Author's response to reviews

Title: Making stillbirths count, making numbers talk - Issues in data collection for stillbirths

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Version: 5 Date: 16 November 2009

Author's response to reviews: see over
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Version: 3 Date: 13 November 2009

Author's response to reviews: see over
Dear Sir,

According to the Editorial request of October 30th, please find our point-by-point responses (in blue) to the reviewers according to the template we received - as an addition to our original cover letter.

Sincerely,
J.Frederik Frøen

Reviewer 1
Reviewer's report
Title: Making stillbirths count, making numbers talk - Issues in data collection for stillbirths
Version: 1 Date: 6 September 2009
Reviewer: Ingela Rådestad
Reviewer's report:
Making stillbirths count, making numbers talk – Issues in data collection for stillbirths
It is not a minute too early; an international consortium of leading figures in the field jointly formulates ways to go forward. Simply counting stillbirths is the first step to prevention. The article encompasses a gigantical intellectual effort, we who are interested in stillbirth and the future parents are to be congratulated for this achievement.
The article addresses an important problem of interest to a broad audience. The authors present novel arguments and insights into existing work and the piece is well argued and referenced. Logical arguments are put forwards and it is easy to follow the reasoning.
Discretionary revision
I have worked hard to try to find suggestions for refinement. The authors may consider taking a stronger position and point out the most important improvements they want to achieve by implementing the Case ID form. Do the authors want to improve research in this field or do they want to improve quality indicators? Is the main goal to retrieve data for comparing high, middle and low income countries? If the goal is to collect quality indicators, some few factors measured with high quality are probably better than to have the ambition to measure many factors. The Case ID form (Figure 4) is very ambitious, nonparticipation may lessen and sensitivity and specificity in the measurement increase by simplifying. Results may be better if fewer variables are measured.
1) The description of the purpose of the specific design of the form has been clarified. Page 15.
Minor essential revision:
The article is well written. Terminology can be more stringent.” Rich vs. poor countries" can be erased and "high, middle and low income countries" or “high and low resource communities” used consequently.
2) Changes have been made accordingly. The entire manuscript has been extensively reviewed for improved clarity and consistency in the terminology.
Accept after minor essential revision.
Level of interest: An article of importance in its field
Quality of written English: Acceptable
Reviewer 2
Reviewer's report
Title: Making stillbirths count, making numbers talk - Issues in data collection for stillbirths
Version: 1 Date: 12 September 2009
Reviewer: Rachel Haws
Reviewer's report:

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Overall comments
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Overall, this is an extremely important assessment of the current poor state of the world’s data on the compelling yet neglected problem of stillbirth, with broad relevance for epidemiologists, maternal and child health program planners, and clinicians. The paper effectively reviews the multifaceted challenges to obtaining complete stillbirth registration and reporting. This is also the first paper I have seen which proposes a comprehensive system for collecting data on stillbirths globally that is both theoretically implementable in and relevant for low-resource settings, and also flexible enough to capture higher-level and informative data which may be available in higher-resource settings (e.g., placental histopathology, etc). There are some areas of potential overlap in the classification system the authors suggest which could cause confusion, and some difficulties imagining how the cause of stillbirth could be ascertained in the lowest-resource settings at the present time. Still, I largely agree with the logic, organization, and classifications proposed and the framework is a welcome new tool in need of testing. The authors have highlighted some of the logistical and methodological challenges involved in implementing this system globally, as well as the need to adapt the data collected for local program and reporting needs. I would have liked to see actual testing of the classification framework in the settings described in the panels, but the system appears to be relatively nascent and untested. Instead, the panels merely describe the capacity of several lowand middle-income sites to adopt and practice stillbirth ascertainment, including cause of death.

The article is also the only analysis I have seen which systematically assesses the strengths and deficiencies of the ICD-10 definition and classification of stillbirth, and compares the requirements, strengths, and weaknesses of the multiple stillbirth classification systems in existence. The use of these systems has important implications for capturing globally usable and meaningful data on stillbirth incidence and causation, which the authors carefully analyze. The article offers a thoughtful consideration of the potential power of verbal autopsy in settings where stillbirths occur outside of health facilities, and rightly highlights that more efforts are needed to improve the usefulness and accuracy of this tool. The focus on the continuum of pregnancy-associated deaths, with stillbirths included alongside maternal and neonatal morbidity and mortality, is compelling and much needed to galvanize political and programmatic action.

