Author’s response to reviews

Title: The economic implication of stillbirth: Exploring the intangible costs

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RESPONSE TO REVIEWERS

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Exploring the intangible costs associated with stillbirth: a structured review and synthesis of the evidence
Chidubem B Ogwuulu, Louise J Jackson, Alex EP Heazell and Tracy E Roberts

Dear Editor

The economic implication of stillbirth: Exploring the intangible costs

(Please note a revised title)

Many thanks for inviting us to revise and resubmit our paper. We thank both reviewers for their helpful comments which have added clarity to the paper. We present a response to all comments below. Where appropriate we have edited the manuscript and presented these in track changes.

We look forward to hearing from you.

Yours sincerely

Tracy Roberts on behalf of the co-authors

REVIEWER 1

(1) - This is an interesting study, more because of the cumulated knowledge of psycho-social and health related effects on parents and the wider family who experience a stillbirth, than because of possible economic, so-called 'intangible' costs.

Response: We thank the reviewer for the comments. We have responded to specific queries below.

(2) - The main objectives for the study are described as: i) identify the consequences (of stillbirth) and ii) describe their economic implications (lines 104-107).

The study succeeded more in reaching the first goal than the second. Because after reading the manuscript it is still not clear how to measure economic implications of a stillbirth.

Response: Thank you. We apologise if we have not been clear. Our study aimed to describe the economic implications of the intangible costs of stillbirth. In order to measure it, we will need to know the duration of its effects and how it affects quality of life. However we acknowledged that most of the papers did not explicitly report this (lines 327/328) and believe that the responses to other comments below will also help to clarify this.
(3) - I am not sure why the word 'evidence' is used in the title: you present a synthesis of the evidence... of what? In my view it is a synthesis of the available knowledge about possible effects of a stillbirth. There has been no attempt to quantify the findings, as is mentioned in the discussion (line 403/4)

**Response:** We agree with the reviewer that the title may be misleading and that a synthesis might imply that some data quantification was done. To clarify this, we have rephrased the title to “The economic implications of stillbirth: Exploring the intangible costs”.

(4) - However, strong conclusions are made, that are not fully supported by the data presented. As main strength of this paper is written that it is the first that has explicitly determined the intangible costs of stillbirth. I would be more careful in my conclusion and say that it has identified possible sources of intangible costs.

**Response:** Thank you. We agree that this phrase may be misleading, given that we have not quantified the cost. So we have rephrased the sentence which says 'The main strength of the paper is that it is the first that has explicitly determined the intangible costs of stillbirth' and it now reads 'The main strength of the paper is that it is the first to identify the probable main sources of the intangible costs of stillbirth” (line 385/386)

(5) - And on the next page the intangible costs are described as having a substantial contribution to the total costs. That conclusion cannot be verified, because the tangible costs have not been quantified.

**Response:** We agree with the reviewer that this conclusion is not verifiable. We have amended the phrase “Firstly, the findings justify the rationale for including intangible costs in economic evaluations of stillbirth by revealing their substantial contribution to the total costs” to read “Firstly, the findings justify the rationale for including intangible costs in economic evaluations of stillbirth by revealing their potentially significant contribution to the total costs” (line 411-413)

(6) - The proposed questionnaire needs more explanation, not only regarding its development, but also regarding its goal to capture intangible costs and consequences. How do you propose to use a questionnaire like this? After the 'refinement' you suggest, would the questionnaire be used in a (large-scale) survey or for individual interviews?

**Response:** Thank you for your comment. We have attempted to rectify this by adding some sentences regarding the goal of the proposed questionnaire in the methods section. Thus we have included the sentence “In an attempt to move towards quantifying the evidence collated in the qualitative synthesis, questions are proposed; which could be developed for use in a large scale survey to provide this quantification.” (line 173-175) on page 8.

Also in the discussion, we have clarified that the questionnaire will be in a large scale survey. “However, by proposing questions that could be asked in
prospective large scale surveys the study developed a framework to guide future studies” (line 400/401)

The appendices are informative, maybe you can make more use of them in the main texts.

Response: We will take editorial advice on how much of the appendices, if any, should be included in the main manuscript – but we hope that by clarifying in the methods the goal of the questionnaire, the link to the appendices will be clearer.

REVIEWER 2

Minor Essential Revision (4)

Response: We thank the reviewer for the comments. We have responded to specific queries below.

(1) - Do the figures appear to be genuine, i.e. without evidence of manipulation? The figures are well designed and clearly structured. It seems that figure 1 contains a spelling mistake in the first box (n=49p81).

Response: Thank you for noting this error. We have corrected it.

(2) - Are the discussion and conclusions well balanced and adequately supported by the data?
The discussion is thorough and well balanced. Maybe the remark in line 376 could be dismissed, as there is no need to devalue positive experiences as little as they are. Also, personally I would avoid direct citations from secondary data.

Response: We agree with the reviewer that the phrase is subjective and have removed that phrase.