Author’s response to reviews

Title: Effectiveness of therapeutic footwear for children: A systematic review

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Author’s response to reviews:

Response to reviewer reports

The Authors appreciate the time it has taken to review our manuscript. A clean untracked version is presented for ease of reviewing. All changes made to the document (inclusive of additions and deletions) are highlighted in a tracked change version located at the end of the untracked submission.

Reviewer 1

Thank you for the opportunity to review the manuscript titled 'Effectiveness of therapeutic footwear for children. A systematic review'. This manuscript is a systematic review of impact of corrective or functional footwear on a range of physical and functional outcomes. It offers an alternative, more specific, review of literature to a related scoping review already published with this journal. The overall aim of the paper is to investigate effectiveness of footwear in aiding mobility impairment in children. In my opinion the review will be of interest to readers and adds to the emerging body of knowledge in this area. The approach to the review is structured and robust and the authors should be commended for tackling the topic. There are, however, some areas that can be improved prior to publication. Some around minor formatting issues but also a few things required clarification, and some attention is needed for layout and presentation of results. At this time I am unable to recommend this manuscript for publication but suspect it can be rectified with some attention to the following issues.

• The authors would like to thank the reviewer for their insightful feedback we have made modifications to the review that we feel address your comments.

General formatting concerns:
1. If you are using a number over 10 it can be numerical. If you have a sentence with a number under 10 and a number over 10, convention allows you use the numerical form for both. Examples where this needs to be addressed are line 232, 389
2. Similarly, when referring to levels of evidence, it is accepted that roman numerals are used (e.g. line 231, Level 11 evidence).
3. Please change '-' to 'to' when discussing age of participants (e.g. 3 to 7 rather than 3-7)
4. When needing to astrix a result, the convention is that the first is allocated *, the second receives a ^, please review a general 'style manual' for further assistance if needed.
5. Line 200 - change would be to were (always past tense).
6. Line 204 - add a comma after geometry.
7. Line 229 - remove the first comma
8. Line 215 - change supplementary to additional

• The text in manuscript and tables has been modified to correct formatting. Tables have been modified to use traditional convention typographical footnote order *, †, ‡, §, |, ¶, #.

Clarification:

1. Line 82, the definition and linking of mobility impairment and the ICF-CY needs reworking.

• Text modified to address these comments.

The following sentence is also ambiguous, does it mean that 2% of the child pop is living with mobility impairment, or that 2% are living with mobility impairment that may require supportive intervention?

• References and text edited to clarify this comment.

2. Please add the PROSPERO registration number.

• Registration number for Prospero has been added.

I would also encourage that the search strategy example be available directly through this article rather than linked to a previous one. Each article should be able to stand alone.

• Search Strategy added as additional file 1

3. Add the word 'trials' behind 'controlled' in line 145.

4. The PICO definitions require attention.
   a. Please change age range to "9 months to 18 years of age" (replace the dash with 'to'),
   b. what definition of 'mobility impairment' was used,
   c. if they ambulated with assistive devices was there any limits on this (e.g. did you include walkers, Zimmer frames, arm or underarm crutches etc.).

• Text modified as requested. Definition of mobility impairment clarified. No limits were applied to assistive aids other than the devices allowed upright ambulation example within the text expanded to highlight this.

5. It is noted that only one reviewer screened the titles and abstracts. This appears to be happening more frequently, but it has limitations. It is confusing to me why two reviewers then screened full text. Can the authors give reasoning for this. On this same theme, only one person extracted data with a second 'corroborating' author review. Can the authors please clarify how this was handled.

6. Were the survey studies (assessed separately using a different tool) also reviewed for quality by two authors? Please clarify this in the manuscript.
• The text had been modified to corroborate that the systematic review followed on from the scoping review. All abstracts that met the criteria for the definition of children’s therapeutic footwear from the scoping review were screened by two reviewers against the eligibility of the current systematic review for full-text assessment. Potential bias from this process has been acknowledged in the limitations of the study.
• Data was extracted from the included studies by one reviewer, the second reviewer then checked the data extracted against the full text of the studies to corroborate the information extracted and to identify any missing data. Quality of the survey was assessed by two reviewers using the survey assessment tool. These points have now been clarified in the text.

