Author’s response to reviews

Title: Walking and weakness in children: a narrative review of gait and functional ambulation in paediatric neuromuscular disease

Authors:
Rachel Kennedy (rachel.kennedy@rch.org.au)
Kate Carroll (kate.carroll@rch.org.au)
Jennifer McGinley (meginley@unimelb.edu.au)
Kade Paterson (kade.paterson@unimelb.edu.au)

Version: 1 Date: 23 Jan 2020

Author’s response to reviews:

To the reviewers and Associate Editor,

Thank you for your time in reviewing and commenting on our paper. We have duly considered all of your suggestions and have amended the paper accordingly. Please refer to our responses to each point below.

In the process of revising our paper, two papers (Henricson E, et al, 2013 and McDonald, CM, et al, 2013) came to light that were very similar to two papers already included in the review in terms of the authors and dates of publication. We have subsequently added the information from these further two papers to the results and Table 1.

Kind regards,
Dr Rachel Kennedy, on behalf of the author group

Reviewer reports:

Reviewer #1: Journal of Foot and Ankle Research
JFAR-D-19-00195

Kids, walking and weakness: A review of gait and functional ambulation in paediatric neuromuscular disease

Overall, this is an interesting narrative review that considers a number of important features related to this topic. Comments are provided below in relation to each of the sections.

Title of paper:
The authors may wish to consider removing the word 'Kids' as it does not offer anything to the literature and it is not consistent in terms of terms - kids and paediatric.
Response:
We have removed the colloquial term “Kids” and replaced it with “Children” in a re-framed title “Walking and weakness in children: a review of gait and functional ambulation in paediatric neuromuscular disease” (Title page, page 1)

Abstract:

The abstract is informative and contains the relevant information that is supported by key words.

Background:

The background section is useful and in particular the opening paragraph sets the scene well to place the topic in context linking NMD and gait and function. The aim of the review is clear and is supported well with figure 1.

Method:

The methods section is relatively short - but is concise, and provides the relevant information required. How far back did the search go back to?

Response:
The literature searches went back to 1946, that is the earliest default date for the databases. This information has been added in the Method section: “Literature was searched in Ovid Medline, Embase and PubMed from 1946 up to and including October 2019” (page 4).

Results:
Good information provided on the studies included (page 4). The subsections of gait speed, six minute walk test, time function tests, other assessments and descriptive measures of gait are useful. Whilst it is traditionally not accepted to have discussion in the results, there is perhaps some information or discussion missing in terms of reporting the papers but then not offering a summative viewpoint - either small sample, inappropriate methods used (not long enough testing - metre length etc). The authors may wish to review this approach a little to sharpen up the content adding a critical component to the narrative.

Response:

We agree that while we had presented the findings of our review, this section lacked some summative viewpoints and some critical appraisal. To add clarity, we have revised the section heading to “Results and Discussion”. As the reviewer points out, this is not the traditional format but we feel it fits well in the narrative of this manuscript. In order to provide further critical appraisal to the narrative we have made a number of amendments throughout the Results and Discussion section, including:

“Studies included” the number of papers in each diagnostic group, the median age of the combined studies and the range of sample sizes (page 4)

“Gait speed” detailed the study assessment methodologies and clarification of normalisation (page 5)
“Six-minute walk test” added detail about the walkway lengths and the effect of different walkway lengths on final six-minute walk distance (page 5); added detail of cohort sizes and MCID/MDC (page 6)

“Timed function tests – 10 metre walk/run” added that this test is a common predictor of disease (page 6)

The clinical implications and future directions section is useful, however the authors should provide a separate conclusion.

Response:
A conclusion section has been added (page 9).

Reviewer #2: This manuscript presents a narrative review of the Kids, walking and weakness: a review of gait and functional ambulation in paediatric neuromuscular disease. The manuscript is poorly well written and it is unclear what this review adds to what is already known and have been published earlier. No clear research question seems to be formulated, the conclusions are unclear and other major concerns with this manuscript that make it not acceptable for publication.

Response:
Thank you for your time and comments. We note the reviewer’s concerns and have attempted to address each of these in response to the specific comments below.

My specific comments are stated below. Overall, several important issues need to be addressed and some are of methodological character which requires a considerable revision of the paper.

