

## PEER REVIEW HISTORY

BMJ Open publishes all reviews undertaken for accepted manuscripts. Reviewers are asked to complete a checklist review form (<http://bmjopen.bmj.com/site/about/resources/checklist.pdf>) and are provided with free text boxes to elaborate on their assessment. These free text comments are reproduced below.

### ARTICLE DETAILS

<b>TITLE (PROVISIONAL)</b>	Parental experiences of end of life decision-making for children with life-limiting conditions in the Paediatric Intensive Care Unit: A Qualitative Interview Study
<b>AUTHORS</b>	Mitchell, Sarah; Spry, Jenna; Hill, Emma; Coad, Jane; Dale, Jeremy; Plunkett, Adrian

### VERSION 1 - REVIEW

<b>REVIEWER</b>	Elizabeth L. Ciemins AMGA (American Medical Group Association), USA
<b>REVIEW RETURNED</b>	31-Dec-2018

<b>GENERAL COMMENTS</b>	<p>This is an excellent and much-needed study that examines parents' perspectives at the end-of-life of their children. These findings will hopefully help clinicians and PICU staff better understand these perspectives so that they can be addressed and ultimately the EOL experience for parents of seriously ill children can be improved.</p> <p>One aspect that was not touched on in this paper is "moving palliative care upstream." This discussion may be beyond the scope of this paper, but perhaps this came up in some of the interviews?</p> <p>One of our papers from the adult literature might be useful to reference:</p> <p>Ciemins EL, Brant J, Mulette E, Kersten D, Dickerson D. A Qualitative Analysis of Patient and Family Perspectives of Palliative Care. <i>Journal of Palliative Medicine</i> October 2014 (17).</p> <p>Additional comments:</p> <ol style="list-style-type: none"><li>1. In abstract, add (ACP) behind first spell out of Advance Care Planning (bullet 4, Results)</li><li>2. Methods: you describe the response rate for the retrospective approach, but not for the prospective approach. E.g., 8 led to interviews. What about the prospective approach?</li><li>3. Last paragraph in recruitment section should clarify if referring to both retrospective and prospective approaches, which I think you are.</li></ol>
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	<p>4. Data Analysis: at what intervals did you compare data? How often? Also, did you calculate interrater reliability based on these comparisons? If you can report, that would be helpful for the reader to have confidence in the three reviewers.</p> <p>5. Table 2. Your link for the children's diagnoses returned an error, "page not found." Not sure how familiar readers will be with this categorization. I looked on the webpage and couldn't find the definitions. Perhaps you can put something more meaningful in the table to help readers understand this categorization. Even a footnote indicating which direction the scale goes in terms of severity would be helpful, at the least.</p> <p>6. Theme 1. You first theme could be worded more strongly and more consistently. I believe the authors are making the point that parents' knowledge and experience and perspective should be acknowledged and integrated into the care process at the EOL. To say they are "relevant" is a bit unclear and understated. Immediately following the list on page 8, you change the wording to "must be taken into account," which is better, stronger, and perhaps could be used in your 2 previous lists of themes.</p> <p>7. Theme 3. What do you want to say about the variability? Just that it exists? Your first two theme are statements that can be acted upon. This theme is a little vague. Is the variability bad? Should it be reduced? Should improvements be made to provide more communication continuity?</p> <p>8. Theme 5. Again, not sure what your point is on this final theme. Can you restate it to be actionable? To just say, "experiencing the death..." is not an active voice sentence and may be difficult for clinicians to act upon. In general, all your themes should be actionable or at least informative statements.</p> <p>9. Summary. Once themes are clarified a bit, the summary might be improved and expanded. Also, integrate very specific recommendations into the recommendations that relates to your stated themes.</p> <p>This is an excellent and important paper that should definitely be published after a few minor edits that will make this a stronger, more actionable paper.</p> <p>The paper is well-written but could use a copy-editor as there are a few grammatical errors, e.g., "Learning from the experiences and perceptions of families is imperative in order to improving practice."</p>
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<b>REVIEWER</b>	Julie Ling European Association for Palliative Care Belgium
<b>REVIEW RETURNED</b>	10-Jan-2019

