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Evaluation of the burdens and benefits of participation in research by parents of children with life-limiting illnesses

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Abstract

Background—Research is needed to improve care and diminish suffering for children with life-limiting illnesses and their parents. However, there are doubts about whether it is possible to conduct paediatric end of life research safely and ethically, as it may unduly burden or inadvertently harm participants.

Aim—To compare and evaluate responses from participants to the assessments of burdens and benefits that were conducted at two timepoints during a phenomenological study that investigated parents' experiences of having a child with life-limiting cancer participate in a Phase I clinical trial.

Discussion—Parents reported that participating in the study was beneficial and resulted in minimal burden or distress. The assessment of benefits and burdens at the first timepoint appeared sufficient to understand participants' experiences.

Conclusion—This study adds to the evidence that research may be safely and effectively conducted with parents of children who are deceased or have life-limiting illnesses. Further research is needed to evaluate the most effective timing of assessments of the burdens and benefits of their participation in research.

Implications for practice—It is important when conducting research with people with life-limiting illnesses or their family members to assess the burdens and benefits of their participation, to understand their experiences and assist in its conduct.

Keywords

cancer; cancer research; clinical trials; end-of-life care; families; research; study participation; patient experience; patient feedback; patients

Introduction

A life-limiting illness is a condition for which there is no cure and that is expected to cause the patient's premature death. The death of a child can cause their parents, siblings, extended

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families, peers and healthcare providers significant physical, psychological, social and spiritual pain (Akard et al 2013). Research into palliative and end of life care that considers the experiences of patients and their families, including bereavement, has become a global priority (Sigma Theta Tau International 2005, Kaasa 2008, Higginson 2016, Clark et al 2018). Children with life-limiting illnesses and their parents must participate in such research, to identify problems accurately, highlight how these can be addressed and ultimately improve outcomes.

There are doubts about whether it is possible to conduct research ethically and safely with children with life-limiting illnesses and their parents, as it may cause additional burden or even inadvertent harm (Hinds et al 2007, Akard et al 2014, Bloomer et al 2017). Children and those who are terminally ill are considered vulnerable populations (Shivayogi 2013); bereaved parents who cope with intense grief are also vulnerable, although they are not usually regarded as a vulnerable population (Shivayogi 2013, Akard et al 2014). Obtaining ethical approval for studies involving children with life-limiting illnesses and their parents can be challenging, as approval boards may assume that research would worsen the participants' already tenuous circumstances (Hynson et al 2006). 'Gatekeepers', such as family carers, healthcare providers and approval boards, can deny researchers access to individuals they deem vulnerable (Juritzen et al 2011, Cook 2012, Crocker et al 2015, Bloomer et al 2017).

Assessing the burdens and benefits to children with life-limiting illnesses of participating in research captures their experiences and allows those participants who suffer harm or distress to be referred for further support (Hinds et al 2007, Bloomer et al 2017). However, it is unclear when to do this. Hinds et al (2007) proposed a research strategy for paediatric oncology end of life studies that included follow-up calls to assess participants' experiences; other studies conducted assessments immediately after completing other activities, such as data collection (Olcese and Mack 2012, Wiener et al 2015, Allen and Kelley 2016, Aoun et al 2017). The authors' search of the literature found nothing comparing the effectiveness of these two strategies.

The authors conducted a phenomenological study of parents' experiences of having children with life-limiting cancer participate in a Phase I clinical trial (Crane et al 2018). This study assessed the burdens and benefits of participating in the trial, to ensure parents were not unduly psychologically distressed by sharing their experiences. The authors conducted these assessments twice: once after collecting data from the participants (timepoint one) and again seven to ten days later in a follow-up call (timepoint two).

The purpose of this paper is to compare the participants' responses at both timepoints and compare the values of the two assessments.

Methods

Primary study results and further details of the study's methods are published elsewhere (Crane et al 2018).

Recruitment

Eligibility criteria for the study included:

- Parents had to be 18 or more years old.
- Parents had to be children's primary caregivers.
- Parents had to speak English fluently.
- Children had to be enrolled in at least one paediatric oncology Phase I clinical trial in the US.
- Children had to be younger than 18 during the trial.
- Children had to have left the Phase I trial at least 60 days before the study, to ensure that the off-study transition was fully experienced.

Parents were ineligible for the study if the child had died within the previous 60 days.