1. I recommend formulating a specific research question and performing a meta-analysis to answer the question.

Response:
Thank you for this comment. While we agree that a systematic review and meta-analysis would be useful, we were requested by JFAR to write a narrative review of gait and functional ambulation in paediatric neuromuscular disease, and not a SR and MA. We clearly stated that this is a narrative review on pages 4 (“Method”) and 9 (“Conclusion”) in our manuscript. We agree that SR and MA would be an interesting contribution to the field, and that our narrative review may provide the impetus for a more analytical approach. As such, we have added the following to our future directions section: “Limitations including the heterogeneity of the study populations and differences in assessment protocols precluded meta-analysis of these studies. A more analytical systematic review might provide further insights.” (page 9).

3. Methods: Was the study registered at PROSPERO?

Response:
Please see our response to comment #1 above. As noted, this is an invited narrative review and therefore PROSPERO registration is not applicable.
4. Methods - literature search and selection: Please outline the exact search string or provide an appendix with the search strategy with specific search outcomes for each search and combinations.

Response:
We agree that this is a useful suggestion. In response, we have now included our search strategy as a Supplementary Material. Please see this additional document attached.

5. Methods - literature search and selection: Did you restrict study selection on any language?

Response:
Studies were limited to English language only due to the authors being native English speakers. We have subsequently re-run the search removing English language as a limiter and found only one non-English paper in Portuguese that reported four case studies of juvenile Pompe disease. From the English language abstract, this paper is unlikely to have been included in this review. We appreciate that limiting our search to English is a possible limitation of this review.

6. Methods. The authors have not performed a systematic review, according to international standards PRISMA Guidelines, so they do not provide specific numerical data.

Response:
Please see our response to comment 1 above that this study is an invited narrative review and not a systematic review. Accordingly, we have provided summary numerical data only in Table 1.

Results: Please, provide clear results and describe them. Use appropriate statistics.

Response:
Thank you for this feedback. The summary results of the studies included in our narrative review are provided in Table 1, and described in detail in pages 4 to 9. As this is a narrative review, no further inferential statistics have been conducted.”

Within your discussion, outline your results, discuss their novelty and their application to practice.

Response:
Our results are presented in Table 1 and discussed on pages 4 to 9. We have provided additional discussion regarding their novelty and application to practice and have added the following to our results and discussion section:

“Studies included” the number of papers in each diagnostic group, the median age of the combined studies and the range of sample sizes (page 4)

“Gait speed” detailed the study assessment methodologies and clarification of normalisation (page 5)
“Six-minute walk test” added detail about the walkway lengths and the effect of different walkway lengths on final six-minute walk distance (page 5); added detail of cohort sizes and MCID/MDC (page 6)

“Timed function tests – 10 metre walk/run” added that this test is a common predictor of disease (page 6)
Also, please include conclusions section in order to reflect only the study findings.

Response:
A conclusion section has been added as advised (page 9).

Associate Editor:
Thank you for developing this manuscript on request from the editorial team for our upcoming special edition. It creates an interesting dialogue for clinicians to consider the interface of gait and physical function of children that have neurological conditions. I note the comments of brevity of methods and while the intent of this invitation was for narrative or editorial discourse, additional information within the methods would assist the reader to understand the sources of literature included within the review.

Response:
Thank you. We have now provided additional information including the search year range, detailed search exclusion criteria, and have added our search strategy as Supplementary Material.

I would also encourage the authors to consider amending:
1. Format of journal includes a conclusion, please review the inclusion in your article.

Response:
A conclusion section has been added as advised (page 9).

2. Availability of data and materials – amend as not all articles may be available on the internet, but not publicly available unless you have a subscription. You may consider a statement such as: all articles are in accessible locations such as article repositories or websites.

Response:
This statement has been amended on page 10.

3. Have the authors considered the different in versions of the some of the assessments described given their difference (ie: BOT versus BOT-2)?

Response:
Thank you for pointing out the differences between the BOT and BOT-2, a good pickup. Only the BOT-2 was included in the reviewed papers. Therefore we have clarified this throughout the manuscript with the term “BOT-2” replacing all references to “BOT”.

4. Checking the reference list for formatting errors (full rather than abbreviated journal title) or missing information. In particular, article numbers: 2, 4, 5, 12, 13, 15, 24, 26, 31, 32, 41, 64, 65, 68, 70,

Response:
All references have been checked and formatted according to the journal specifications.

Additional references: