

PEER REVIEW HISTORY

BMJ Paediatrics Open publishes all reviews undertaken for accepted manuscripts. Reviewers are asked to complete a checklist review form and are provided with free text boxes to elaborate on their assessment. These free text comments are reproduced below.

This paper was submitted to a another journal from Archives of Disease in Childhood but declined for publication following peer review. The authors addressed the reviewers' comments and submitted the revised paper to BMJ Paediatrics Open. The paper was subsequently accepted for publication at BMJ Paediatrics Open.

ARTICLE DETAILS

TITLE (PROVISIONAL)	Comparing regional neonatal mortality rates: the influence of registration of births as live born for birth weight <500g in Taiwan
AUTHORS	Wang, Liang-Yi; Chang, Yu-Shan; Liang, Fu-Wen; Lin, Yung-Chieh; Lin, Yuh-Jyh; Tsung-Hsueh, Lu; Lin, Chyi-Her;

VERSION 1 – REVIEW

REVIEWER	Reviewer name: Smith, Lucy Institution and Country: University of Leicester, Health Science Competing interests: None
REVIEW RETURNED	12-Apr-2019

GENERAL COMMENTS	<p>This is a very interesting and important area and the contribution of these findings from different countries adds to our knowledge of the impact of perviable births on mortality rates. The paper is concise and well written but has some methodological issues that require addressing.</p> <p>The introduction covers the key issues but misses papers that have looked at trends over time in the reporting of periviable births as live or stillborn. A recent Lancet paper(1) looked at this across a wide range of European countries. A more expansive literature review would be helpful here to put this paper into context e.g. (2).</p> <p>The methods do not identify the legal registration requirements which are key to exploring reporting of stillbirths and neonatal deaths at periviable gestations. If there is a gestational age cut-off for stillbirths but not neonatal deaths this will have a major impact on comparisons. Also there needs to be a discussion about ascertainment of stillbirths over time and between areas as this may have changed over the period as research has highlighted the impact of stillbirth on parents at earlier gestations and the need to recognise and monitor stillbirth from an earlier stage. Furthermore a discussion of termination of pregnancy is required. Information on legal limits for termination of pregnancy and inclusion or exclusion of terminations of pregnancy is extremely important as this could impact greatly on the findings (3). This is a fundamental issue to the paper and needs addressing before interpretation of the findings can be made.</p> <p>With regard to the results, exploring these trends over time with just 20 regions is limited. The emphasis is on interpreting the change in variation over time.</p>
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	<p>While there appears to be a reduction in variation over time, the scatterplot suggests the influence of Taitung county on the regression in the first period should be explored. A graphical representation of changes over time by region would be of interest to identify whether changes are uniform across regions or some regions are having a large influence. Further exploration of the influence of data in the regression is required.</p> <p>1. Lancet. 2018 Nov 3;392(10158):1639-1646. doi: 10.1016/S0140-6736(18)31651-9. Epub 2018 Sep 27. Quantifying the burden of stillbirths before 28 weeks of completed gestational age in high-income countries: a population-based study of 19 European countries.</p> <p>2. Paediatr Perinat Epidemiol. 1999 Jul;13(3):278-87. Changes in the registration of stillbirths < 500 g in Canada, 1985-95. Fetal-Infant Mortality Study Group of the Canadian Perinatal Surveillance System.</p> <p>3. CMAJ. 2013 May 14;185(8):E345-51. doi: 10.1503/cmaj.121372. Epub 2013 Apr 8. Determinants of increases in stillbirth rates from 2000 to 2010.</p>
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VERSION 1 – AUTHOR RESPONSE

Reviewer: 1

Comments to the Author

This is a very interesting and important area and the contribution of these findings from different countries adds to our knowledge of the impact of periviable births on mortality rates. The paper is concise and well written but has some methodological issues that require addressing.

1. The introduction covers the key issues but misses papers that have looked at trends over time in the reporting of periviable births as live or stillborn. A recent Lancet paper(1) looked at this across a wide range of European countries. A more expansive literature review would be helpful here to put this paper into context e.g. (2).

Reply: The original manuscript was submitted as 'short report' and could cite only five references. In the revised manuscript we have cited 21 relevant references.

2. The methods do not identify the legal registration requirements which are key to exploring reporting of stillbirths and neonatal deaths at periviable gestations. If there is a gestational age cut-off for stillbirths but not neonatal deaths this will have a major impact on comparisons.

Reply: We have added one paragraph to explain the situation in Taiwan. (the third paragraph in the Introduction, page 3).

3. Also there needs to be a discussion about ascertainment of stillbirths over time and between areas as this may have changed over the period as research has highlighted the impact of stillbirth on parents at earlier gestations and the need to recognise and monitor stillbirth from an earlier stage. Furthermore a discussion of termination of pregnancy is required. Information on legal limits for termination of pregnancy and inclusion or exclusion of terminations of pregnancy is extremely important as this could impact greatly on the findings (3). This is a fundamental issue to the paper and needs addressing before interpretation of the findings can be made.

Reply: This study is not focusing on 'stillbirths'. Furthermore, we did not have more information on the stillbirth on parents at earlier gestations or termination of pregnancy.

4. With regard to the results, exploring these trends over time with just 20 regions is limited. The emphasis is on interpreting the change in variation over time. While there appears to be a reduction in variation over time, the scatterplot suggests the influence of Taitung county on the regression in the first period should be explored. A graphical representation of changes over time by region would be of interest to identify whether changes are uniform across regions or some regions are having a large influence. Further exploration of the influence of data in the regression is required.

