative was that their child would almost certainly die:

> *"if we leave the child as she is, she's going to pass away anyway. But if we do the, if we put her on the machine there's a chance that she's going to have a problem – said well it's a no brainer, you know. What's the point? You know, if you're going to leave her, she's going to pass away anyway. You might as well on the machine, what have you got to lose?" (Father 9)*

Clinical uncertainty was a common experience and was particularly confusing and difficult for parents when they were used to making decisions based on precise medical explanations and test results. In this situation, parents hoped for consensus amongst their HCPs:

> *"You couldn't help but feel -- not quite them and us, but was more -- it was case where you thought, 'Are they all on the same page?' Well, they probably were on the same page…. But if they're not, we've got a fight on our hands …" (Father 4)*

Parents' expertise extended to the ability to recognise that their child was dying when that time came. Many of the parents described recognising that their child was dying before any open conversations had occurred with healthcare professionals about this. Frequently there was a tacit realisation, knowing "*in my heart*", or having a feeling that "*she just wasn't right*". For some, the realisation was associated with seeing their child's increasing dependence on life-sustaining interventions:

> *"Because there was really nothing and there was no improvement whatsoever … The machine got knocked and … it was only for a second, it just jumped in and his blood pressure crashed down low and his heart rate got down very low and, off literally just for a second" (Mother 4)*

For others, it was a realisation that their child's condition was deteriorating despite the treatments that they were receiving:

> *"… you then don't necessarily need to be a doctor to understand that you're on a bit of a one-way street" (Father 1)*

> *"If I'm honest, the moment they ventilated her, I think deep down we knew she wasn't coming back that time. … I didn't want to say it out loud and I didn't want to admit it, but I think that we all knew" (Mother 10)*

2. Trusted relationships with healthcare professionals are key to supporting parents making end of life decisions: *"I've got to admit they went a little bit above and beyond their duties"*

Trusted relationships with healthcare professionals (HCPs) were highly valued. Continuity of care was a key factor underpinning the development of such relationships. Parents often identified, by name, the individuals who they particularly trusted. These were often HCPs from PICU but were also members of other teams involved in their child's care. Such HCPs had usually advocated for the child or the parents at difficult times and offered extra support, such as providing a personal contact number or going into work on a day off to see the child and family:

> *"So he gave me his mobile number, so 'Just text me or something and I'll make sure that one of my team would come down and see you and then we'll make sure that she's getting the right sort of ….' That was really very kind of the [Doctor] to do that and that made life a lot easier for us, a lot easier for everybody" (Father 9)*

These individuals were particularly important at times when key decisions were being made.

> *"He was very good at explaining things, he was and he would answer any questions … from my perspective when he was telling us 'I'm very sorry. There's nothing else that we can do.' And then it was believable, I didn't feel like we're being fobbed off, or anything like that" (Father 4)*

Parents recognised that these HCPs had their own emotions and stresses to cope with, and empathised with how this affected their work.

> *"the doctor that helped us at the end was lovely. … Couldn't have asked for a better doctor and it turns out that unfortunately five people died on [intensive care] that day. So he had a bad day" (Father 7)*

Relationships with HCPs were fragile and trust was easily compromised. Parents described occasions when they were provided with conflicting advice were described as difficult:

> *"One would be happy to do something or they'd put him on something. And then another one would come in and say, 'Now take him off that…' So that was where we found it a bit hard -- it's different" (Mother 4)*
>
> *"it's a great idea to have one consultant that will oversee because there's so many doctors in-out, in-out, in-out, you know, and obviously everybody's got different opinions as to how things should be done. I think for [Child010] she would have really benefited from having one person that had one say" (Mother 10)*

One mother described how her trust was compromised when she discovered that a meeting had been held about her child's care without her involvement; "*you have broke[n] my trust again completely*" and "*don't lie to me*" (Mother 9). It also happened when parents discovered that an aspect of their child's medical treatment was not being openly discussed with them:

