Author’s response to reviews

Title: Parental decision-making following a prenatal diagnosis that is lethal, life-limiting, or has long term implications for the future child and family: a meta-synthesis of qualitative literature

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Reply to reviewers’ comments

Ms. Ref. No.:

Title: Parental decision-making following lethal or life-limiting prenatal diagnosis: a meta-synthesis of qualitative literature, BMC Medical Ethics

Reviewer 1’s comments: Lisa Crowe

1. I have read your paper with great interest. Unfortunately, I cannot recommend it for publication in it's current form. "Life limiting" - you use this term throughout but make no attempt to define it. This term will mean very different things to different people, and having a definition as to how you have defined it for the purpose of this paper is important. I would also be interested in how your definition marries up to how the papers you have discussed define life limiting. For example, you have included papers that discuss Down Syndrome, and I know from my own research that this is a condition with conflicting opinions as to whether a TOPFA can even be justified under Clause E, suggesting differing opinions as to whether it is life limiting. I am not saying it is incorrect to use those papers, just to be clear in how you interpret life limiting.

Our reply: Thank you for your comment and suggestion. This issue was also noted by the second reviewer and for this reason we decided to amend references to lethal or life limiting within the text to ‘lethal, life-limiting to severely debilitating’. We have also amended the title of the paper accordingly.
The title of the paper is now ‘Parental decision-making following a prenatal diagnosis that is lethal, life-limiting, or has long term implications for the future child and family: a meta-synthesis of qualitative literature.’

References to lethal or life-limiting have been changed throughout the paper to ‘lethal, life-limiting, or severely debilitating diagnosis.

2. Literature review - you have some quite dated references, for example line 29 you cite a 33 year old reference to make your point here. Off the top of my head, you could have Cited Lotto's papers here (2016, 2017).

Our reply: We would like to thank the reviewer for drawing our attention to this issue. We were unable to find the reference on line 29 on any of the pages within the manuscript. On page 5, line 15 there is a reference which we believed you may be referring to.

In line with your request, we have added in more recent studies to support this older paper, including one of Lotto’s papers (2016). We also added in additional references (3,8,11,12, & 24) to support older references within the introduction.

3. Page 5 Line 35 - this statement about identification of several factors appears to contradict your sentence on line 19 where you say little is know about factors influencing parental decision making.

Our reply: We are sorry for this contradictory information. We have revised the text accordingly. The text on page 5, line 8 now reads “However, no concise review detailing all factors influencing parental decision-making when parents are faced with the option to continue or terminate a pregnancy affected by lethal, life-limiting, or severely debilitating prenatal diagnosis has been provided; hence there is a need for further clarification to ensure adequate support is provided during this period.”

4. Page 5 - Reference 17 is not included in the bibliography so I am unable to comment on it.

Our reply: We would like to apologise for this omission. We have now included the reference, which has changed in number due to additional references being added.

5. Page 6 Line 1-7. There are studies outside of TOPFA literature that could be alluded to here about healthcare teams supporting parents, (Butterfly Project on multiple loss) as well as papers within the TOPFA literature (again off the top of my head Lotto, 2016, 2017, and possible Fisher various years). Upon having a look at the systematic reviews you cite on line 31, Lou et al., discusses examples of how health professionals can acknowledging them as parents, and acknowledging the child, which I would suggest are examples of how healthcare teams can support parents.

Our reply: Thank you for drawing our attention to these papers and studies. The initial sentence has been re-phrased to highlight that there are no current guidelines for teams in supporting parents in making decisions, rather than in supporting parents during this time.

The text on page 6 now reads “Whilst studies provide examples of factors influencing parental decision-making, they do not infer how maternal healthcare teams working with parents should best support them in making decisions, leaving important recommendations to better prepare maternal healthcare teams for this supportive role.”

6. Page 7 line 18 - you state that Lafarge et. al., systematic review is about the experience of TOPFA rather than the decision making process, but they have 2 sections discussing decision making in their paper 'Ambivalence' and 'losing and regaining choice'.

Our reply: Thank you for raising this issue. Lafarge considered decision making within the overall experience of parents. Studies weren’t excluded from the review if they did not consider decision-making, and as such a lot of papers included by Lafarge were ineligible for inclusion in our study.

Lafarge’s review also considered decision-making after baby was lost/terminated, whereas our study focussed solely on decision-making during pregnancy. However, we have amended text as shown to clarify this:

The text (on page 7, line 3) now reads as follows: “Lafarge et al.’s meta-ethnography22 reviewed 14 English-language studies which addressed women’s experiences of choosing to terminate a pregnancy, following prenatal diagnosis. They identified that women expressed feelings of guilt and hopelessness when deciding to end their pregnancy. However, they also considered the baby’s potential quality of life as well as the impact on them and their wider family. The review focussed on the women’s experience of the termination process, addressing women’s decision-making in two sub-themes, whilst the remaining ten sub-themes focussed on the experience of women. Studies were not excluded from this review if they did not consider parental decision-making.”
7. Page 8 - I am not an expert in systematic reviews by any means, but I am wondering if ASSIA should have been included in the database searching.