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Major compulsory revisions

1) The authors should be applauded for acknowledging that how a woman, her family, and her caregiver experience a stillbirth (and their overall cultural, practical, and financial importance and meaning) may influence data quality on stillbirth incidence. I was disappointed, however, that there are multiple instances where the abstract concept of “culture” is treated monolithically as a barrier to stillbirth ascertainment (rather than a set of factors shaping how stillbirths are viewed and discussed in a society), with few examples given and a little attention to the “whys” of stillbirths being sensitive events, etc. Designing effective ways of increasing registration and disclosure rates will depend on the local context and meaning of a stillbirth. These issues and “whys” are left largely unexplored in the paper, therefore making the recommendations regarding incentivizing pregnancy registration and survey measurement somewhat simplistic. The anthropological literature on stillbirth and stillbirth disclosure is limited, and I grant that this is not a social science paper. However, because its focus is interdisciplinary, I would encourage the authors to include a few examples from the social science literature that highlight the gravity of both pregnancy and stillbirth in low-resource settings (and even many high-resource settings), and to use caution when considering the abstract notion of “culture” as a barrier when other more tangible and universal concepts and issues apply. Additionally, the grief and emotional aspects of perinatal loss (which may also impact disclosure in surveys) are essentially unaddressed. Both of these issues are germane to the larger issue of the quality of stillbirth data collection, and require investment in operational and formative research to understand the social dynamics of loss, how these dynamics affect stillbirth ascertainment, and whether the accuracy of measurement methods would improve if data collection were more culturally resonant and sensitive. If the section on underreporting could be better organized and nuanced, the paper would offer a more evenhanded assessment of the social, emotional, and setting-specific context in which measurement occurs, and more constructively suggest pragmatic ways of improving data collection (as it is daunting to try to change “culture”).

3) As noted in the original cover letter, we appreciate this thoughtful comment. A new rewritten section specifically addressing all these questions has been added starting on page 9 (last para.) to page 11 (first para.)

Some other specific instances where social science literature could be cited to help the reader understand:

a) P. 6, third para. Please flesh out what you mean by “Cultural resistance to registration can be considerable.”


4) Now modified (page 7), and only an introduction to the section described in 3).


See 3)
need for culturally sensitive data collection methods. Also mention both women’s
grief responses and cultural proscriptions forbidding mourning of a stillbirth in
many settings that could encourage underreporting.

See 3)

d) P. 9 – Reconsider saying that “religious beliefs and cultural habits” are keeping
women from delivering in facilities. Apart from cost and distance issues, rural
women may not deliver in facilities because care in facilities is not culturally
sensitive or perceived as poor quality, or because laboring women are not
permitted to be accompanied in the labor & delivery ward. Policy and bedside
manner changes in facilities, attractive incentives to register pregnancies and
birth, and respect of privacy and women’s and their family’s emotions and
expectations both during pregnancy and surrounding a stillbirth (which are poorly
understood and for which more research is needed in different settings) would
likely make a bigger impact on our ability to count stillbirths than changing
culture.

5) Changes have been made accordingly. Rewritten section on page 11, (2. & 3. para.) See
also 3) – specifically last para. of that section
e) P.9/10, discussion of prenatal registration – important to acknowledge the
possibility that in certain settings, registration incentives may have to be
extremely attractive if they are to override norms of concealing pregnancy and
risks women may perceive they face if their pregnancy becomes public.
Otherwise, the denominator may still be an underestimate (Stokes E et al, BJOG.
2008 Dec;115(13):1641-7). Also interesting that you suggest (without citation)
that women are more likely to report a stillbirth in hopes that it will prevent
recurrence – some studies have found that women keep quiet about stillbirth
because publicizing their loss is thought to increase the likelihood of gossip and
witchcraft, which is associated with recurrence (discussed and cited in Lawn

6) Changes have been made accordingly. Rewritten section from last para. page 11 & first
para. page 12.
f) P. 12 – Please specify and cite some example of cultural practices and clarify
the “lack of understanding” of the importance of the placenta you mention. In
many cultural settings (e.g., West Africa) the placenta is regarded as having
extreme and even spiritual importance, and is to be immediately buried or eaten,
and thus families are unwilling to allow it to be subjected to medical investigation.
Birth attendants may similarly appreciate its symbolic power but not its role in the
pathophysiology of stillbirth or neonatal morbidities – please clarify this in the
text.

7) Changes have been made accordingly. Rewritten section in first para. page 17.