> *"It's that trust relationship, you trust so openly because your consultants, doctors, registrars, nurses, these are like Gods and you're looking at them and thinking come on, I know this happened. It doesn't change what's happened, we know that [complication]'s part and parcel and this is a risk, we know." (Mother 10)*

Trust was also compromised if parents felt that they were not being listened to or felt that they had to repeat their concerns over and over to many members of staff:

> *"you're having to explain what [the child's condition] is, and it becomes "groundhog day" that you're doing it over and over again"(mother 6).*

3. Verbal and non-verbal communication with healthcare professionals impacts on the family experience: "*somebody needs to sit with you and explain why you can't have this or why you can't have that*"

Parents described their experiences of receiving information both at the bedside, and during more formal meetings. Both verbal and non-verbal communication made a difference to parents. In terms of verbal communication, parents described how important it was for information to be presented in a clear and sometimes brutally honest fashion. It helped if this information was given by a trusted HCP, but that sometimes depended on the uncertainties of the shift roster:

> *"I always asked her from the beginning if I'm looking at mortality, then tell me. And a few nights before the end, she said, 'You're looking at it.' So yes, but that was more to do with kind of -- I don't know, good luck or whatever you want to put in that. It might not have been her, if it would have been someone else, I wouldn't have had that at all." (Mother 6)*

Meetings to discuss end of life care with the clinical team were challenging experiences for parents. They were frequently outnumbered by an "*overwhelming*" number of staff which they interpreted as an indication of the severity of the situation:

> *"[Child 4] was very popular with the staff in the hospital and so a lot of people took an interest in him. They just wanted to be there at that meeting and we very much appreciate them wanting to do that. But I think it was a bit disconcerting" (Father 4)*

Parents' described a realisation that their child was dying informed by actions rather than explicit communication, such as a move to particular area of PIC or being given priority to use the parent's bedroom:

> *"And we moved over to [letter] side; that was awful. Because there you're surrounded by a lot of other sick children and then you think, 'Well, that must mean [child]'s really sick as well'" (Mother 3)*

> *"And so when she [nurse] came over … I said 'oh I'm sorry, I've left all my stuff in the parents' bedroom, I'll get it out in a sec so you can use it' and she said 'oh no, no, nobody's using it'…. and she said 'nobody's need is greater than yours today' and I thought 'shit'. And just hearing that, I thought 'oh hang on, so you're telling me basically I've got the sickest child on the unit'. And it was a bit of a wake-up call" (Mother 11)*

One parent described a palpable shift in the manner in which HCPs spoke to her as her child was dying:

> *"my relationship with all the consultants on the unit shifted and they all of a sudden became very business-like and very, how I'd seen them with other parents but never with me. It had always been quite a chatty friendship almost and very comfortable with each other, chatting to people. And now they were very matter of fact, very focused and very negative. So Sunday morning we're all stood round the bed and each and every one of them are just looking at [Child] with this grave look on their face and they're clearly all upset, shocked and uncomfortable … So I found them quite difficult and quite brutal really even though I could see why they were doing it……. They were all of a sudden now, a bit like barriers were going up and they were stepping back from, like 'we can't be chatting, we can't be your friend now, we've got a job to do and we need to focus on this'". (Mother 11)*

4. Engaging with advance care planning can be emotionally overwhelming, but becomes possible if parents reach a "place of acceptance"

Parents experienced wide-ranging, intense emotions towards the end of their child's life which impacted on their ability to take part in end of life care decision-making. They described a range of conflicting emotions related to a hope that their child would not suffer, and a simultaneous fear that their child was going to die. These emotions created stress, anxiety, a state of inner conflict and cognitive dissonance. Parents felt intensely vulnerable as their children approached the end of life. They described feeling *"mentally worn down"* and "*not being able to think straight*", as below:

> *"looking back, I realise just how, you know, mentally worn down with an overload of information I was because I remember them asking me to sign the consent form for the [treatment] and I was looking at the form and … my mind had just completely gone … I just couldn't physically remember how to sign my own name" (Mother 10)*