Reply: We appreciate reviewer's good comment on this point. We have deleted the analysis of trends over time because of small number of births <500g in many cities/counties. In the revised manuscript we focused on 2015-2016 only and focused on the changes in city/county ranking of neonatal mortality rate including and excluding live births <500g. We also followed the tables presented in series of studies in Canada in the revised manuscript.

VERSION 2 – REVIEW

REVIEWER	Reviewer name: Rachel Hilliam Institution and Country: The Open University, UK Competing interests: None
REVIEW RETURNED	15-Jun-2019

GENERAL COMMENTS	<p>This paper is of interest to the readers of this journal and the authors should be commended for undertaking this review.</p> <p>The authors have edited the paper in light of previous reviewers comments, however there is limited analysis of the data in terms of confounding factors, therefore I feel whilst the paper is expanded it should be a short article rather than a full research paper.</p> <p>The main reason for this is the limitation in terms of other factors which would need to be measured in order to make claims regarding how the rankings have changed when the still births are excluded. There are several reasons, most notably that the change in rankings is mainly due to the large percentages in three regions/cities, however two of these cities have a fairly small number of births and therefore there will be a large natural variation in the percentage of live births from one year to another precisely because the number of births <500g is so small in these two cities.</p> <p>It would be helpful to know how the jump in the number of (still-live) birth which is seen in Table 2 as suddenly for 2015 is distributed across these different regions. If as you claim there is a difference in the reporting, then it is likely this change has taken place in only some of the regions, so identifying how the increase in 2015 was distributed would give some insight into this issue. Again something that could be highlighted in a short article as needing to be done for future research.</p> <p>This is alluded to in the paper, but the point needs to be made more strongly. I realize that it won't be possible to collect data to analyse why these variations take place, but in order to substantiate the claim in the abstract that 'excluding live births <500g is an essential step to ensure a fair comparison of city/country NMR' much more detailed analysis needs to be carried out.</p> <p>What the paper does is highlight there is an issue and a first exploration of where the issue might occur, the article should then should expand on what further research is needed, using the current analysis as a first exploration of the issues. Hence why this is a short article not a full research paper.</p>
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	<p>There are a few typographical errors so careful reading of the paper will help. Perhaps one that is not so easy to spot is the second paragraph of the introduction the calculation $848/217385$ does not agree with the value of 217386 in Table 1, so it would be worth checking these values.</p> <p>With a few tweaks to the claims and results, so this reflects just what the paper is trying to achieve - that is awareness of the issue and the need for further analysis into why there are such variations, this paper will be of interest and sit well as a short article in the journal.</p>
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VERSION 2 – AUTHOR RESPONSE

Reviewer: 1

This paper is of interest to the readers of this journal and the authors should be commended for undertaking this review.

1. The authors have edited the paper in light of previous reviewers comments, however there is limited analysis of the data in terms of confounding factors, therefore I feel whilst the paper is expanded it should be a short article rather than a full research paper.

RESPONSE: We agree that this study was limited by not taking into account of the confounding factors affecting the changes in the ranking of county/city neonatal mortality rates. We have added this limitation in the discussion. Please see the third line in the third paragraph on page 8.

2. The main reason for this is the limitation in terms of other factors which would need to be measured in order to make claims regarding how the rankings have changed when the still births are excluded. There are several reasons, most notably that the change in rankings is mainly due to the large percentages in three regions/cities, however two of these cities have a fairly small number of births and therefore there will be a large natural variation in the percentage of live births from one year to another precisely because the number of births <500g is so small in these two cities.

RESPONSE: The three cities (Taipei City, Kaohsiung City and Taichung City) with large change had large number of births <500g. Please see the last paragraph on page 7.

3. It would be helpful to know how the jump in the number of (still-live) birth which is seen in Table 2 as suddenly for 2015 is distributed across these different regions. If as you claim there is a difference in the reporting, then it is likely this change has taken place in only some of the regions, so identifying how the increase in 2015 was distributed would give some insight into this issue. Again something that could be highlighted in a short article as needing to be done for future research.

RESPONSE: Thank you for this important point. We have identified several cities/counties showing marked increase in number of live births <500g in 2015 or 2016 and suggested future research to discern the possible reasons. Please see the first paragraph on page 8.

4. This is alluded to in the paper, but the point needs to be made more strongly. I realize that it won't be possible to collect data to analyse why these variations take place, but in order to substantiate the claim in the abstract that 'excluding live births <500g is an essential step to ensure a fair comparison of city/country NMR' much more detailed analysis needs to be carried out.

RESPONSE: We have cited the way OECD Stat did and gave a moderate conclusion as “We recommend of presenting city/county NMR using both criteria (with or without minimum threshold of gestation period or birthweight) for better interpretation of the findings of comparisons of city/county NMR.”

5. What the paper does is highlight there is an issue and a first exploration of where the issue might occur, the article should then should expand on what further research is needed, using the current analysis as a first exploration of the issues. Hence why this is a short article not a full research paper.

RESPONSE: We have added suggestions for future researches. Please see the last two sentences in the first paragraph on page 9.

6. There are a few typographical errors so careful reading of the paper will help. Perhaps one that is not so easy to spot is the second paragraph of the introduction the calculation $848/217385$ does not agree with the value of 217386 in Table 1, so it would be worth checking these values.

RESPONSE: We have corrected this typo error. Please see the second line in the second paragraph on page 3.

7. With a few tweaks to the claims and results, so this reflects just what the paper is trying to achieve - that is awareness of the issue and the need for further analysis into why there are such variations, this paper will be of interest and sit well as a short article in the journal.

RESPONSE: We have added suggestions for future researches. Please see the last two sentences in the first paragraph on page 9.