Thank you for raising this issue. We developed our search strategy in line with expertise within the team and guidance by the University’s librarians and believed that our search strategy was comprehensive. We searched a total of 13 databases. Furthermore, MEDLINE alone is known to consistently generate the most relevant ‘hits’. However, after reviewing the ASSIA database, it appears that the comprehensive list of other databases utilised within the review covers journals listed in ASSIA.

8. Page 9 - I think it is reasonable to state which author did what regarding coding. Themes: You have lots of sub themes. To me, the discussion feels a bit fragmented and doesn't flow. I am not sure why you haven't focused on less sub themes in more depth. Your subthemes are also unbalanced - Theme 1 you have 5, theme 2 you have 2 theme 3 you have 3. you have also referenced an excluded study in your results (reference 31 - page 9 line 23 you say it is excluded, page 10 line 59 you reference it).

We agree with the reviewer and decided to indicate who the coders of the complete data set were. A comprehensive discussion of which authors were involved in the coding has now been added to Page 9. However, we do not agree with the need for an equal number of subthemes having to contribute to themes. We described the analytic process in the text and these are the subthemes and associated themes we identified and agreed on within the research team.

The excluded reference was added in error and should have reflected a quote from an included study. The referencing has now been addressed, and only included studies are referenced in results.

A further paragraph has been added to the meta-synthesis section on page 9, stating “The synthesis was implemented in three stages of analysis described by Thomas and Harden27. Stage 1 of the process involved both the first author (CB) and second author (DMS) reading each of the included studies a number of times, before independently implementing line-by-line coding within the texts of the results or findings sections of each paper in order to develop codes describing the meaning or content. This process was repeated for each included study, enabling the translation of concepts between the studies, as well as identification of new themes. This enabled a higher level of analysis to be conducted in stage 2, when both researchers grouped together codes to form descriptive themes apparent across and between papers. The third and final analytical stage of interpreting consistent and inconsistent themes within papers enabled the presentation of analytical themes and sub-themes. This final stage relied on researcher inference and judgement regarding the meaning behind each code. To ensure reliability, both researchers coded and grouped themes independently, before discussing their views with each other and forming the final themes which were then discussed with and agreed by all authors.”
9. Themes: You have put a couple of quotes in that seem a bit random as there is not explanation before or after them (for example, page 14 line 54, and page 16 line 51).

In line with guidance on the presentation of qualitative data (see Creswell, 2007), we decided to embed shorter quotes within the text to support the theme these quotes were linked to.

10. Page 17 line 38-44 - this feels like a bit of a repeat of an earlier point. If it is to stand alone separate, it needs some more unpicking.

Theme 3, ‘a life worth living’, is important in describing how parents considered the factors which would influence their own ability to care for a child with a severe condition, and their own individual life circumstances which may impact this ability. It also describes concerns parents described around the quality of their unborn child’s life.

Whilst the themes presented in theme 3 do influence, and are influenced, by others presented in theme 1 and 2 (as highlighted by Figure 2), we believe they are distinct and present a clear reflection of the themes emerging from the data available.

11. Page 18 line 31 - I am not convinced you have produced a novel finding. For example "Another pregnancy is generally soothing but can be bitter-sweet, another illustration of ambivalence. A new pregnancy is seen as a leap of faith requiring courage and determination, but which is eventually rewarding: "no guts, no glory" " - This is a direct quote from Lafarge et al., who you reference. I may be incorrect as I have not had time within the time frame of completing this reference to go back to some of the literature and check, but I am not convinced this is novel.

The novel findings relate to the parent’s perception of their own ability to conceive again (i.e. age), rather than their doubts about whether they could emotionally prepare themselves for a further pregnancy.

Lafarge’s ‘surviving the ordeal’ theme does not make reference to women’s understanding of whether they can physically have another baby again, or whether they have lost an opportunity (potentially their only opportunity). It details how women prepare themselves for potential setbacks when pregnant following initial loss.

Page 19, line 7 reads “Unlike previous reviews which reflect on how parents prepare themselves for potential setbacks in future pregnancies, this review describes how parents also reflecting on their ability to conceive another child in the future and their own personal factors which may influence this, such as advancing age.”
12. My knowledge of psychological theory is limited, however, I would have expected any theoretical discussion to come much earlier in the paper, with the discussion used to demonstrate how the findings support the theoretical argument presented earlier.

Following your recommendation, more theoretical background has been added to intro from discussion section. We feel the discussion section provides a reflection on this prior knowledge on page 19, line 21.