> *"It was such a busy few hours that morning. And the noise was incredible. And it probably wasn't any more noisy than normal, but it felt massively noisy. I just, probably because I was just exhausted and I'd had sleeping tablets and I think I was still recovering from them and I couldn't think straight about anything. … And then the next significant thing was they said 'we need to go and have a chat". (Mother 11)*

In these contexts, end of life care decision-making could feel overwhelmingly difficult for parents, particularly when they were being asked to make decisions about the withdrawal of life-sustaining treatments:

> *"I think the hard thing was that, you know, they were kind of, like, 'what do you think?', which is great that they wanted to ask our opinion but, at the time, we were like so overwhelmed. And I remember thinking, why are they asking us! No, I understand it's good to give parents that power but I was like, you know, 'I have no idea, they're the experts'" (Mother 8)*

Clear guidance and the support of trusted clinicians was critical. The manner in which they were engaged in the decision-making process was important, for example feeling that they have made a choice to "*say goodbye*" rather than having to make a choice to withdraw life-sustaining treatments.

> *"that decision didn't come about easy. It didn't -- people think oh well, you chose to switch his life support off. Yeah, we did but we also chose to say, we had to choose to say goodbye to him, you know what I mean?" (Father 5)*

> *"So whilst I know it comes under sort of end of life decision, it didn't feel like we were having to decide whether he'll live or not. It was more about agreeing that it was time to stop, which I think was the right way to do it. All the way through this, we've been led by the medical teams" (Father 4)*

There was some comfort in making decisions that they considered to be "*best*" for their child.

> *"As much as it did hurt us to let him go, we were thinking what was best for him to be comfortable and not in pain" (Mother 2)*

The need to preserve their role as parents, providing love and care for their child, was strongly apparent in their accounts. One parent described how much she valued being given the opportunity to hold her child:

> *"all you can see is that your child is just hooked up to everything possible and they made my day when they lifted her up once she was – because she was relatively flat with all the drugs and what not – and lifted her up so I could actually have a cuddle and put her on me. Oh, even now, I'm so grateful that they did that" (Mother 10)*

Some parents expressed a desire to know what to expect when it came to their child dying, although they appreciated that this may not be something that other parents would want:

> *"...not knowing what death is and what it's going to look like... when you're seeing it for the first time, when you're kind of dealing with it, both as an experience of death but also as your baby …. I would like to have known that…sorry … Not everybody would ..." (Mother 6)*

Where parents felt they were missing crucial information, they sought it out from other parents:

> *"We did need to know what happened if we switched the machines off. And so I did ask another parent about what happened to their friend when at the hospital, because I wanted to imagine that scenario if it was going to happen …" (Mother 1)*

Not all of the parents were aware of advance care planning (ACP), and many had not experienced this for their child. There were opposing views, with some parents feeling that ACP "*would have been very useful*", and others that a plan which considered the child's death was not acceptable; "*never an option*". Parents reported that the timing of conversations with respect to ACP was important, but could be particularly difficult where there was uncertainty about the likely outcome of a treatment or procedure, such as surgery or a new medical intervention:

> *"We knew that his life would be short … but we never planned for a negative, we always planned for positives. I don't think anybody told us the potential negative of that [intervention] and I don't think they knew the potential negatives that could happen because they weren't expecting that to happen" (Father 7)*

Parents described the need to be in a "place of acceptance" in order for ACP conversations to take place:

> *"I think you have to come to a place yourself to kind of accept what's going to happen. And until you've got to that acceptance, I think it can make people very aggressive. … And for me, I kind of accepted that was it at that point." (Mother 6)*

Parents who had made a formal ACP for their child made practical suggestions related to the information and knowledge that should be considered for parents in order for them to make an informed decision about whether to create an ACP. They suggested that it was necessary to observe and understand the implications of particular interventions, such as ventilation, before considering this in an ACP:

