Author’s response to reviews

Title: Prioritising the development of severity distributions in Burden of Disease studies for countries in the European region

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Author’s response to reviews:

Dear editor and reviewers,

Thank you for devoting your time to considering our commentary article for publication, it is very much appreciated. Also thanks for providing helpful comments which we feel have improved the clarity of the commentary. 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 commentary 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, Neil Chalmers, Gerry McCartney, Diane Stockton

AOPH-D-19-00284
Prioritising the development of severity distributions in Burden of Disease studies for countries in the European region
Grant MA Wyper, MSc; Ian Grant, PhD; Eilidh Fletcher, PgDip; Neil Chalmers, PhD; Gerry McCartney, MD; Diane Stockton, PhD
Archives of Public Health
Dear Mr Wyper,

Your manuscript "Prioritising the development of severity distributions in Burden of Disease studies for countries in the European region" (AOPH-D-19-00284) has been assessed by our reviewers. Although it is of interest, we are unable to consider it for publication in its current form. The reviewers have raised a number of points which we believe would improve the manuscript and may allow a revised version to be published in Archives of Public Health.

Their reports, together with any other comments, are below. Please also take a moment to check our website at https://www.editorialmanager.com/aoph/ for any additional comments that were saved as attachments.

If you are able to fully address these points, we would encourage you to submit a revised manuscript to Archives of Public Health. Once you have made the necessary corrections, please submit online at:

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I look forward to receiving your revised manuscript soon.

Best wishes,

Brecht Devleesschauwer
Archives of Public Health
https://archpublichealth.biomedcentral.com/

Reviewer reports:

Reviewer #1:
Thank you for adding this topic to the scientific discourse if it comes to Burden of Disease estimates. I really appreciate your work.

Our response – Many thanks for your comments and taking the time to review our article. We are glad that others acknowledge that work can be done to drive improvements for severity distributions.

Nevertheless, I would like to give you some comments or notes on your manuscript:
Row 73: What do you mean with "developing a composite approach"? Do you mean to mix the GBD distributions with national one?

Our response – Thank for you highlighting this sentence, we realise it appears slightly ambiguous on re-read. We have edited this sentence to now read “Researchers from independent national studies have been left with either: using the same approach as the GBD study; or developing their own country-specific severity distributions for all, or a subset of, causes”. The point we were trying to make was that some studies are restricted to using the GBD severity distributions whilst there is a degree as to which other studies are able to work with their own: either for all conditions, or for a subset of conditions. Examples are then discussed in the sentence which follows.

Row 79 - 83: Before removing the "asymptomatic group" from calculating YLDs, the specific disease (and therefore the specific context) has to be taken into account. For some diseases it is plausible to consider a asymptomatic group. For example, "Asymptomatic IHD" describes persons who survived an initial IHD event and are living in an interval without symptoms of AMI, heart failure, or angina" (Moran et al. 2012, p. 321). Thus, even if we are using records (claims data) to identify prevalent IHD cases, it is plausible that a group of patients are in an asymptomatic state and should be removed from YLD estimates. So, I would abstain from generalisation. Please make clear that your suggestion is in the context of your data frame and
As side note, we had a debate about asymptomatic COPD-cases because if we can identify prevalent cases in claims data those have to be mild at minimum (from a medical perspective).

In my opinion, this paragraph should start with a brief discussion about the question, if we can define a asymptomatic group of individuals within the context of a given data frame because that is the important point. And then, you could specify your suggestions.


Row 80: It hast to be clear that asymptomatic equals a state of "no health loss".

Our response – Thank you for your comments. We have taken them both into account and made this a standalone paragraph that has now been restructured. This starts off by separating asymptomatic and symptomatic cases and how this distinction is important. We have discussed our example for cocaine dependence cases in more detail and included more rationale for why this approach can be beneficial.

Row 103 - 104: I do not really understand the benefit of using absolute differences between the highest and lowest weights for prioritising diseases. Do you want to make clear that the difference in weights means an underestimation of YLDs? Because in conclusion this would be the overall topic. The difference between the highest and lowest weight is just an indicator and shows the variation of health loss within a disease. If it comes to a suggestion for prioritising there are far more relevant issues, e.g. data access (individual level), measure of life quality in relationship with diagnosis or health state within surveys (see Burstein et al. 2015) or identification of sequela within surveys or claims data. Those have to be taken into account before starting any analysis. I would argue that the 20 leading diseases concerning the estimation of YLDs are itself a good starting point because they produce the highest burden.