“Studies have examined decision-making as a psychological construct, with normative models, such as Classical Decision-Making Theory50, aiming to rationalise a complex and individual process51. Whilst this model provides insight into how best to approach a decision, it does not necessarily describe how people choose in real-life situations. Descriptive models, such as Prospect Theory51, provide insight into how individuals make real-life choices. Descriptive models suggest that schemata, encompassing prior life experiences, are drawn on to make decisions52, considering potential outcomes of each option available to them, and the likelihood of each option occurring.52 According to Emotion-Imbued Choice Theory53, individuals imagine their perceived emotional response to each available outcome as a way of mediating decisions. Individually salient impacting factors, such as culture54, current emotional state55, delivery of information56 and time constraints57, must be considered when supporting parents making decisions. “ has been added to introduction on page 7, line 21.

13. A). Page 20 - line 18 - I am not convinced you have included all papers anyway, but I will again concede I may be incorrect as I have not had the opportunity in the time frame to consult the literature to check my thoughts.

Thank you for raising this issue. We have previously responded to concerns regarding the inclusion of the ASSIA database. Hopefully, given the overlap between this database and others included in the review, this alleviates some fears regarding any missing papers. We did search 13 databases.

It is also important to note that our inclusion criteria focussed solely on papers which considered decision-making during the pregnancy, which opposes a number of papers included in Lou and Lafarge’s reviews.

We are confident that we have included relevant papers sourced through stringent searches and inclusion criteria.
B). Page 21 line 1-10. """"All staff involved in the care of a woman or couple facing a possible termination of pregnancy must adopt a non-directive, non-judgemental and supportive approach" This is a direct quote from the Royal College of Obstetric and Gynaecology guidelines for professionals supporting parents undergoing TOPFA. So while I agree with what you say in that care teams should be aware of parents' beliefs etc, again, this is not a new conclusion and something already stated in professional guidelines.

The RCOG guidelines described do state this recommendation. However, this does not advise care-teams to avoid pre-emptive considerations or rely on their experience with other patients. The guidelines also do not provide suggestions that staff should be mindful and aware of factors which may be important to the parents, such as advancing age, religious beliefs, personal attachment to fetus, as we have discussed in our findings.

14. References: Some more up to date references needed. Nothing against older references, but if you want to cite them, I would include an up to date one wherever possible to show the continued relevance of this discussion.

We have added in some newer references to introduction. However, some older references relating to the established decision-making theory remain as they provide the most relevant references for this area of research.

15. I would have expected to see some references in your paper - the Royal College of Obstetrics and Gynaecology for example, FASP guidelines, as well as some authors (Robyn Lotto 2016, 2017 in particular I would have expected to see - one of her papers discussed parents, the other is professionals). References 64> don't appear to have been cited in the paper?

We have added in RCOG ‘Perinatal Management of Pregnant Women at the Threshold of Infant Viability -the Obstetric Perspective’ and Lotto’s (2016) paper to the introductory text.

FASP guidelines do not talk about decision-making or supporting parents’ decision-making. As such, we have not included them within the paper.

The latter references you have queried are cited in Table 2. Please see references in text and in the reference list.
Reviewer 2’s Comments: Steven Leuthner, MD, MA

1. Overall, I think this article is a nice paper that gathers qualitative information from many studies of prenatally diagnosed conditions. Having performed prenatal consultation for 24 years now as a neonatologist and palliative care physician, I find many of the authors’ themes to hold true from experience and have been described in my publications as well.

We would like to thank the reviewer for his encouraging comment. We are also pleased that our findings match up with his clinical experience.

2. The most concerning issue I have, however is that I disagree with the title of the paper, and feels it is misleading. My main concern is that I do not agree that the articles they reviewed are all in regards to what one standardly considers a fetal lethal or life-limiting conditions. At least in the US, when we discuss Down Syndrome or myelomeningocele, or cardiac defects, or some level of neurodevelopmental delay - while these are very serious prenatal diagnosis that lead women to make these very same difficult decisions, and while sure they are somewhat life-limiting, most people do not label them as lethal or life-limiting. This is then misleading and can lead us to compare apples to oranges.

We appreciate the reviewer’s comments and decided to change the title in line with his suggestion to:

"Parental decision-making following a prenatal diagnosis that is lethal, life-limiting, or has long term implications for the future child and family: a meta-synthesis of qualitative literature."

3. I do think that the authors’ thematic analysis is true for many if not all of these diseases, but I just don't label them all as lethal or life-limiting. In fact, for the case of a true life-limiting to lethal conditions the hope of some long term survival that requires sibling care, for example is not real. For someone with severe disability it is.

References to lethal or life-limiting have been changed throughout the paper to ‘lethal, life-limiting, or severely debilitating diagnosis

4. The easiest way to rectify this is to change the title to "parental decision-making following a prenatal diagnosis that is lethal, life-limiting, or has long term implications for the future child and family: a meta-synthesis of qualitative literature" - or something like that.

We revised the title accordingly as described above.