A pilot interview was conducted before the main study with a volunteer parent, who was recruited based on knowledge of the parent's advocacy efforts. The main study recruited participants from two children's hospitals and parent advocacy groups between March and December 2016. Parents of children attending the hospitals were identified from clinical trial records and sent letters by the hospitals containing details of the study and instructions for opting in or out; parents who did not respond were contacted two weeks later by phone. Parent advocacy group leaders were contacted by email and asked to share recruitment materials with group members; interested parents then contacted the authors.

Procedures

If parents met the eligibility requirements, they were asked to complete an electronic form confirming their agreement to participate and collecting their demographic data. They then took part in an unstructured telephone interview lasting approximately one hour, in which they were asked to share the entire story of their children's participation in Phase I trials; the depth of the discussion was then enhanced by using further probing questions, such as 'Tell me more about that' or 'What did that mean to you?'

At the end of the interview (timepoint one), participants completed an assessment of the study's burdens and benefits using the modified Pessin et al (2008) instrument, and were asked if they had any additional information or concerns to share. Parents whose child had died were then offered local bereavement services. A pre-determined safety plan was in place for distressed participants.

Seven to 14 days after the interview (timepoint two) [Q1: In Introduction it says seven to 10 days. Which is correct?], participants were contacted for a follow-up call. This was conducted by a research assistant unknown to the participant using a modified version of Pessin et al's (2008) instrument and Hinds et al's (2007) research strategy. The protocol included two attempts at calls at various times of the day, at least three days apart. Only one voice message was left with the first call attempt. Participants were asked:

- To share their overall impression of the primary interview.

- If they wanted to share any additional or clarifying information.
- To complete another assessment of the study's burdens and benefits using the modified Pessin et al (2008) instrument.
- If the follow-up telephone call was appreciated and, if so, why?
- If the follow-up telephone call was difficult or inconvenient and, if so, why?

All interviews and follow-up calls were recorded, transcribed by an independent, third-party professional, and verified for accuracy by the second author.

Modified Pessin et al (2008) instrument

The modified version of Pessin et al's (2008) instrument consisted of two questions, one asking if participation in the study was burdensome, the other if it was beneficial (Akard et al 2014). Answers were scored on a five-point Likert scale, with options ranging from 'not at all' (zero) to 'very much' (four). Participants who responded that participation was more than 'a little bit' burdensome or beneficial were asked a closed, multiple-choice follow-up question to clarify why participation was burdensome or beneficial.

The original instrument was modified for this study from the original by omitting the options in the follow-up question that did not apply to parents of children with life-limiting illnesses or to the study design. For example, the original instrument had several inapplicable reasons why participation was burdensome ('I was too weak or too ill at times' and 'it interfered with other activities that I wanted to do') or beneficial ('I enjoyed having the company' and 'it helped pass the time/kept my mind busy').

Results

Recruitment

In total, 11 parents were recruited to the study, not including the pilot participant who did not complete any assessments of burden or benefit. The participants were six of the 17 parents (35%) approached at the first children's hospital, three of the nine parents (33%) approached at the second children's hospital and both of the two eligible parents found through parent advocacy groups.

Most were female ($n=9$, 82%); 10 (91%) specified they were white, while one did not state their ethnicity. The mean age of their children at the time of enrolment in the first Phase I trial was 12.2 years old (SD 4.9 years; range: three to 17 years old) and the mean number of children in the household was 1.5 (SD 1.9; range: zero to six). Most parents ($n=8$, 73%) held a college or professional degree or higher. Eight parents (73%) were bereaved.

One parent failed to complete the interview, resulting in a 91% retention rate. No parents reported significant distress from participating in the study and the safety plan was not implemented for any participants. No parent reported at either of the two timepoints any burden resulting from participation.

Timepoint one

At the first timepoint, parents reported a median benefit of participation as ‘quite a bit’ (median=3, mean=2.6). The most frequently selected reasons on the modified Pessin et al (2008) instrument for this were:

- ‘It was helpful or a relief to talk about this issue with someone’ ($n=9$, 90%).
- ‘It made me feel good to help others/contribute to society’ ($n=6$, 60%).
- ‘It helped me think about these topics’ ($n=5$, 50%).