2) The paper is acceptably written and very well-organized, but suboptimal word
choices, typos, and minor punctuation errors occlude the authors’ meaning in
some places. I have suggested some corrections (through about pages 8/9), but I
would recommend that the paper be carefully reviewed to minimize these
grammatical errors and improve the clarity of the evidence and arguments set
forth in the paper.

See 2)

3) Panels – only Panel 4 is referred to in the text. Given that examples of
setting-specific stillbirth registration/measurement are promised in the
introduction and that these illustrate the ways in which stillbirths can be
registered and analyzed in resource-poor settings, please incorporate these
panels into the text. Additionally, some minor editorial changes could harmonize the tone so that these panels are more parallel presentations of the current situation, what is currently measured, and whether the system proposed in the paper is implementable. Has this template been tested/implemented anywhere yet? This would be the strongest evidence to put forth rather than discussing potentialities as these 3 panels do.

8) Reference to panels in the text, and a clarification of their purpose has been added. Page 15, para. 3

Minor essential revisions

1) P. 4 Abstract, first/second line has missing word: “and yet ARE invisible”
2) P. 4 Abstract, last sentence has missing word: “systems, AND effective registration and reporting systems.”
3) P. 5, first para, end of sentence beginning with “Accurately reported stillbirths…”: Change to “the first step toward any improvement.”
4) P. 5, first para.: Change “Millennium goals” to “Millennium Development Goals”
5) P. 5, second para, change “challenges in governance, infrastructure, and workforce in developing countries” to “LIMITED governance, infrastructure, and workforce in low-resource countries”

Changes have been made accordingly for all above, see 2)
6) P. 5, last para: add footnote (same as/similar to footnote 1 on next page?) for “For every 50 publications on unexplained infant death…”
7) P. 6, second para, last sentence has missing word: “to serve AS a sensitive indicator”
8) P. 7, first para under international definition: be more specific about what is meant by “different health care planning than those reporting only third trimester stillbirths.” Meaning not entirely clear.

Changes have been made accordingly. See 2) and page 8
9) P. 7: Citation and preferably more explanation needed for “differences in cultural understanding of pregnancy duration”
10) Changes have been made accordingly on page 8 second para.
10) P. 8: Under identification of stillbirths, should say “Today ninety countries worldwide lack any data on the occurrence of stillbirths.” This is the second instance of this statistic (first on p. 4) – one time may be enough.
11) Deletion has been made accordingly.
11) P. 13 – Even in the most resource-rich settings, the cause of stillbirth remains a mystery in a substantial proportion of cases (which you describe on p. 15). This seems like a very important caveat to consider at the challenges/implications para at the end of the section on p. 15, as you recommend that stillbirths be classified by cause, but even in the best circumstances, these causes may not be documentable.
12) The reason to classify stillbirths consistently across populations even in cases of unexplained stillbirths (versus those of unknown cause) is dealt with on page 21 and figure 6. Attention to this has been added on page 15, second para.
12) P. 14 – would be good to explain why under-diagnosis of malformations occurs in low resource settings – generally because the lack of autopsies and other diagnostic assessments allow identification only of gross external
malformations.

13) This section has been expanded accordingly on page 14 para. 2.

13) P. 17, under “2)” – “hydranENcephaly” misspelled. Changes have been made accordingly. See 2)

14) P. 18/19 – please include a clear distinction between unexplained and unknown (presumably, in unexplained, all possible diagnostic measures were performed but failed to generate a diagnosis/cause of death, whereas in unknown these measures were not taken?)

14) Unexplained stillbirth has been defined and ref. given to the original definition on page 21, 2. para.

15) Figure 1 – 2nd column – this column seems somewhat redundant with figure 5 in terms of both content and organization. This is not inappropriate per se, but is this purposeful or drawn from any particular source? Under the second column, does unknown intrapartum include non-cord fetal hypoxic events – e.g., those linked to uterine hyperstimulation, fetal distress, etc?. In third column, what do you mean by “pregnancy complications? Additionally, is there a reason why breech is not considered in the third column (it clearly falls under intrapartum malpresentation in 2nd column) somewhere near multiples [which is increasingly linked with assisted reproductive technologies in developed countries but not as frequent in low-resource settings]. Also, use of plus sign and divisor sign on arrows seems non-parallel. Meaning still comes across, but multiplication/division or addition/subtraction signs are better pairs.

