Author's response to reviews

Title: Systematic review of chronic ankle instability in children

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Author's response to reviews: see over
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Editors-in-Chief

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Dear Prof Hylton Menz and Mr Mike Potter,

Thank you for your review of our paper “Systematic review of chronic ankle instability in children”. We have been able to address the issues raised by the editorial team and reviewers and would like to take the opportunity to resubmit our revised manuscript. We have provided a revised version of our manuscript. In addition, we enclose the reviewer comments (verbatim) and our responses, point by point (with page numbers of any changes made).

Thank you for your consideration of our systematic review. Please let us know if you require any further information.

Yours sincerely,

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On behalf of the co-authors: Fereshteh Pourkazemi, PT, MSc, Amy Sman, BHealth (Physiotherapy), Joshua Burns, PhD, Claire E Hiller, PhD
1. Abstract (background): misspelled “focused”
   Authors’ response: This spelling error has been corrected.

2. Abstract (results): please be more specific on how 14,263 papers became 9
   Authors’ response: Some additional information has been added to the abstract to more clearly describe the screening process. This section now reads as:
   Following the removal of duplicates, 14,263 papers were screened for eligibility against inclusion and exclusion criteria. Nine full papers were included in the review.

3. Abstract (results): The last sentence seems out of place with no explanation.
   Authors’ response: This sentence has been eliminated from the results section and the topic is raised in the discussion section of the manuscript only.

4. Abstract (results): 100% reinjury is not a correct #
   Authors’ response: One study reported that all children (100%) who experienced an ankle injury were re-injuries. To be consistent with the reworked reporting of results, we have now chosen to report the only study that reported the prevalence of recurrent sprains. The sentence now reads:
   A history of recurrent ankle sprain was found in 22% of children.

5. Background (paragraph 1): not necessary to include the parentheses after mechanical instability unless you explain the other ones as well
   Authors’ response: Thank you for your suggestion. The explanation of mechanical instability has been excluded from the text. The sentence now reads as:
   Three main components of CAI exist; perceived instability, mechanical instability and recurrent sprain [2].

6. Background (paragraph 1): change to: people may experience one, two, or all…
   Authors’ response: This suggestion has been incorporated into the text.

7. Background (paragraph 2): Not just here, but throughout the paper- you cannot start a sentence with an acronym (CAI), must be spelled out if it is the start of a sentence.
   Authors’ response: The acronym helps reduce the word count. We would be happy to spell out the acronym if the Editor prefers.

8. Background (paragraph 2): The 2nd and 3rd sentences are confusing. 32% have symptoms of CAI, but 72% are impaired. Please clarify.
   Authors’ response: The second and third sentences of the Background highlight that 32% of people who experience an acute ankle sprain will develop CAI. Of these, 72% will have their function impaired. To clarify, this section now reads:
   CAI is common, with many adults enduring negative impacts long into the future. Following ankle sprain, up to 32% of people will develop CAI [6]. Of these, 72% will have their function impaired [6].

9. Background (paragraph 3): misspelled “focused”
Authors’ response: This spelling error has been corrected.

10. Background (paragraph 3): I fail to see the importance of addressing CMT as it is not mentioned at all in the rest of the manuscript. Further, CMT is a nervous disorder that is not caused by a previous injury as CAI is. Please delete or expand on its importance.

Authors’ response: We believe this information regarding children with CMT is relevant and enhances the background, highlighting the importance of this research and its broader applications. To clarify the importance of CAI in populations such as children with CMT, this section has been adjusted and now reads as:

Research to date has been heavily focused on adults and there appears to be little attention on the prevalence of CAI specific to the pediatric population. The limited body of research on CAI in children reports that it is commonly suffered by children following sporting injuries [9], hypermobility [10] and in those with inherited neuropathies such as Charcot Marie Tooth disease (CMT). CMT is a peripheral nerve disease which commonly inflicts symptoms of a cavus foot deformity, muscle atrophy, decreased sensation, and peripheral weakness [11,12]. This results in ankle unsteadiness, causing trips, falls and ankle sprain injuries [13].

11. Methods (inclusion criteria): you stated that there were no language restrictions and in your acknowledgements you thanked an individual for interpreting an article. Please include that information in your results.