> *"it'd be nice to have that little bit of a tick box 'Has the parent seen a ventilator?' I know they've made this decision but you know like when you have at the end, 'cause some just don't want anything, and that's fine and some have just the oxygen. Some want IVs, you know you've got that little paragraph at the bottom, so just underneath it you know, it would be nice 'Have they seen it?' and at least then you'll know. And maybe then that's the time when the consultant will say 'Would you like to see it?' you know just, we can do a little session for you where you can go in and have a look" (Mother 9)*

5. Families perceive benefits to receiving end of life care for their child in a PIC: *"the support that we received on [PICU] was just amazing"*

The parents all provided poignant accounts of their child's death. Some were shocking and traumatic, for example when they witnessed resuscitation as their child's terminal event:

> *"And then I think for me the last straw was when the last time they were doing the [cardio-pulmonary resuscitation] CPR, is when the guy came with the drill and he started to put a drill in her" (Father 9)*

> *"then at some point we knew they'd been working for ages, they were going at it and then I heard one of them crack a rib … we'd obviously heard that and we knew that there's no way her chest was coming back from that, she was struggling already and it's not their fault, it happens, you know, it is what it is" (Mother 10)*

However, even in these traumatic circumstances, parents valued the reassurance provided in PICU that all possible treatment options had been explored for their child:

> *"There was nothing that they could do for him and they had made that clear – they'd tried everything. They even tried things they thought weren't going to work" (Mother 2)*

> *"By this time I knew a lot of what was goes on in the unit and I knew that [intervention] wasn't a good place to be. I knew that I hadn't seen many kids come off [intervention] and go home but, at the same time, I thought well it's worth a try and I also knew if they didn't think it was worth doing, they wouldn't have done it. It's expensive. And I know you can't put a price on life, can you, but I knew that they wouldn't have tried that unless there was a chance" (Mother 11)*

Being in PICU and having more days of life than might have been possible in other clinical settings was also highly valued:

> *"we had [doctor] worked several nights through with her, where he didn't leave her bedside for lines and things like that; of which in hindsight, she was going to go anyway. But by doing that, he gave me an extra couple of days of which, if we'd gone to a hospice you can't do things like that. She would have just gone. So for me, I wouldn't have wanted that" (Mother 6)*

Discussion

Summary

This study provides important insights into the experiences of end of life care decision-making of parents of children with life-limiting and life-threatening conditions who die in a PICU. The study has identified important themes that affected their experience. The parents who participated in this study had expert knowledge of their child and their child's condition, and wanted this to be taken into account in medical decision-making. Parents were often aware that their child may be dying before this was openly acknowledged by their HCPs. Trusted relationships with HCPs were critical to their experience of end of life care decision-making, as were both verbal and non-verbal communication. Every situation was unique; making decisions about care at the end of a child's life was described as "overwhelming" by some. The parents in this study expressed a preference for end of life care discussions to be conducted by a trusted HCP in small meetings.

Parents described the need to have come to a “place of acceptance” in order to be able to take part in end of life care decisions. The idea of an ACP was received positively by some parents, but was completely unacceptable, even in principal, to others. The end of life was traumatic for parents to witness if associated with attempts at resuscitations and invasive medical procedures, however the continuity of care provided in PICU, reassurance that all possible treatment options had been tried for their child, and the extra hours of life that could be provided were all perceived as important benefits by parents.

Strengths and Limitations of the study

Recruiting to research about end of life care in children is known to be challenging (19). The study was conducted with parents whose children had died from a diverse range of life-limiting conditions. However, the number of participants is relatively small, and they were all recruited through the same PICU which may limit the generalisability of the findings. While data saturation was reached around the key themes reported here, it is likely that the parents who felt unable to participate may have had views, experiences and perceptions that were different. There were several emerging themes in our data analysis which are not reported here, including the experience of end of life care meetings, the care of siblings, spiritual needs and bereavement care; all are worthy of further research. Furthermore, the study’s findings are based on retrospective accounts that may have been re-framed over time. We did not capture the experiences and perceptions of families who are currently in the process of making end of life care decisions for their children, or the views of any children or young people regarding their own end of life care decision-making.