15) The consistency of causes and conditions across figures is intentional, and drawn from the CODAC classification, as this is one of the two classifications we report to be adhering to the requirements of table 1. This classification is used as an example to present the main components of mortality and to exemplify how expandable layers or levels in a classification enable the same classification to be used in multiple settings. The CODAC classification is referenced both in the text and in figure legends, and the interested reader will find all the subgroups Dr. Haws is requesting and / or asking about discussed and included in that classification while it was seen as beyond the scope of this paper to present these details. In addition, in relation to Dr. Kirby’s comments (See 20) below), it has been intentional by the group of authors not to use CODAC as more than an example and avoid to present it as “the final & recommended” solution. While the group finds CODAC to be useful for exemplifying core concepts, we try to avoid making CODAC the issue of this paper. The figure has been changed according to the comments on use of signs.

16) Figure 2 – please include a color legend for pink (deaths?) and blue (time period?). Also consider moving days of neonatal life and birth/neonatal, which might fit best at the bottom, near or incorporated with pregnancy trimester or pregnancy term (which keeps all time period issues together).

16) Color legend has been added to the figure legend accordingly. Placement of neonatal days have been kept separate from pregnancy weeks intentionally as they are not on the same scale, as commented also by Dr. Kirby (See 24) below). The difference in scale has been kept to provide clarity in presenting the neonatal portion of perinatal mortality even in a figure reduced in scale for print. This portion would be almost invisible if kept in the same scale as pregnancy.

17) Figure 3. For clarity, please reference the discrepancy between stillbirth rates by birthweight and gestation in the figure caption.

17) These are original data for this paper from the Norwegian Medical Birth Registry, as indicated in the figure legend.

18) Figure 4. Template questions: Under delivery/care level, why are general
practitioners/licensed doctors who may not be specialists not included? Under maternal characteristics, what is the value of including terminations on this template, given that in most settings (especially where induced abortion is illegal), this outcome is hugely underreported (I appreciate the importance of knowing this discussed on P. 14, but think that its accuracy very importantly depends on setting and on how the information is elucidated)? Under quality of care, would it be feasible to include a box for “month when antenatal visits began”? In many settings, quality of care may be good but starts in the 7th or 8th month.

18) The form is updated according to the comments on timing of antenatal care and attendance at delivery. We do appreciate the problem of underreporting of abortions, but as discussed in the paper, the optimal will be to include this information, and we believe it should be included for the same reasons of consistency and setting of goals as we discuss in the paper.

19) Panel 2. Change the tense “The hospital now moves towards introducing…” to “The hospital is moving toward introducing…”

20) Panel 3. The first-person voice (“we”) throughout this panel breaks from the tone of the rest of the article. Consider changing to third-person.

21) Panel 4. The first-person voice (“we”) throughout this panel breaks from the tone of the rest of the article. Consider changing to third-person. Also important to note that a disadvantage of this system is that while it helpfully distinguishes between fresh and macerated SBs, diagnostic data to determine cause of death are not generally available.

Changes to all above have been made accordingly. See 2)

Discretionary revisions

1) Suggest use of “low- and middle-income”/”high-resource” or “low resource”/”high-resource” rather than “developing”/”developed” (e.g., “developing regions” p. 8)

Changes have been made accordingly. See 2)

2) Figure 5. Just a technical question - does verbal autopsy allow for multiple contributing conditions, as you helpfully suggest be included in the template (Figure 4)?

19) The technique of verbal autopsy can definitely be enabled to identify significant conditions of pregnancy in addition to concluding with one probable main cause of death (although not all available tools may have this today). Gathering not only the cause of death, but also the scenario in which it happened (the associated conditions) is probably a much stronger tool to identify improvement opportunities than the cause alone. This is discussed by the authors of the CODAC classification in their BMC Pregnancy and Childbirth paper of 2009.

3) Throughout the paper, the authors suggest areas for future/needed research. For digestibility and clarity for readers, consider extracting and putting these into a panel to highlight research gaps and priorities (e.g., need to investigate variations in local meaning and management of stillbirth and relationship to ability to capture stillbirths, testing effectiveness of incentivization of pregnancy registration, feasibility and accuracy of placental examination—possibly by birth attendants—in low-resource settings, improvement of verbal autopsy strategies, testing of classification systems and impact on stillbirth prevention/rates).