Authors’ response: Information was provided in Paragraph One of the results section regarding the translation of a German paper. The section now reads as:

Initial searching resulted in 31,299 papers. Following the removal of duplicates, the titles and abstracts of 14,263 papers were screened for potential eligibility. After initial screening, 219 articles were identified as potentially eligible and full texts were sought. Succeeding full text review and the translation of a German paper [19], nine full papers were included in the review (Figure 2) [17,19-26].

12. Methods (inclusion criteria): under 18 yrs of age was included, however there were 2 papers that included participants over the age of 18. If the data could not be retrieved and separated out, these papers cannot be included in this review.

Authors’ response: In addition to our inclusion criteria of participants aged less than 18 years old, we have also stated that we included participants classified by the author as children or adolescents, which can be found on Page 4 of the manuscript: “Participants aged up to 18 years old were included along with studies including participants classified by the author(s) as children or adolescents.” We felt it was important to include results from papers where authors classified and treated participants as ‘children’ or ‘adolescents’ rather than setting an arbitrary age limit as the definition of childhood differs in many cultures and countries. To meet the purpose of this review, to systematically find and collate all available information on CAI in children and adolescents, it was important to include these papers due to their author’s definition of the participants.

13. Methods (exclusion criteria): more specifics are needed here. In Figure 2, 14,044 articles were excluded.
A more detailed explanation of why they were excluded is needed. Also, it should be clearly stated that studies involved participants over 18 yrs of age are excluded.

Authors’ response: Please refer to above response regarding the age-related comment.

14. Methods (exclusion criteria): terminology needs to be clarified. Here, the term “children” is used. However, later on in the manuscript, the terms “children” and “adolescents” are used. Please clarify on the terms used.

Authors’ response: The terminology within the manuscript has been revised to use only the term “children” to ensure consistency and improve clarity.

15. Methods (search strategy): You highlighted all of the databases searched, but then separated out the search strategy for only Medline. Please clarify.

Authors’ response: Figure 1 illustrates the search strategy utilised for Medline. It is explained in the manuscript (Page 5, Search Strategy) that this was modified for each database search: “Figure 1 illustrates the search strategy utilised for the Medline database, which was modified for each database”. This was an example of the search methods employed for each database.

16. Methods (assessment for trial inclusion): why is it “trial” inclusion? What trial is being included?

Authors’ response: This subheading has been changed to “Method for Study Inclusion” to enhance clarity.

17. Methods (assessment for trial inclusion): It is mentioned that 2 examiners screened titles, however there are 3 initials for the examiners. Is it 2 or 3 examiners? Please clarify. (this occurred in 2 places)

Authors’ response: Three authors shared the screening of the papers, as described in the manuscript: “MM and either AS or FP”.

18. Results (paragraph 1): Please clarify the 14,263 papers screened. Can you honestly say that you read 14,263 abstracts? In Figure 1, you should include the # of articles found using each set of search terms. That way the reader can truly see how many relevant articles were actually found and truly screened.

Authors’ response: Yes, we read the titles and abstracts of over fourteen thousand abstracts for eligibility. Figure 1 has been adjusted to ensure the clarity of our methodology. All search terms were combined with an ‘AND’ and/or ‘OR’ to produce a single list of titles/abstracts to be screened. It is therefore not possible to provide fractions of each list in Figure 1.

19. Results (perceived instability, paragraph 1): Please state what tools or questionnaires were used to identify perceived instability in children.

Authors’ response: Further information regarding the tools utilised to measure perceived ankle instability has been included in this paragraph:

Perceived instability was measured using tools including the Cumberland Ankle Instability Tool (CAIT) [14] and the Foot and Ankle Outcome Score (FAOS) [22], and through medical [20,24] and self reports [19].

20. Results (perceived instability, paragraph 1): One of the listed population groups investigated were ones that experienced severe ankle trauma. Please clarify the “severe ankle trauma from that subject group.
Authors’ response: The paper by Hollwarth et al. did not clearly define what was considered as a ‘severe’ ankle injury. This is reflected in the low assessment of the quality of the paper. The sentence has been altered to allow for readers to understand the use of undefined description:
Symptoms of ankle instability were investigated in specific populations including dancers [17], soccer players [22], children who were overweight [24], or who had experienced “severe ankle trauma” (undefined by authors) [19,20].

