Author’s response to reviews

Title: How do World and European Standard Populations impact Burden of Disease studies? A case study of Disability-Adjusted Life Years in Scotland

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Author’s response to reviews:

Dear editor and reviewers,

Many thanks for devoting your time to considering our manuscript for publication, we very much appreciate it. Also thanks for providing helpful comments which we feel have improved the manuscript. We have provided an itemised response to each of the comments raised, including the actions taken in response to each comment.

We hope the responses and actions taken to the reviewer comments are sufficient for acceptance of the paper for publication in BMC Archives of Public Health. We look forward to hearing from you regarding these revisions. We would be glad to respond to any further questions and comments that you may have.

Yours,

Grant MA Wyper, Ian Grant, Eilidh Fletcher, Gerry McCartney, Colin Fischbacher, Diane Stockton

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Reviewer reports:

Reviewer #1: Thank you for the possibility of reviewing this interesting manuscript.
The results of which are however quite obvious: if you adjust rates to a standardised population, rates will change according to the population adopted. If you adopt a younger population, you will see the rates of diseases and injuries typical of the younger population acquiring more weight, and those of the elderlies decreasing their weight.

Of course, if you choose the ESP2013, which is almost identical to the Scottish population, rates will not change much if compared to the crude rates.

Now, the problem behind this analysis is that ASRs are useful if you compare trends or if you compare different geographies. If you need to compare diseases or injuries by ranking, you better adopt crude rates, if ranking is a way of understanding how prevalent or how important in terms of frequency or burden is a disease or injury.

ASRs should never be used to discuss absolute instances.

Specific comments

Line 41: "As expected…". As expected by whom? It is not clear at this stage why it should be expected. Of course, you later explain that GBD WSP is a younger population and this reduces the DALY rates because in Scotland DALYs are mainly concentrated on the elderly. But this is not clear in the abstract.

Our response: Thank you for this comment. We have changed this by removing “As expected”, its inclusion was out of context (L41-42).

Line 84: "calculations" is repeated twice.

Our response: Thank you for this comment, we have now removed this (L84).

Line 87: I do not agree that international comparability becomes a secondary aim. I agree that subnational estimates are crucial to policymakers. However, comparing our country with neighbouring countries, or to countries with similar socio-economic conditions, or with similar health systems, is also vital to policymakers.

Our response: Thank you for raising this. We do agree this remains very important. I appreciate that the current wording may downplay its importance. Our choice was to consider that the primary aim is to get it correct for the nation, and then to look at comparing. We have changed the structure of this, to make it more of an ordered process, rather than to be upfront about down playing the role of international comparisons. It now reads as “BoD studies are becoming an increasingly popular way to assess national and local population health as a means to influence national and local policy decisions for within-country resource allocation. It is therefore essential that estimates used to set national and local policies are based on the needs of the populations they represent and are a valid reflection of the relative burden of different causes of ill-health and
mortality. Once this assessment has been made then comparability between different locations are other important approaches which can be usefully utilised.” (L85-92).

Line 114-116: Better specify that the aim of the study is to compare crude vs. adjusted, AND adjusted with ESP2013 vs. GBD WSP.

Our response: Thank you for this suggestion. We have included the following sentence to give more information about the aims “This was carried out by comparing crude and age-standardised rates, and assessing differences between age-standardised rates derived using different standard populations (ESP2013 and GDB WSP).” (L117-119).

Line 168. As previously mentioned, if the Scottish burden weights more on the elderly, it is evident that if you standardise the rates to a younger population, the burden will reduce. While ESP2013 practically does not standardised, given that it almost coincides with the Scottish population. I suggest changing throughout the manuscript the tone of the results, which appear to be less surprising than described.

Line 179-181: As previously mentioned, the point is not to compare crude rates vs. ASR, but to compare ASR for different years of geographies.

Line 204: This is obvious once you establish that GBD WSP is a younger population than the Scottish or the ESP2013.

Line 230: This is because the two populations (Scottish and ESP2013) are almost the same.

Line 232: This is because these populations are different.

Line 249-250: When you adopt a standard population, you should then compare results considering these are ASRs, and comparisons make sense if you compare years or locations. In relative terms, not is absolute terms.

Line 297: Of course you need to supplement with crude rates or numbers! There are comparisons which require crude rates or numbers, and others that are facilitated by the use of standardised rates. Each comparison has its own reason. Of course is you want to use ASRs for absolute reasons, it will work only if your standard population is almost identical to your population. But this makes standardisation pointless.

Our response: Thank you for your suggestions. We have dealt with these comments under the one rebuttal, as we feel they all relate to the same underlying issue. We absolutely understand and agree with the points which you have made from an epidemiological perspective, but much of our thinking for this study has been around how epidemiological information is used by both experts and non-experts, and the types of difficulties that may arise. A large justification for this study was to illustrate that it is not possible to carry out everything that is needed using the one method (i.e. crude or age-standardised). Difficulties can arise in examples such as results in
published academic papers (restricted space, or key results hidden in appendices), or presentations that misconstrued messages can arise from. For example, if a study compares change over time, lay messages about the current contributions of health loss would be incorrect, because the main study aim was to compare across time. It is useful to prepare ourselves for such situations, as the focus from the media, or other interested parties may decide to focus on other aspects.

