Author’s response to reviews

Title: Obstetric fistula in low-resource countries: an under-valued and under-studied problem - Systematic review of its incidence, prevalence, and association with stillbirth

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Author’s response to reviews: see over
Dear BMC Pregnancy and Childbirth Editor,

Thank you for the opportunity to review this manuscript. Below, please find our point-by-point responses to the reviewers’ comments.

Reviewer 1

Thank you for this thorough and thoughtful review; we have made the changes you requested, and believe they strengthen the paper.

Major Compulsory Revisions (which the author must respond to before a decision on publication can be reached)
1. Title: I recommend that you alter the title given the largest proportion of your review is dedicated to evaluating the incidence and prevalence of obstetric fistula.

We have revised the title to:
Obstetric fistula in low-resource countries: an under-valued and under-studied problem – Systematic review of its incidence, prevalence, and association with stillbirth

2. Background, pgh 3: Please be more explicit about your rationale for combining the available population and facility-based estimates of OF and the association of OF and stillbirth.

We have added the underlined phrase below (line 81) to explain our reliance on facility-based data for reports of the association between obstetric fistula and stillbirth:

This paper reviews the literature reporting original population-based estimates of OF incidence and prevalence rates in low-resource countries and assesses their precision and risk of bias; in the absence of population-based data linking OF and SB, it also reports facility-based estimates of SB in births that caused OF.

3. Discussion: What are the author’s particular recommendations for improving the availability and quality of data on obstetric fistula based on the systematic review?

We have added this sentence (lines 230-234):

House-to-house studies that systematically enumerated female residents aged 10 and above while actively searching for cases, then confirmed cases by physical exam (and, ideally, offered fistula repair surgery), could be a viable tool for reliably establishing population prevalence in an area. Adding information about birth outcomes could help to better quantify the association between OF and SB.

4. Conclusion: While the conclusion is true, it not directly supported by the particulars of the study. Perhaps you can restructure this somewhat in reference to the rationale of the study (as
Thank you for this suggestion; noting your comment below in response to the BMC-proposed questions, we have revised the conclusion in the body of the article to match that in the abstract and to better align with the particulars of our study (lines 277-281):

In summary, OF remains a significant obstetrical problem in low-resource countries. It is strongly associated with stillbirth, as both are related to obstructed labor in the absence of emergency obstetrical care. Reliable data on OF and associated SB in low-resource countries are lacking, underscoring the relative invisibility of these issues; sound numbers are needed to guide policy and fund responses to these neglected conditions of poverty.

Minor Essential Revisions (such as missing labels on figures, or the wrong use of a term, which the author can be trusted to correct)

1. Some copy-editing will be necessary prior to the manuscript being ready for publication including ensuring proper placement of commas, spacing, etc.

2. Background, pgh 1: I suggest that you revise your reference to prolonged obstructed labor to exclude the comma between prolonged and obstructed.

Done.

3. Background, pgh 2: Your background information on the social sequelae of OF would be improved if you could add a small amount of text tying in the contribution of the stillbirth to the social sequelae.

We added the underlined text (line 57):

Women with OF also suffer significant psychosocial repercussions, including isolation, divorce, loss of social roles, including the role of mother, for those whose infants are stillborn, loss of income, stigmatization, shame and diminished self-esteem.

4. Methods, pgh 1: Please clarify what you mean by no review protocol exists.

We revised this to clarify that we did not register a formal review protocol (as recommended in the PRISMA guidelines) (line 89):

no protocol for the review was registered (see PRISMA item 5).

5. Methods, pgh 1: Please revise your search terms (or supplementary text) S3 to S2 (this is as it is presented in your supplementary file).

We could not find a reference to S3 in the paper. The PRISMA checklist has been labeled Additional file 1, the search terms Additional file 2, and the two tables have been submitted as Additional files 3 and 4 as directed in the authors’ checklist.
6. Methods, pgh 2: Please clarify the sources of grey literature. Were these all sourced from the CAB Global Health Database or have you done a broader search – this is important for readers to understand.

We have clarified that the grey literature we included we obtained through ancestry searches or in response to an all requests (line 95-96).

7. Methods, pgh 3: Please indicate the number of reviewers that worked together in data extraction.

We have indicated that KDC, AKN, and JB performed the data extraction (line 103).

8. Methods, pgh 4: Perhaps you want to address the assessment of overall study quality vs. risk of bias. Did you use any systematic protocol for assessment of study quality?

We assessed risk of bias per the PRISMA checklist (item 12); in retrospect, a tool like STROBE would have been helpful to assess study quality.

9. Table 2: Why did you not include assessment of risk of bias here as you did for Table 1?

In the last paragraph of the Methods section, we stated (revised text underlined; lines 116-117):

For studies reporting the proportion of SB among births that led to OF, we did not perform an explicit assessment of bias, as these estimates were generally not part of the studies’ main objectives. Instead, we commented on factors that might decrease the validity of estimates...

Discretionary Revisions (which are recommendations for improvement but which the author can choose to ignore)

1. Background, pgh 1: Where you state “Underdeveloped pelvic bones are also a risk factor” you may want to consider ‘underdeveloped pelvic bony structure’ as it is the confluence of the bones that are the issue and perhaps exclude the ‘also’ for this statement as you are beginning to talk about risk factors at this point.

Done, and done (line 53).

2. Background, pgh 1: You may consider moving the last few sentences beginning with “In countries where emergency obstetric care . . . .” to the beginning of the last paragraph in the background section for improved flow.

Done (line 59).

Reviewer 2

Minor essential revisions:
1. Page 7, first para: the authors talk about post-partum women.
OF occur usually in the first week post-partum. It would be good to state how many days post-partum these women were.

As shown in Table 1, the time post-partum for the women in these two studies varied between one and twelve months. We have revised the text to read, “1-12 month post-partum women” (line 141-142).

2. Prevalence/incidence of OF is very difficult to establish. That point is made very clear. When population surveys are conducted to make an attempt at establishing prevalence this can be misleading without examination of women. In unpublished studies in Ethiopia, when women are encouraged to come forward for treatment of incontinence, on examination it is often found that about 50% of women who have urinary incontinence the cause is not OF but prolapse of uterus.

The point that physical examination is required to really establish OF needs to be made with greater emphasis.

Thank you.

We have added a line (line 205) stating that, “Thus, physical examination is required to reliably establish OF.”

We have also included discussion of a recently published paper that emphasizes the importance of physical exams (lines 220-227):

“Aft er our review was completed, Maheu-Giroux, et al. published a meta-analysis of DHS and Multiple Indicators Cluster Surveys (MICS) data (grey literature reports) on vaginal fistula (VF) collected between 2005 and 2013 from nineteen sub-Saharan African countries [52]. In this excellent paper based on rigorous analysis using a hierarchical Bayesian approach, the authors’ best estimates of lifetime and point VF prevalence per 1000 women aged 15-49 years in these countries was 3.0 cases (95% credible interval (CrI) 1.3-5.5) and 1.0 case (95% CrI 0.3-2.4), respectively. However, they caution that data from these surveys may include false positive cases, since the surveys are not followed by gold-standard gynecological exams.”

We also inserted the word “published” in line 209.

And finally, we have proposed that active case-finding and physical examination can provide reliable population-based estimates of OF (lines 230-234).