21. Results (perceived instability, paragraph 1): How were the initial injuries diagnosed?

Authors’ response: Initial ankle injuries were diagnosed by a medical practitioner or were recalled by participants via self-report. This information has now been included in Perceived instability, Paragraph 1:
Injuries were self-reported via recall or diagnosed by a medical practitioner.

22. Results (perceived instability, paragraph 2): This whole section read quite roughly. You interchange within the same paragraph discussing one paper to another. Maybe you should highlight each article to summarize its contents in a sub-category paragraph.

Authors’ response: The reporting of the results was remodelled to improve the flow of the manuscript and synthesise the main findings to create an overall picture of CAI in paediatric populations. We chose not to summarise each paper as this information is provided in the tables and we did not wish to duplicate results. The results for perceived instability now reads as:

Five papers investigated perceived instability including pain and impaired ankle function [17,19,20,22,24]. Symptoms of ankle instability were investigated in specific populations including dancers [17], soccer players [22], children who were overweight [24], or who had experienced “severe ankle trauma” (undefined by authors) [19,20]. Symptoms of perceived instability, pain, weakness, swelling or paraesthesia were investigated in dancers [17] and in children following ankle injuries [20,24]. Ankle injuries were self-reported via recall or diagnosed by a medical practitioner. Perceived instability was measured using tools including the Cumberland Ankle Instability Tool (CAIT) [14] and the Foot and Ankle Outcome Score (FAOS) [22], and through medical [20,24] and self reports [19].

Perceived instability and impaired ankle function during activity was common. Prevalence of perceived instability ranged from 31% in children with severe ankle injuries [19] to 71% of children who were dancers [14] (Table 2). The risk of perceived ankle instability was greatest for children who were overweight (≥ 85th percentile for Body Mass Index [BMI]) [24] of a younger age [20] and in those with abnormal talar tilt [19]. For every unit increase of BMI, the risk of having long term symptoms of instability increased 0.66% (OR, 1.07; 95% CI, 1.02-1.12; p=0.01) [24]. Of note, “permanent symptoms” of instability (lasting up to 12 years) were more frequent for injuries sustained by children under 10 years, compared to children aged over 10 years who were prone to more temporary symptoms (lasting 3 years, p<0.05) [20]. Subjective complaints of poor ankle
functioning were most notable in those who had a history of ankle injury [23] and in children following severe ankle trauma with abnormal talar tilt (>5°) [19].

23. Results (perceived instability, paragraph 2): it is mentioned that some symptoms last up to 12 years. First, “permanent” is not a correct usage of the word here. Second, please explain these symptoms and how they were identified. Was this a part of a longitudinal project? Please clarify the details of this paper.

Authors’ response: Although the term ‘permanent’ may not be the most correct use of this word, it was utilised in this paragraph as a direct quotation from the author. Quotation marks have been included to illustrate the direct quotation from authors of the paper. The sentence now reads as:

Of note, “permanent symptoms” of instability (lasting up to 12 years) were more frequent for injuries sustained by children under 10 years, compared to children aged over 10 years who were prone to more temporary symptoms (lasting 3 years, p<0.05) [20].

The study was not a longitudinal project. Full details regarding the symptoms explored in the paper were not included as they were beyond the scope of this review.

24. Results (perceived instability, paragraph 3): Sentences here are essentially the same as the paragraph above. See comment above to organize these paragraphs.

Authors’ response: Please refer to response to comment 22 above regarding the re-organisation of this section.

25. Results (perceived instability, paragraph 3): abnormal talar tilt should be included in the mechanical instability section, not perceived.

Authors’ response: Results of children with abnormal talar tilt is included in the perceived instability results because information regarding ‘subjective complaints’ of ankle instability are provided. This sentence has been reworded to clarify this explanation and now reads as:

The risk of perceived ankle instability was greatest for children who were overweight (≥ 85th percentile for Body Mass Index [BMI]) [24] of a younger age [20] and in those with abnormal talar tilt [19].

26. Results (mechanical instability, paragraph 1): Again, see comment from above. This paper would read much smoother if it was reorganized into sub-categories summarizing each paper included in the review.