<b>GENERAL COMMENTS</b>	<p>General comments: The paper would benefit from structural review. It would be helpful to have sections clearly delineated. introduction Aim, method, results, conclusion.</p> <p>Abstract: I think the abstract could do with better structure and be more concise. I also wanted to read more about the method in the abstract. e.g. results: Who were the interviews with? How many mums? How many dads? ACP needs to be in brackets after it is written in full.</p>
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	<p>Introduction:  Living with uncertainty could be expanded to include the care that parents provide and the impact that this has on family life, siblings etc. and the role of parents in providing sometimes very technical care for their children.</p> <p>Does the PIC involve or have access to palliative care services? And does this have any impact on the care that families receive? Does Birmingham have a palliative care team? I think this is key issue - not to say that all cases should involve palliative care but certainly if children have LLI's then are palliative care not involved at some level?</p> <p>Where/who were referrals for family inclusion in the research received from?</p> <p>Refer to table for the type of LLI - include a list as an appendix? 59 parents of how many children?</p> <p>I am not sure of the ethics and what was included in the ethical approval. If parents were recruited who had lost a child in the last 12 months, it is possible that some were newly bereaved - their experience would possibly be impacted by this compared to someone who has been bereaved for nearly a year.</p> <p>There are two separate groups here. Identifying people prospectively - it is well-documented in the literature that it may impact on findings if people are still receiving care compared to retrospectively. Maybe the two should be identified separately?</p> <p>Was the interview schedule piloted?</p> <p>Was any support offered to parents post-interview if respondents were upset or distressed? or even just to check if they were ok after being interviewed and talking about a distressing experience.</p> <p>P8. Line 31 - Losing control when their child is admitted is a key issue for parents and this could be included here.</p> <p>P16. Line 3 - There is literature in children's palliative care about these issues and it would be helpful for the authors to acknowledge and refer to this.</p> <p>Home as a preference - could the authors say more as to why children are dying in PIC? were parents offered a choice to take their children home? Is support available if they wanted this?</p> <p>Key words: add the UK 'paediatrics'</p> <p>Overall:  I enjoyed reading the paper and found it interesting. I think taking a broader view may improve the content</p>
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### VERSION 1 – AUTHOR RESPONSE

One aspect that was not touched on in this paper is “moving palliative care upstream.” This discussion may be beyond the scope of this paper, but perhaps this came up in some of the

interviews? – the idea of early referral to palliative care services is frequently championed by those in palliative care in the UK, but not often referred to as “upstream”.

1. In abstract, add (ACP) behind first spell out of Advance Care Planning (bullet 4, Results) corrected, thank-you.

2. Methods: you describe the response rate for the retrospective approach, but not for the prospective approach. E.g., 8 led to interviews. What about the prospective approach? Data added

3. Last paragraph in recruitment section should clarify if referring to both retrospective and prospective approaches, which I think you are. Clarification added

4. Data Analysis: at what intervals did you compare data? How often? Also, did you calculate interrater reliability based on these comparisons? If you can report, that would

be helpful for the reader to have confidence in the three reviewers. We have added further details of the data analysis, detailing how the study team worked together on the analysis. The data analysis was deliberately inductive, with the focus on the subjective experience of the participants, and we did not apply a formal framework.

5. Table 2. Your link for the children’s diagnoses returned an error, “page not found.” Not sure how familiar readers will be with this categorization. I looked on the webpage and couldn’t find the definitions. Perhaps you can put something more meaningful in the table to help readers understand this categorization. Even a footnote indicating which direction the scale goes in terms of severity would be helpful, at the least. Added as a third table and link updated, thank-you for bringing this to our attention.

6. Theme 1. You first theme could be worded more strongly and more consistently. I believe the authors are making the point that parents’ knowledge and experience and perspective should be acknowledged and integrated into the care process at the EOL. To say they are “relevant” is a bit unclear and understated. Immediately following the list on page 8, you change the wording to “must be taken into account,” which is better, stronger, and perhaps could be used in your 2 previous lists of themes.

7. Theme 3. What do you want to say about the variability? Just that it exists? Your first two theme are statements that can be acted upon. This theme is a little vague. Is the variability bad? Should it be reduced? Should improvements be made to provide more communication continuity?

8. Theme 5. Again, not sure what your point is on this final theme. Can you restate it to be actionable? To just say, “experiencing the death...” is not an active voice sentence and may be difficult for clinicians to act upon. In general, all your themes should be

actionable or at least informative statements.

We have reviewed and revised the theme subtitles, including some further illustrative quotes in these subtitles. We have also amended the content and quotes within each theme to help to ensure that the key messages from each theme are clearer.

9. Summary. Once themes are clarified a bit, the summary might be improved and

expanded. Also, integrate very specific recommendations into the recommendations

that relates to your stated themes. We have revised the discussion and conclusion which now relate more closely to the stated themes.

The paper is well-written but could use a copy-editor as there are a few grammatical errors,

e.g., "Learning from the experiences and perceptions of families is imperative in order to

improving practice." We have tried to correct the grammatical errors and ensure consistent use of abbreviations.

Reviewer: 2

General comments:

The paper would benefit from structural review. It would be helpful to have sections clearly delineated. introduction Aim, method, results, conclusion. We have followed the authors guidelines for BMJOpen to structure our paper.

Abstract:

I think the abstract could do with better structure and be more concise. I also wanted to read more about the method in the abstract. e.g. results: Who were the interviews with? How many mums? How many dads? Added in to the abstract

ACP needs to be in brackets after it is written in full. Corrected, thank-you.

Introduction:

Living with uncertainty could be expanded to include the care that parents provide and the impact that this has on family life, siblings etc. and the role of parents in providing sometimes very technical care for their children. We agree but this was not directly relevant to the aims of the paper.