Comparison with existing literature

There is a lack of empirical research examining end of life care decision-making in PICU, perhaps because admission to PICU does not tend to be explicitly for end of life care (20). Our findings contrast with previous research that suggested an awareness amongst HCPs that a child may be dying before this is recognised by their parents (21). In keeping with our finding that parents often had an unspoken knowledge that their child was dying, there are published case studies and parental narratives which also suggest an earlier recognition amongst parents (22, 23). A situation of “mutual pretence” may be reached between parents and HCPs, where this knowledge is unspoken; this situation of mutual pretence could potentially be acknowledged more openly in order to start discussions about palliative and end of life care, including referral to specialist paediatric palliative care services, where available (24).

Previous studies have described influences on parental decision-making as the child's diagnosis, prognosis and the extent of their pain, discomfort or suffering (21, 25). Our findings indicate that clinical uncertainty, unpredictable outcomes of treatments, a sense of loss of control, and the intense emotional burden that can exist for both parents and HCPs add complexity of end of life care decision-making in PICU. As medical treatments advance, and information about such treatments becomes more available particularly in media reports, these clinical and ethical complexities are becoming ever more prominent. In this context, for parents who may already be finding it difficult to understand that their child's condition is incurable (26), conversations about end of life care may represent a significant change from previously cure-focussed management plans. Previous research has suggested that parents do not always need to fully acknowledge their child's situation in order to place emphasis on the relief of suffering (21). It may therefore be possible to reach a situation earlier on in a child's illness which provides the opportunity for conversations about end of life care, ACP or referral to specialist paediatric palliative care services, through skilful acknowledgement of uncertainty and the conflicting emotions that parents may be experiencing, including fear and hope.

Parents value affirmation in their decision-making from a HCP who is known and trusted and who has witnessed the magnitude of their child's illness (27). Trusted relationships with HCPs were critical to the experience of the parents in this study as they tried to make decisions which were "best" for their child. Continuity of care, and a visible commitment to understanding and addressing the end of life care needs of the child and their family, were important in achieving such relationships. In keeping with previous studies, there were times when conflicting advice brought about through changes in staff could cause parents to feel confused and abandoned (28). Provision of a working environment that allows for continuity of care warrants attention, as does care for the workforce as they face these emotionally charged, ethically challenging situations with children and their families.

Parents were reassured by the knowledge that all possible treatment options had been explored for their child, and valued the extra days of life provided by the delivery of high intensity treatments in PICU. Given the rising numbers of children with life-limiting and life-threatening conditions and the complexity of their needs, there is a need to consider how this care and associated reassurances can be offered to children in environments other than PICU, including high dependency units, and children's hospices. There is a careful and highly individual balance to be found for each family between the knowledge that everything possible has been tried for their child, alongside preparation for the time when high intensity treatments may become futile and potentially harmful towards the end of a child's life (29).

Recommendations

Wider recognition of the complex factors that relate to end of life care decision-making in PICU, and an organisational commitment to providing a clinical environment in which continuity can be provided to families, could both assist with the implementation of policy guidance related to end of life care decision-making.

An important area for research is further investigation into the child and family perspectives of ACP, and the impact of earlier integration of palliative care into a child's care. A recent study suggested that children who received SPPC were five times less likely to receive high intensity treatments at the end of their lives (30). Another study examined the potential impact of routine referral to SPPC when a child was commenced on extracorporeal life support in PICU (31). This approach to the introduction of SPPC could be further explored. However, SPPC services are inconsistently funded and provided both in the UK and internationally (10); ACP and end of life care decision-making therefore depends on the principles of palliative care being practiced amongst the wider workforce.

There is more work to be done to understand how end of life care can be effectively achieved in PICU for individual children and families. Future research into the views of children regarding their own end of life care decision-making and ACP, as well as research to further understand the experiences of families who are currently in the process of making end of life care decisions for their children would be of value.