Authors’ response: We have remodelled this section to align with the structure and flow of the results for the ‘Perceived Instability’ section. The section now reads as:

Mechanical instability following ankle sprain was investigated in two studies using four measures. Prevalence of mechanical instability was between 18% of children following severe ankle trauma [19] and 47% children who were dancers [17] (Table 1). A modified anterior drawer test identified increased laxity in adolescent dancers to be associated with lower CAIT scores, indicative of higher ankle instability (r=-0.484, p<0.01) [17]. Stress x-ray and talar tilt revealed a high prevalence of abnormal talar tilting (>5°) in 42% of children six years after severe ankle trauma [19].

27. Results (mechanical instability, paragraph 1): if 2 studies identified mechanical instability, the prevalence cannot “range” from 18-47%. Please reword.

Authors’ response: This sentence has been reworded to:
Prevalence of mechanical instability was between 18% of children following severe ankle trauma [19] and 47% children who were dancers [17] (Table 1).

28. Results (mechanical instability, paragraph 1): see previous comment regarding the terminology of children vs adolescents.

Authors’ response: The terminology included in this section has been revised to improve the clarity of the manuscript. The term “children” is now used consistently throughout the manuscript.

29. Results (recurrent sprain, paragraph 1): Again, see comment from above. This paper would read much smoother if it was reorganized into sub-categories summarizing each paper included in the review. It is especially confusing when new articles are people included.

Authors’ response: Please refer to the response to comment 26 above regarding the re-organisation of this section.

Six studies investigated recurrent sprain or re-injury rates using self or medical reports (Table 2) [17,21,23-26] using self [17, 24, 26] or medical reports [21, 23, 25]. A history of sprain ranged from 22% in football players [25] to 50% in dancers [17]. Only one study reported results of recurrent ankle sprain across a population, finding 22% of dancers had a history of recurrent sprain [17] (Table 1). The prevalence of recurrent sprain in children who had previously sprained their ankle ranged from 16% of normal weight children presenting to an emergency department for an ankle injury and 100% of ankle injuries sustained by physical education students [26]. Overweight children experienced a higher incidence of re-injuries to the ankle than those of normal weight (Table 1) [24].

30. Results (recurrent sprain, paragraph 1): reporting a 100% reinjury is incorrect. According to your Table 1, in Weir & Watson’s paper, 230 injuries occurred while, most of them were ankle injuries (though the actual number of ankle injuries was not reported in Table 1). And there were 7 ankle reinjuries. Unless there were only 7 ankle injuries out of the 230, the reinjury rate is not 100%. Please clarify or fix in all of the manuscript where you report 100% reinjury rate.

Authors’ response: Only seven injuries were ankle injuries of the 230 injuries experienced by participants. The most common single injury experienced was an ankle sprain. The information in Table 1 has been revised to clarify these percentages:

7 overuse injuries of the ankle were incurred. 100% of overuse injuries of the ankle were re-injuries.

31. Results (recurrent sprain, paragraph 2): It is stated that incidence of reinjury is self-reported, but then also say it was documented by a healthcare practitioner. Which is it? Please clarify.

Authors’ response: Either self-reports or medical reporting were used for each of the studies included. This sentence has been revised to improve clarity for readers and now reads:

Six studies investigated recurrent sprain or re-injury rates using self or medical reports (Table 2) [17,21,23-26] using self [17, 24, 26] or medical reports [21, 23, 25].

32. Discussion: much of the discussion is repeating the findings of the reviewed articles—which, if organized correctly, should be included in the results section. Thus, the discussion section should be left for the interpretation of what was found in the current body of literature and what should happen from here. Therefore, the whole discussion section needs to be revamped to discuss the findings, not summarize the
findings.