Layout and presentation of results:
This is where the most work is required. The authors are to be applauded for attempting to give the reader all the information, but it would be preferred that we get the important information, neatly displayed.

1. Table 1 and Figure 2 need to be combined into one. The figure is nicely presented but it adds nothing to the overall manuscript. I'd also consider adding to this table but simplifying into the following headings: Study, Intervention (CTF or FTF), Modifications/Additions (list extra shanks, lifts etc), N = x (and only report end of study numbers), Condition (simplify and name the condition only e.g. CTEV, Flexible pes planus, Down's syndrome, Typically developing (rather than healthy children), CP - for the one study where there are multiple conditions don't give them numbers, just state 'CP + other' and put a footnote stating a variety of neurological and developmental disorders were present), Duration of study, Gender (report the % of females or males only- 1 column), Age (mean + SD), Mass (mean + SD), Height (mean + SD), BMI (mean + SD) PLUS OUTCOME MEASURES USED (e.g. radiography, physical, functional etc.).

• Table 1 have been modified to reflect these comments. Figure 2 has been added as an additional file.

2. Table 2 can be an additional file

• The authors respectfully disagree with the reviewer, as the primary focus of this review is about therapeutic footwear, we feel the descriptions are integral to the paper and should remain in the main manuscript.

3. Table 3, 4 and 5 are too data heavy - I would, at the least, recommend moving the duration of the study to table 1, and then really consider if this is the best way to display outcomes. Forest plots (without meta-analysis) might visually show the impact better. Please also consider what value is there in some of the outcomes reported. For example, does it help the reader that you're separating skeletal geometry into anterioposterior, lateral etc.? and reporting on outcomes where CTF or FTF aren't used (e.g. Group2 BD and Own footwear, in table 3 + Aboutorabi etc.). All results where CTF or FTF aren't used should be removed completely or tabled in an additional file as extraneous results. I would recommend that several of the outcomes from Zabjek et al. and potentially other studies be tabled as 'repetitive outcomes' in an additional file and the authors decide and report on the key relevant results only (e.g. perhaps review 'eyes closed' at one point in time rather across three). Also do not need to report the type of statistical test used (readers can review the study directly if this level of detail is required).
Tables 3, 4, and 5 have been modified following a number of the reviewer’s suggestions. Duration of study has been moved to table 1, extraneous results from the Chen et al., Ramstrand et al., have been moved to an additional file (Additional file 3). However, the authors felt that it is crucial to present the results of the comparators to showcase the effects of therapeutic footwear (e.g. spatiotemporal) or lack of effect (e.g. skeletal geometry and corrective footwear) in order to inform the reader of the potential benefits or the indifferent effects of therapeutic footwear interventions.

Radiographic outcomes were grouped according to the view to avoid repetitive description and allow clarity to the tables (Radiographic has now been added to the view description in skeletal geometry outcomes).

To reduce the size of the table, but still enable the reader to interpret the statistical results, the column concerning statistical testing has been merged with the statistical result column.

The authors agree with the reviewer; forest plots would indeed showcase trends in outcomes, however, due to the disparity of the conditions and the footwear interventions considered this could also lead to misleading interpretation by the reader.

4. Please also refer readers to the table early in the paragraph when discussing results.

Text modified to reflect this comment.

5. This is opinion but I’m not sure acknowledging the duplicates in the text of the result section is required (line 208). It is cleaner to state 5003 unique articles were yielded, with a further x….

Text modified to reflect this comment.

6. QI outcomes are currently interspersed across results - whilst it’s more conventional to often discuss findings as a separate section it is helpful that you’ve mentioned them directly before the text discussion for each outcome.

Thank you for this helpful comment. The QI are placed in this arrangement to allow the reader to understand how these results apply directly to the various groupings of therapeutic footwear rather than in a combined section of the results.

7. The Discussion is overly long and needs more work. The majority of the discussion currently revisits and re-reports the findings. This is not the point to a discussion. More synthesis of outcomes would direct the reader on what the findings of this review mean to them as practitioners. Should clinicians look to use CTF or FTF? Are they important in mobility impairment and if so, when and how? If I see a child next week with CP and a limb length difference, should I be discussing footwear over orthoses?