Conclusion

Learning from the experiences and perceptions of families should inform improved policy and practice. This study highlights the need for recognition of parental expertise and experience, and the critical importance of a trusted relationship between families and their HCPs, which can often be established through repeated admissions or prolonged stays in PICU. Whilst trust is vital to the relationships between families and HCPs, it is also fragile, and can be easily lost. Parents are highly vulnerable and may be exhausted, confused and uncertain at the times when they are asked to engage with end of life care decision-making. Earlier acknowledgement of clinical uncertainty and the conflicting emotions that parents may be experiencing could help in earlier discussions about end of life care and introduction to specialist paediatric palliative care services, where available.

The needs of each family and their readiness for involvement in decision-making is highly individual. ACP is not well understood by parents, and appears to be more helpful for some than others. End of life care of a child on PICU provides potential benefits from a family perspective, and there is a need to consider how the care and reassurances they perceive can be provided in other environments.

There is also a pressing need for greater understanding of the child's experience, which should be the focus of further research.

Author Contributions

The study was conceptualised by SM, AP, JC and JD, informed and guided by Patient and Public Involvement, with specific guidance and advice provided by EH. JS conducted the interviews. AP managed the conduct of the study. SM, JS and AP drafted the article with contributions from JC and JD. EH acted as PPI co-author and reviewed the article to ensure relevance to the family situation. JC and JD reviewed the article for intellectual content and edited the final version. All authors reviewed, edited and agreed this version.

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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

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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, it's 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.

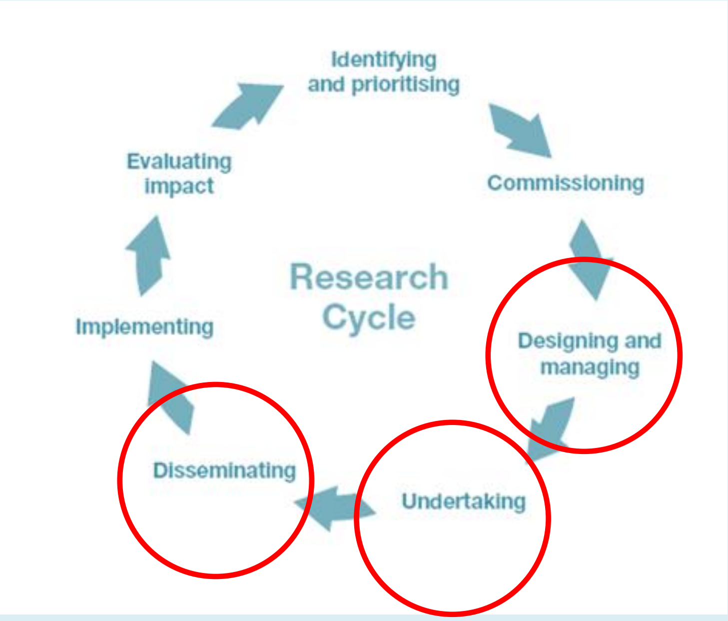
Aim

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.

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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	1) Stop the interview 2) Allow time for the participant to regroup and offer support 3) Assess further with following questions: a) How are you feeling right now? b) What thoughts are you having? c) Do you feel able to continue with your day? Decide if they are experiencing **acute emotional distress beyond what would be normally expected in an interview about a sensitive topic.**		
Display behaviours suggesting they are too stressed (crying uncontrollably, struggling to speak clearly)	1. Stop the interview 2. Allow time for the participant to regroup and offer support 3. Assess further with following questions: a) How are you feeling right now? b) What thoughts are you having? c) Do you feel able to continue with your day? Decide if they are experiencing **acute emotional distress beyond what would be normally expected in an interview about a sensitive topic.**		

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.

Adapted by J. Spry from Drauker, Martsolf & Poole (2009)

Reporting checklist for qualitative study.

Based on the SRQR guidelines.

Instructions to authors

Complete this checklist by entering the page numbers from your manuscript where readers will find each of the items listed below.

Your article may not currently address all the items on the checklist. Please modify your text to include the missing information. If you are certain that an item does not apply, please write "n/a" and provide a short explanation.