Timepoint two

At the second timepoint, only one (10%) of the parents responded to the first call attempt; a voice message was left for the remaining nine parents. A second phone call was made between three and ten days after the attempt, to which two parents responded, for a total of three parents (30%) completing the second assessment of burdens and benefits.

Two of the three responding parents were bereaved. No parents shared new information about their experiences of when their child was in a Phase I trial. One parent was asked to clarify details shared in the interview – for example, to describe more fully their experience when their child was rejected from participating in a Phase I trial at a distant hospital. Only one parent changed an answer they had given to the modified Pessin et al (2008) instrument at the first timepoint – they decreased the benefit of participation from ‘quite a bit’ to ‘somewhat’ (Table 1).

The parental feedback regarding the need for the follow-up call was mixed.

- Parent 1: they did ‘see the need for follow-up phone calls... unless there are remaining questions’ and that it was inconvenient ‘having to carve out time’ for the follow-up call.
- Parent 2: the follow-up call ‘was appreciated’ and not inconvenient.
- Parent 3: reported being ‘neutral’ regarding the need for the follow-up call and that it was not inconvenient.

Discussion

It is important when conducting research with individuals with life-limiting illnesses or their family members to assess the burdens and benefits of participation, to understand their experiences and assist the conduct of the study. This study reinforces that participation in such research by parents of children who are deceased or have life-limiting illnesses causes them minimal or no burden or distress and is personally meaningful to them (Hinds et al 2007, Olcese and Mack 2012, Akard et al 2013, Wiener et al 2015, Allen and Kelley 2016, Weaver et al 2018). It also shows that negative psychological reactions from participating in research are uncommon and generally mild in severity for these parents, so research can be safely and effectively carried out with them (Dyregrov 2004, Hinds et al 2007). Parents who participate in end of life research with their children do so out of altruism, for autonomy and to optimise care (Kavanaugh and Campbell 2014). Benefits include a sense of

empowerment, time to reflect on their experiences and the opportunity to find meaning (Gysels et al 2012, Kavanaugh and Campbell 2014, Aoun et al 2017).

This paper evaluated the use of assessments of burden and benefit when conducting end of life research with parents of children with cancer. The assessment of benefit and burdens at the end of the interview appeared sufficient to understand participants' experiences during the study, the follow-up calls seven to 14 days later providing no additional information. However, the generalisability of these results is limited because of the very low response rate ($n=3$, 30%) to the follow-up calls. It is difficult to interpret this lack of response by the other participants, but the authors hypothesise that it primarily indicates a lack of interest or time to respond; bereavement processes could also have affected parents' responsiveness.

There are implications for researchers and participants to including a follow-up call in a protocol for conducting end of life research with children and parents. These include:

- Additional logistics.
- Increased costs from hiring a research assistant and a transcription service to complete, transcribe, verify and evaluate the follow-up calls.
- Greater time required of participants and researchers.

The lack of value added by the follow-up calls in this study leads the authors to hypothesise that it is possible to evaluate the burden and benefits of participation in a study sufficiently following collection of the data without a subsequent follow-up phone call.

Limitations

The primary limitation of this study was the very small sample, which represented a relatively homogeneous population. This significantly decreases the generalisability of the results.

It is also likely there was a bias created by respondents self-selecting during recruitment – for example, similar participants might respond similarly to the follow-up call attempts.

A further remaining question is whether the frequency or timings of the follow-up calls caused the low response rate. Hinds et al (2007) reported a rate of more than 75%, for example, but did not disclose the protocol used to achieve this.

Further research

Further research with larger, more diverse samples is warranted. It is also needed to clarify the most effective timing and frequency for conducting follow-up calls, given that each time a participant is contacted it is an additional burden that must be considered.

Conclusions

This study adds to the evidence that research may be safely and effectively conducted with parents of children who are deceased or have life-limiting illnesses. Further research is needed to evaluate the most effective timing of assessments of burdens and benefits of

participation, including whether these assessments are best conducted at the end of data collection – versus in a follow-up phone call – and the most effective timing and frequency of follow-up calls.

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Table 1.

Reports of burdens and benefits of participation at the two timepoints

Parent	Timepoint one		Timepoint two	
	Burden	Benefit	Burden	Benefit
1	Not at all	Quite a bit	Not at all	Somewhat
2	Not at all	Not at all	Not at all	Not at all
3	Not at all	A little bit	Not at all	A little bit

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