Repetition of the results in the discussion has been removed. Analysis and synthesis of the results have chiefly focused on the issues with study design quality and recommendations for improvement in methodology. Some limited cautionary recommendations for clinical practice have been made, with functional footwear suggested as having the potential to offer broader assistance to mobility impairment than corrective footwear, however, the lack of consideration in the current research of meaningful comparators precludes even the recommendation of functional therapeutic footwear over that of standard footwear. The text has been modified in the discussion to highlight these
8. Line 414 is a long bow and could be toned down - yes asymptomatic pes planus may not require intervention (yet is often intervened upon) but the full story is not available to us. I would keep mention of the anomaly, but perhaps in a less accusatory way.

- Text modified to reflect this comment with supporting annotations.

9. Limitations need expanding. The requirement of an English abstract needs discussion, the age and data collection method of participants increases change for error and a lack of description of shoe types and other forms of intervention also require acknowledging. Thank you for the opportunity to review this manuscript.

- Limitations now expanded to include these highlighted points.

Reviewer 2

In providing this peer review, I recognise the considerable amount of work that has been undertaken by its authors to form this systematic review. The SR has collated studies on a very useful area of practice that is relevant to the many health professionals involved in the prescription of this footwear. The review is very important is driving the quality of future studies in this field. I feel that this SR does warrant publication and the aim of my comments below are to improve the presentation and clarity of the information to the reader so that the extensive data presented can be better understood. With the complexity of the information provided by a SR it is never easy to present such information in an easy-read format. The first section of my comments are minor corrections that have identified typos / grammatical errors and a few points for clarification. The second section is suggesting a more detailed consideration of the results section and how the outcome criteria are included.

- The authors would like to thank the reviewer for their time and constructive feedback we have made modifications to the review to address your comments.

1. Background Line 82 - This doesn't read very well - I would suggest "The World Health Organisation International Classification of Function (…) recognises that mobility impairment affects the body structure and function of a child and it may lead……"

- Text modified to reflect this comment.

Line 89 - "Footwear has been..." change to "footwear is...."
Line 91 - no comma after including, Small p for pes planus
Line 92 - no comma after cerebral palsy and,
Line 93 - remove comma after professional,
Line 97 - remove comma after effects,
Line 100 - "highlighting that children's (add in "conventional"?) footwear research has grown rapidly"
Line 102 - "was represented in (add in "just"?) a small population…"
Line 105 onwards - why are certain words in bold? - this is not necessary.
Line 107 - add comma "further to this," Methodology
Line 146 - remove comma after randomised,
Line 147 - "case studies and case report studies (remove comma) were not....."
Line 151 - remove comma between children, and.....

- Text modified to reflect these comments.

Line 164-167 - need to state what adverse effects were being looked for specifically and state whether these were in the primary or secondary outcomes.

- Methodology has been expanded to qualify this.

Line 180 - remove comma from between developed, and ..... Results Line 229 remove comma "These three studies, focused on....." Also change semi-colon after alignment to a colon
Line 231Start a new sentence for "....all randomised controlled (level 2 evidence). Two were …"
- Text modified to reflect these comments.

Line 235 - "The sample size of two of the studies gave sufficient power to detect a moderate change". This needs further defining - what is meant by a moderate change?

- This was in reference to the statistical power calculation used in the quality assessment and was set at medium effect. This has now been clarified in the text.

Line 249 - "in the development of asymptomatic pediatric pes planus" - does this mean " in the correction of developmental asymptomatic pes planus"? or "in the development of asymptomatic pes planus"? It is unclear if this study would be eligible if it was looking at the development of the condition since the criteria for the review is footwear for correction rather than influencing normal development.

- The authors note this ambiguity. The studies included assessed children presenting to clinical settings with pes planus that was asymptomatic and did not define the subjects as typically developing. One study did look at the effects of corrective footwear on typically developing children, and this was excluded from the review during full text screening.

Line 250-256 - please be consistent whether splint is capitalised or not.
Line 262 - add "with an" between studies (with an) age range....
Line 263 - participants needs an apostrophe "participants' "
Line 264 - add "the" - "that (the) small sample size...."
Line 265 "amongst" ???
Line 281 - "medial" or "medical"?
Line 282 - be consistent in Downs or Down's syndrome
Line 284 "three of the four studies [.....]" needs a space before bracket and full stop after.

- Text modified to reflect these comments.

Line 295 - could you clarify in the text whether the "no difference were noted between the stabilising footwear and taping comparator..." is referring to the Down's study or to a different study?
Text modified and citation attached to the adjoining sentence to clarify this.

Line 299 remove the semi-colon between study and however

Text modified to reflect this comment.

Line 298-300 - there is a contradiction here "No SD was found in the step symmetry however the regular shoe and orthotic demonstrate a significant increase in step symmetry…"

Text modified to clarify this point.

Line 301 please define "meaningful effects"

Text modified to statistically significant.

Line 302 - please explain what "torqheels" are.

Additional text and appropriate annotation added.

Line 323 - "offered sagittal stability (add comma here) study design….."
Line 324 "[40]; both studies had a fair QI"
Line 331 - add full stop after [39]
Line 335 - AFO's change to AFOs as it is a plural.

Text modified to reflect these comments.

Line 339 - Is MBTs considered "therapeutic" footwear? Maybe consideration of this should be brought out in the discussion as it is not prescribed, it is bought in standard retail shops.

The authors note this point however the footwear was used from a functional therapeutic perspective in the study, and it could be argued that other forms of children’s therapeutic footwear such as stability footwear may be purchased by the public without prescription.

Line 341 - remove the semi-colon and use a full stop.
Line 342 - why is the "a" in bold?
Line 334 - need to remove the capital letter from "Motor"

Text modified to reflect these comments.

Line 350 - I come back to this point later about outcome criteria. There is a mention of the number of falls in this subgroup that has not been considered elsewhere. Was 'number of falls' a pre-set outcome criteria and if so, why is it not mentioned for any other study?

The biomechanical outcomes were not limited to specific outcomes due to the broad nature of the mobility impairments and therapeutic footwear considered. This is discussed further on to address the reviewer’s later points.
Line 354 - Again another inconsistency between subgroups - why is intention to threat analysis mentioned for the MBT study but not for any other study?

- This is reported for studies where there was an apparent loss to follow up or failure for all participants to be assessed across testing occasions the text of the results is now modified to include this comment alongside the Wenger et al. corrective footwear paper.

Line 358 - is adding a raise to standard footwear meeting the criteria of inclusion? If a raise was added internally in the form of a flat insole it would have the same effect but would not be classed as therapeutic footwear - or would it, in which case did you search for this treatment?

- Only if the structure of the footwear was modified such as a permanent sole adhesion would this qualify as therapeutic footwear, simple raise issued via removable inlay (insole) would not qualify as lift therapeutic footwear. This was established in the scoping review, studies that had used inlays as lifts were excluded from the scoping review. It was perceived that external lifts were applied where the level of limb inequality was greater than could be accommodated with an inlay.

Discussion Analysis and synthesis

There are no adverse effects reported in the results, is that because no study included them? Maybe reiterate this important point is that was the case and discuss relevance.

- This is discussed in the results in lines 225 -226 and the potential consequence of not recording adverse events is addressed within the discussion (lines 45-546).

The quality of the studies is reported in the discussion. It would be more useful here to summarise the quality points and then discuss the relevance. - it would benefit the review if the overall result was summarised here and discussed. Line 484 - as in the comment above for again quality of studies, the results of the pain outcome should summarised and then discussed here.

- Some repetition of the results in the discussion has been removed, with more focus on the relevance of study quality and how it impacts on bias and the ability to make clinical recommendations.