Upload your completed checklist as an extra file when you submit to a journal.

In your methods section, say that you used the SRQR reporting guidelines, and cite them as:

O'Brien BC, Harris IB, Beckman TJ, Reed DA, Cook DA. Standards for reporting qualitative research: a synthesis of recommendations. Acad Med. 2014;89(9):1245-1251.

		Reporting Item	Page Number
	#1	Concise description of the nature and topic of the study identifying the study as qualitative or indicating the approach (e.g. ethnography, grounded theory) or data collection methods (e.g. interview, focus group) is recommended	1
	#2	Summary of the key elements of the study using the abstract format of the intended publication; typically	2

		includes background, purpose, methods, results and conclusions	
Problem formulation	#3	Description and signifcance of the problem / phenomenon studied: review of relevant theory and empirical work; problem statement	4
Purpose or research question	#4	Purpose of the study and specific objectives or questions	4
Qualitative approach and research paradigm	#5	Qualitative approach (e.g. ethnography, grounded theory, case study, phenomenolgy, narrative research) and guiding theory if appropriate; identifying the research paradigm (e.g. postpositivist, constructivist / interpretivist) is also recommended; rationale. The rationale should briefly discuss the justification for choosing that theory, approach, method or technique rather than other options available; the assumptions and limitations implicit in those choices and how those choices influence study conclusions and transferability. As appropriate the rationale for several items might be discussed together.	5
Researcher characteristics and reflexivity	#6	Researchers' characteristics that may influence the research, including personal attributes, qualifications / experience, relationship with participants, assumptions and / or presuppositions; potential or actual interaction between researchers' characteristics and the research	5

		questions, approach, methods, results and / or transferability	
Context	#7	Setting / site and salient contextual factors; rationale	5
Sampling strategy	#8	How and why research participants, documents, or events were selected; criteria for deciding when no further sampling was necessary (e.g. sampling saturation); rationale	5
Ethical issues pertaining to human subjects	#9	Documentation of approval by an appropriate ethics review board and participant consent, or explanation for lack thereof; other confidentiality and data security issues	3
Data collection methods	#10	Types of data collected; details of data collection procedures including (as appropriate) start and stop dates of data collection and analysis, iterative process, triangulation of sources / methods, and modification of procedures in response to evolving study findings; rationale	4
Data collection instruments and technologies	#11	Description of instruments (e.g. interview guides, questionnaires) and devices (e.g. audio recorders) used for data collection; if / how the instruments(s) changed over the course of the study	4
Units of study	#12	Number and relevant characteristics of participants, documents, or events included in the study; level of participation (could be reported in results)	5

Data processing	#13	Methods for processing data prior to and during analysis, including transcription, data entry, data management and security, verification of data integrity, data coding, and anonymisation / deidentification of excerpts	4
Data analysis	#14	Process by which inferences, themes, etc. were identified and developed, including the researchers involved in data analysis; usually references a specific paradigm or approach; rationale	4
Techniques to enhance trustworthiness	#15	Techniques to enhance trustworthiness and credibility of data analysis (e.g. member checking, audit trail, triangulation); rationale	5
Syntheses and interpretation	#16	Main findings (e.g. interpretations, inferences, and themes); might include development of a theory or model, or integration with prior research or theory	5-12
Links to empirical data	#17	Evidence (e.g. quotes, field notes, text excerpts, photographs) to substantiate analytic findings	5-12
Intergration with prior work, implications, transferability and contribution(s) to the field	#18	Short summary of main findings; explanation of how findings and conclusions connect to, support, elaborate on, or challenge conclusions of earlier scholarship; discussion of scope of application / generalizability; identification of unique contributions(s) to scholarship in a discipline or field	13

Limitations	#19	Trustworthiness and limitations of findings	`13
Conflicts of interest	#20	Potential sources of influence of perceived influence on study conduct and conclusions; how these were managed	3
Funding	#21	Sources of funding and other support; role of funders in data collection, interpretation and reporting	3

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