Besides this, I checked the result for migraine and did not come to the same result. Because in my opinion the difference between the highest and lowest weight is 0.441 - 0.000 = 0.441 (with asymptomatic) and 0.441 - 0.223 = 0.218 (without asymptomatic). Please check this.

If you want to stay with this figure I would suggest to be more clear what is the benefit.

Our response – Thank you for your comments. Our approach to look at the range of disability weights reflecting the most extreme scenarios (under-/over-estimates) that may occur e.g. the lower bound could only be achieved if 100% of cases were asymptomatic (or mild) and the upper bound could only be achieved if 100% of cases were severe (or the health state reflective of the highest disability). Although these are largely implausible, we do not have a great understanding of the potential variation and the findings of our PLoS ONE paper (https://doi.org/10.1371/journal.pone.0221026) indicate that we may want to remain open minded upon the range of potential variation.
We’ve opted to use these to identify the potential variation that resulting severity weighted-average disability weights could possibly lie between. Although a simple exercise, we gain some important insights into where we may be uncomfortable with uncertainty and focus efforts (such as opioid use disorders or major depressive disorders) or where we may feel more comfortable using GBD severity distributions because the impact of shifts in health state prevalence will have less of an effect (such as asthma or osteoarthritis), meaning we have potentially less to gain from efforts focused here.

Much of ours and other studies current efforts are based on existing data sources and making use of what data is available. This is a good starting point for any study, but longer-term aims should also consider what can be done for conditions without available data. If we can identify key areas that we are likely to be the most uncertain about which are likely to have a high impact on final results, it encourages us to move towards either changing or influencing the way source data is collected, or the commissioning of new data collections to fill the gap.

We thank you for pointing out the issue with the migraine disability weight. This comes from the fact that we applied the GBD duration to it to standardise its use of disability across the same time period as the other conditions. We have changed this to reflect the non-duration adjusted disability weights (including the migraine medication overuse headache health state), and in-text we have made reference to the fact that duration is another condition-specific factor which uncertainties may arise from.

In the conclusion section, we have add some texts to indicate that data availability often drives the focus. This was missing before, and we thank you for raising this.

Reviewer #2:

Overall I found this commentary very useful and interesting.

I found it relevant to highlight the bias induced by the use of standard severity distribution in the calculation of YLDs at the national level, particularly when it comes to comparing different populations, and in particular to interpret the extent of disability in the most deprived populations, with the risk of increasing socio-economic inequalities if these estimates are used in public health policies. You provide answers to essential questions that any researcher leading a national burden of disease study must ask himself. Finally, you propose a concrete approach to guide action priorities in the development of severity distribution at the national level.

Our response – Many thanks for your comments. We are glad that others value this work and the approach we have suggested.

74: I would have briefly explained what this composite approach consists of, and added a reference (if this approach has already been used in another country).
Our response – Thank for you highlighting this sentence. We provided a response to this as the first response to reviewer 1’s comments, to reflect that we meant that some countries are embarking on more ambitious efforts that others (such as developing severity distributions for lots of causes, where as some are restricted to a handful of causes and need to rely on the GBD severity distributions for the remaining causes).

82: it is interesting to mention the possibility of redefining the severity distribution without asymptomatic cases when the available data only include symptomatic cases, and to highlight the importance of remaining consistent in the choices and definition of health states.

Our response – Thanks for this comment. Our original wording did not reflect the link to symptomatic health states, although implied it, so we have re-worded this sentence to read: “This approach can be achieved if the data captured is a robust and consistent proxy for health states representative of individual’s suffering from disease symptoms.”

94: Even if it is another component, I would have mentioned somewhere the existence of a potential bias induced by the use of the same disability weights, subject to variations in interpretation according to context, values and cultural differences.

Our response – Thanks for this comment, it is an important one to raise, especially when we are suggesting that the range of disability weights can be used to identify priority areas for the development of severity distributions. We have introduced the following sentence before we present Figure 2: “A potential limitation of this approach is that disability weight estimates, and other factors such as duration, are also subject to uncertainties such as differences in culture, social values and healthcare access and effectiveness.”

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