Please see comments in our original cover letter.
Reviewer 3
Reviewer's report
Title: Making stillbirths count, making numbers talk - Issues in data collection for stillbirths
Version: 1 Date: 21 September 2009
Reviewer: Russell Kirby

Reviewer's report:
In this paper the authors provide a rationale for why stillbirths need to count worldwide. The paper is a review article that attempts to summarize the information on timing and circumstances of death, associated conditions, and underlying causes, as well as the utility of a classification system to register stillbirths. The authors start the paper framing the problem of stillbirth in a global scale noting the absence of fetal mortality tracks in the Millennium Development Goals, while citing other relevant publications on the topic. The authors argue that simply counting fetal deaths (stillbirth rate) is insufficient for preventing stillbirths. It is necessary to increase political commitment, financial and human resources, consensus on definitions and reporting mechanisms, and information on the causes and associated conditions. The authors suggest a data set designed to respond to the information needs on the underlying causes of stillbirths that can adapted to the local conditions and different scenarios in developed and developing countries.

Another important aspect of the article is that it brings several important issues to discussion, such as the need for tools that could be implemented in both developed and developing settings while retaining most of the information possible even in scarce resource settings (e.g. use of verbal autopsy and adaptation to local indicators). The information provided in this paper could be used to design further studies in the area of perinatal mortality, classification systems, and prevention programs.

Critique: The authors make a very compelling case for why is critical to count stillbirths, and their underlying causes. They also make evident that a better classification system that can integrated into the current reporting mechanisms and definitions while allowing for continuous improvement and tracking of health care priorities will facilitate this process worldwide.

However, there are still insufficient arguments to support the use and utility of the dataset template (CODAC) they proposed (what makes it the optimal classification). Although, some may argue that this information was detailed in Table 1, it was only implicit in the text. Perhaps, reporting preliminary research findings and comparing to previous studies may strengthen the value of this paper.

20) Please see 15) above. The group of authors agree that there is insufficient evidence to conclude that any existing classification system, including CODAC, is the optimal system. In fact, the manuscript specifically states (Page 19) that “... a perfect all-purpose system is
unthinkable.” We have revised the text referring to the figures to underline that CODAC was only used as an example of the concepts being discussed, page 20, 1. para.

Although the paper was grammatically well written, the style and structure of the manuscript are difficult to follow. There appears to be a section missing between the introduction and the discussion. While the purpose of the paper as described in the introduction is clear, the text that follows doesn’t flow naturally from it. The voice used by the authors is sometimes monotonous and limited description of ideas.

Major required revisions: The authors should revise the text to establish a consistent sequence from the beginning. For instance, the outline presented in the intro is this: “we suggest a dataset to cover core needs in registration and analysis of the different categories of stillbirths with causes and quality indicators. We discuss the specific challenges of stillbirth registration, with emphasis on implementation. We point out gaps that need attention in ICD, and review the qualities of alternative systems that have been tested”. Instead, the order presented in the discussion is: Gaps in ICD, Identification of SB, Datasets, Causes and Conditions to be capture (which really should come before datasets), and Classification. The latter is one of the strongest areas in this paper, particularly the ‘basic virtues’ a system should have. The authors may wish to expand on this section.

21) As indicated in our original cover letter, changes have been made accordingly and the paper restructured. Also see 2)

The authors also identified an important area in need of research - pilot testing improved classification systems, and program evaluation studies utilizing homogenous and inclusive definitions. The main question to be answered is: Does better classification improve the prevention of stillbirths, and improve perinatal outcomes? The summary should be framed to guide researchers toward meeting this need.

22) Changes have been made accordingly on page 23, 2. para.

Tables and Figures:
Table 1 contains a great deal of useful information, but requires better formatting (or typesetting) to achieve clarity. Each of the classifications in the first column should be defined if acronyms are used. The second column heading is not clear to a reader not already familiar with stillbirth classifications.

23) Changes have been made accordingly, in particular in rephrasing the column headings. We trust the BMC editorial services will ensure readability after implementing the BMC typesetting for tables and figures.

Figure 1: at the bottom of the diagram, the endpoint ‘stillbirth mortality’ is redundant. All stillbirths represent mortality. The figure summarizes a great deal of information, but might need better formatting relative to the final printed page.

Changes have been made accordingly, see also 23).

Figure 2: same issue re formatting. Units differ between weeks of gestation and days of neonatal life – could the spacing be made consistent across the figure? Some space could be gained by making the row labels smaller. The notes in text boxes at bottom could also be smaller.

24) Please see 16) above

Figure 3 needs a Y-axis label.

25) Changes have been made accordingly.

References: some of the material not cited in journals may be difficult for readers to access. If any of these items are available online, include URLs and date of
access. Examples include ref 72 and 86, but there are others.

26) We have added the information on accessibility as online items to all references where this is an option.