2. Outcome criteria One of my main concerns with this systematic review is the lack of very specific, pre-determined outcome criteria. The selection of important outcomes is a feature that sets a SR apart from a literature review and the prospective planning of the outcome criteria reduces any bias from the authors. Without detailed prospective planning in a review it is unclear if authors are consistent in extracting data or are choosing new outcomes as they start the data extraction and new ideas develop. Whilst I appreciate that the general outcome criteria stated in this SR were considered prospectively, they are not specific in detail and the presence / absence of all the selected criteria is not made clear for each study in this review. By not having strictly specified outcomes in this review, the authors have provided a mixture of outcomes in an overall summary of each paper and therefore there is no consistent representation of outcomes between the studies (this is particularly noted for the secondary outcomes, for example, number of falls, pain or adverse effects). In this situation, the piece of work has a systematic literature search but is not a systematic presentation of results. The primary outcomes are stated as "biomechanical and skeletal geometric measures". These ideally needed to be considered in smaller outcome groups. The authors needed to identify what are the most important biomechanical outcomes they would be looking for - for example, measures of pronation, and they should state what
measures of pronation they would include, for example, foot posture index, navicular drop, arch index. The same would apply for skeletal geometric measures - this needs to be reduced to outcome measures such as transmalleolar axis, metatarsus adductus angle, MLA angles, other recognised X-ray measures. Whilst I appreciate that these might be numerous, the authors needed to identify prospectively, which ones are useful to include, for example only valid measures. I note in the discussion section (Line 437) that five studies considered velocity as an outcome. If the authors had chosen velocity as a predetermined outcome in the "biomechanical" outcomes, then they might have been able to apply meta-analysis to this outcome or at least presented the data on a forest plot to identify a trend across the studies.

• The authors appreciate the reviewer’s comments and feel that predetermined specific biomechanical and skeletal geometry outcomes would be most applicable to present a clear summary of the results and trends across these results if a specific children’s therapeutic footwear type and or a specific children’s mobility impairment was to be considered for the review. However, from the scoping review, it was established that there was a broad range of footwear types used across a range of conditions. The authors felt there was a need to summate the evidence of children’s therapeutic footwear rather than isolating a specific mobility impairment or therapeutic footwear type. The scoping review identified that studies on therapeutic footwear had broadly considered biomechanical and skeletal geometry outcomes. Outcomes considering EMG or physiology (oxygen and metabolite consumption) had not been used. If the authors had focused on specific outcomes that were appropriate for one mobility impairment, this might have overlooked those of another (pes planus vs talipes equino varus, hypertonia vs hypotonia). The authors agree forest plots would better showcase trends in outcomes; however, due to the disparity of the conditions and the footwear interventions considered, this could also lead to misleading interpretation by the reader.

• As you point out “there is no consistent representation of outcomes between the studies”, however, this is not due to the lack of pre-determined outcome criteria for the review this is due to the wide range of outcome measures used across the different studies. While we agree that ideally “authors needed to identify what are the most important biomechanical outcomes they would be looking for”, and acknowledge the recent work in developing Standard Outcome Sets (i.e. ICHOM Standard Sets), we consider that this is not currently possible; with no agreement within this area on what the most important outcomes are.

The secondary outcomes are expanded a little more: quality of life, physical activity, social participation, self-esteem, pain. Line 440 in the discussion also states that compliance was a secondary outcome but it does not appear in the listing of the secondary outcomes in the method section, creating the possibility that it was chosen as an outcome at the point of data extraction and not pre-determined in the protocol. In the results section, the authors have given an overview of the outcome measures of interest when they have been included by each study, but they have not clearly stated when outcomes of interest were not included in the studies. In the therapeutic footwear section, it is clearly stated at the end of the first paragraph that no study considered adverse effects. In the corrective footwear subgroup, it would also be important to state that none of those studies considered any of the secondary outcomes. In that way the reader will know that those outcomes were not considered by the study as opposed to the authors not including the data for other (possibly biased) reasons.

• Compliance was not considered as a secondary outcome; the text has been modified to remove this confusion, with the issue of compliance in the reporting of the studies now discussed separately.

• The authors note the reviewer’s helpful suggestion in relation to corrective footwear and secondary outcomes and this has now been addressed in the text.
• Discussion of the split of the outcomes towards skeletal geometry in the corrective footwear grouping and biomechanical in the functional footwear groupings has been provided and made more explicit in the results section with potential reasoning for this suggested in the discussion.

I appreciate that it is not possible now to go back and be specific about the breakdown of outcome criteria considered, unless it was done at the time of protocol planning - but I am thinking it was not as the protocol is also general in the description of the chosen outcomes, but I think the results section could be tightened up little by making sure that all the outcomes of interest have a statement against them if the study did not include any of the pre-selected outcomes. And perhaps the lack of systematic presentation of outcomes needs a comment in the limitations section.

• All biomechanical and skeletal geometry outcome measures reported in the included studies are presented in the review; we did not limit to specific pre-selected outcomes within these areas. The range of conditions and the corresponding broad range of outcomes precluding a collective analysis is now discussed in the limitations section of the paper.