Does the PIC involve or have access to palliative care services? And does this have any impact on the care that families receive? Does Birmingham have a palliative care team? I think this is key issue - not to say that all cases should involve palliative care but certainly if children have LLI's then are palliative care not involved at some level? The children in the study generally did not have access to palliative care services, because they were so limited both in the community and in the hospital setting, and because the idea of palliative care was far from the aims of the treatments that they were receiving. We have provided further details.

Where/who were referrals for family inclusion in the research received from? We have outlined the recruitment process in more detail

Refer to table for the type of LLI - include a list as an appendix? Added as a third table

59 parents of how many children? Clarified

I am not sure of the ethics and what was included in the ethical approval. We have included the study protocol which details the ethical concerns in as a supplementary file..

If parents were recruited who had lost a child in the last 12 months, it is possible that some were newly bereaved - their experience would possibly be impacted by this compared to someone who has been bereaved for nearly a year.

There are two separate groups here. Identifying people prospectively - it is well-documented in the literature that it may impact on findings if people are still receiving care compared to retrospectively. Maybe the two should be identified separately? We chose not to identify these as two separate groups since the start of recruitment to the study did not represent any change in the care that the children received in PIC. The retrospective approach was included to try to enhance recruitment. We anticipated being able to recruit more parents than we did overall. We have highlighted these challenges, and also referred to the differences in the length of time since bereavement as a potential limitation of the study

Was the interview schedule piloted? Yes, detail added

Was any support offered to parents post-interview if respondents were upset or distressed? or even just to check if they were ok after being interviewed and talking about a distressing experience. Details are included in the study protocol

P8. Line 31 - Loosing control when their child is admitted is a key issue for parents and this could be included here. Added

P16. Line 3 - There is literature in children's palliative care about these issues and it would be helpful for the authors to acknowledge and refer to this. Home as a preference - could the authors say more as to why children are dying in PIC? were parents offered a choice to take their children home? Is support available if they wanted this? There is very little evidence to support place of death as the preferred place for children and families, and in fact the parents we interviewed described benefits to remaining on PIC for the end of life care of their child, which we have described further.

Key words: add the UK 'paediatrics' Added, thank-you

### VERSION 2 – REVIEW

<b>REVIEWER</b>	Elizabeth L Ciemins AMGA USA
<b>REVIEW RETURNED</b>	07-Mar-2019

<b>GENERAL COMMENTS</b>	The authors have adequately addressed all of my suggestions and concerns. Nicely done.
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<b>REVIEWER</b>	Julie Ling European Association for Palliative Care Belgium
<b>REVIEW RETURNED</b>	20-Feb-2019

<b>GENERAL COMMENTS</b>	I am reviewing this paper for the second time and wonder if the authors could please address some of the issues I raised.
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	<p>I think there still needs to be more information in the methods - was the questionnaire piloted? In the results - duration and location of interviews (phone, home, PICU)? I am still not sure that you have addressed the issue of the mix of prospective and retrospective interviews... there are some real differences in what parents will say before and after their child has died... maybe it needs to be more explicitly addressed in the limitations? Was support available for parents during or after the interviews... they are a vulnerable population - please be explicit. I asked this in my original review... were any of the families receiving palliative care? Was taking their child home with support an option? Do the authors think, that following their study that there needs to be better synergy between PICU and palliative care?</p>
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### VERSION 2 – AUTHOR RESPONSE

I think there still needs to be more information in the methods - was the questionnaire piloted? Yes the interview was piloted with the PPI parents, as detailed on p. 5/6.

In the results - duration and location of interviews (phone, home, PICU)?

I am still not sure that you have addressed the issue of the mix of prospective and retrospective interviews... there are some real differences in what parents will say before and after their child has died... maybe it needs to be more explicitly addressed in the limitations? All of the parents who were interviewed were bereaved, none were interviewed before their child had died, as outlined in the inclusion criteria. Perhaps our use of the term “prospectively identified” has caused some confusion. We have revised the relevant paragraph in Recruitment on page 5 and hope is now clearer.

Was support available for parents during or after the interviews... they are a vulnerable population - please be explicit. Full details of the ethical concerns raised in the study are provided in the study protocol, which we have submitted as a supplementary file. We developed a distress protocol for use during the interviews, and all participants had access to support from the hospital bereavement team. We have added a sentence on page 6 to detail this.

I asked this in my original review... were any of the families receiving palliative care? Was taking their child home with support an option? The children in the study generally did not have access to palliative care services, because they were so limited both in the community and in the hospital setting, and because the idea of palliative care was far from the aims of the treatments that they were receiving. We have provided further details in Study Setting on page 5.

Do the authors think, that following their study that there needs to be better synergy between PICU and palliative care? This is an important point and one we have considered in the discussion – on pages 16 and 17 we have tried to suggest ways in which the concept of palliative care, referral to specialist paediatric palliative care services and the introduction of end of life care decision-making could be improved in practice, while acknowledging how difficult this can be for families and healthcare professionals.