Our contribution is such that enabling rates to be standardised by a population that is closer to some national populations may prevent some of those mixed messages from developing. It is important that the methods are correct (and in this case they still remain correct), but we feel that we also need to be prepared to mitigate against contradictory messages, and if alternative approaches allow us to do this, then we should be looking to use them. We have included this in the abstract conclusion, as it wasn’t currently clear from the abstract (L51-52).

Line 307: This does not enable harmonisation. This simply makes ASR closer to crude rates. Again, crude rates are useful for understanding the absolute weight of a disease burden, while ASRs allow us to understand if, not considering population changes, the burden of a disease or injury or risk factor has changed in time or is different between geographies. Let us take breast cancer. In twenty years the incidence of breast cancer might change. This change might be due to a reduction in risk factors, or to changes in the population structure. If you want to exclude the latter, you standardise. Of course, once you standardise, you will not be able to talk about absolute burden. If your aim is to always compare with proximity situations, of course you would better choose a standard population that is the closest possible to your actual population.

Our response: Thank you for pointing this out. We have reconsidered the inclusion of this sentence and agree that harmonisation was not appropriate here. We have instead supplemented it with sentence “This would limit the potential development of mixed messages, or incorrect conclusions, being drawn by non-experts when using BoD estimates.” We believe this now better reflects our stance with respect to limiting the potential for incorrect conclusions being drawn, particularly by non-experts (L313-315).

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Reviewer #2: The study makes a comparison of the estimated DALYs when different standard populations for the age standardisation are used. It shows that when performing an age standardisation, researchers must usually compare the estimates with the crude rates in order to understand better the results. The paper arises the awareness that the usage of standard reference populations can significantly affect the estimates and in case of the DALYS, their ranking. The paper is well written and presents interesting results.

Comments and suggestions are listed below:

General: I would advise to change the abbreviation of BOD to BoD
Our response: We have made the changes throughout the document to refer to BoD.

Page 2, line 41: As expected, all-cause DALY rates (...) -&gt; here it is not clear why it was expected.

Our response: Thank you for this comment. We have changed this by removing “As expected”, as Reviewer 1 had also pointed it its inclusion was out of context, and not relevant in the results section. (L41-42)

Page 4, line 74: morbidity or mortality -&gt; is here meant morbidity AND mortality?

Our response: Thanks for pointing this out. We have changed this to “and” (L74).

Page 4, lines 93-94: "achieve" used twice here. For better readability, may be use a synonym (eg accomplish).

Our response: We have changed this to “accomplished” for better readability, as per your suggestion (L96).

Page 5, line 103: The primary aim of a BoD study is to identify the impact of health problems in a consistent (...) -&gt; I would change to: the impact of health problems and causes of death

Page 5, line 105: (...) what is currently causing health loss -&gt; I would change to (...) what is currently causing death and health loss.

Our response: Thank for you pointing this out in both instances. We had used ‘health problems’ as a capture all for disability and death, in the context of ‘health loss’ being equivalent to DALYs. We see why this may be misleading as they are not synonymous, so have added in “causes of death” as you have suggested for avoidance of doubt (L105, 108).

Page 6, lines 130, 133 and 139: all-causes and 68 causes of disease/injury -&gt; Is here meant all-cause mortality (all causes of death)? Please, clarify in text.

Our response: We have clarified in all three instances by now making references to all-cause DALYs and DALYs for 68 causes of disease/injury. Thank you for this suggestion, we believe it should be much clearer now that the previous wording (L39, 134, 137, 143).

Page 6, lines 134-135: under 1 years -&gt; Change to: under 1 year.

Page 7, line 164: The number of DALYs lost over the (...) -&gt; Please, delete "lost".
Page 7, lines 172-173: at the end of the first sentence, please insert a reference to Figure 2.

Our response: Thank you for pointing out these issues. We have make the changes as you indicated, and inserted a reference to Figure 2, following the first sentence that it refers to data from (L139, 168, 177).

Page 10, lines 237-238: the sentence "The ranking of conditions also changed (…)" does not make any sense.

Our response: We agree that the current wording was not clear. We have changed this to “The ranking of conditions also changed due to the differences in age-groups weights between the two different standard populations” (L241-242).

Page 10, line 239: (…), such as neonatal disorders, congenital birth defects and sudden infant death syndrome, where the burden is experienced early in the life course, saw slight increases in rate -&gt; This is contradictory to the results, where it is mentioned that these causes have the largest gains in ASR of DALYs (page 9, lines 218-219).

Our response: Thanks for highlighting this. We focused on absolute change here. To avoid contradiction, we included a summary of both absolute and relative. It now reads as “Some causes of disease/injury, such as neonatal disorders, congenital birth defects and sudden infant death syndrome, where the burden is experienced early in the life course, saw slight large relative, but small absolute, increases in rate, such as neonatal disorders, congenital birth defects and sudden infant death syndrome” (L242-245).