Authors’ response: In areas where the discussion has been linked to the results of the paper, the manuscript has been revised and remodelled to improve the organisation of the manuscript. Two paragraphs of the discussion were altered and now read as:

The prevalence of perceived instability was as high as 71% in children following ankle injury across the specific populations studied, with many participants reporting symptoms lasting up to 12 years [20]. Characteristics of perceived ankle instability via self-reporting of symptoms were noted in dancers [17], soccer players [22] and children who were overweight [24]. A systematic review of ankle sprains in adults reports a prevalence of perceived instability following acute ankle sprain ranging between 7% and 53% [27]. Perceived ankle instability in particular has been shown to have a large impact in adults, leading to changes in sporting and occupational activities [8]. The prevalence found in children is higher than the reported prevalence in adults. This may not be reflective of a true difference due to different testing methodologies employed. Perceived instability was measured with adult questionnaires including the CAIT and the FAOS, or the recording of subjective complaints. No pediatric-specific tool was available to measure this construct in children, which may account for the higher rate as items may have been misunderstood. Improving the measurement of perceived ankle instability in children would allow for any discrepancies due to questionnaire misinterpretation to be eliminated.

The prevalence of mechanical instability was reported to be as high as 47% in children who were dancers using the anterior drawer test [17] and in 42% of children using stress x-ray [19]. In previous reports, 25% of adults who had experienced lateral ankle sprain within six months prior to the study were found to be ‘moderately lax’ with an anterior drawer test [28]. The higher occurrence of mechanical instability in dancers may be due to the increased general joint laxity in this population [29].

33. Discussion (paragraph 2): please explain how prevalence of perceived instability is greater in children than adults (perfect for the discussion section of the paper)

Authors’ response: This paragraph has been reordered to ensure the clarity of this argument and may be seen above in response to comment 32.

34. Discussion (paragraph 2): you report a range of 7-53% of perceived instability in adults but only provide 1 citation. Please clarify.

Authors’ response: The citation provided is for a systematic review where a range for prevalence was reported. Information has been provided in the manuscript to clarify that this prevalence was found in a systematic review. The sentence now reads:

A systematic review of ankle sprains in adults reports a prevalence of perceived instability following acute ankle sprain ranging between 7% and 53% [27].

35. Discussion (paragraph 3): write out “AP” it hasn’t been defined.

Authors’ response: This term has now been defined.
36. Discussion (paragraph 3): please cite the statement that dancers are more lax

Authors’ response: A citation for the statement regarding laxity in dancers has now been provided.

37. Discussion (paragraph 4): Why was 100% previously reported as the reinjury rate, but here it is 56%?

Authors’ response: This paragraph has been adjusted to discuss the highest re-injury rate of 100% in the paediatric population. The section now reads:

Prevalence of recurrent sprain was high across most groups of children and adolescents studied. Medical and self-reports highlighted that in up to 100% of participants who experienced an ankle injury, it was a re-injury to the joint [26].

38. Discussion (paragraph 4): If there is a 56% reinjury rate in children, then why is there only a 34% reinjury rate as an adult. Shouldn’t this number only increase, not decrease? Please explain.

Authors’ response: We endeavoured to explain in the manuscript that this re-injury rate is high as it is taken from children in specific sporting populations who are more heavily exposed to experiencing an initial acute ankle sprain, placing them at an increased risk of developing CAI. This prevalence has been compared to adults in the general population who may not be active or involved in sports that increase the risk of ankle sprain. More research into re-injuries in the general paediatric population is required to make a true comparison to the general adult population.

39. Discussion (paragraph 5): again, from previous comment, define “children” and “adolescents”

Authors’ response: This terminology has been revised. Please refer to the response to the response to comment 28 above.

40. Discussion (paragraph 5): many stats from this paragraph was already stated in the introduction, therefore, unnecessary to repeat it again.

Authors’ response: Thank you for your suggestion; some of the repeated statistics have been omitted from the discussion.

41. Discussion (paragraph 6): It is unnecessary to “define” CAI again.

Authors’ response: The definition of CAI has been deleted from this paragraph.


Authors’ response: All of the additional data from Hiller et al.’s research pertaining to this review was reported in the manuscript. There was no other relevant information available to include.

43. Discussion (paragraph 6): Last sentence of this paragraph seems to include the discussion of FAI. Please expand.

Authors’ response: The final point of this discussion endeavored to link to a well-known argument that perceived ankle instability may not necessarily be related to mechanical instability.

44. Discussion (paragraph 7): The CAIT and FAOS are questionnaires for perceived functional instability. They do not measure mechanical instability. Please revise the first half of this paragraph.

Authors’ response: This paragraph has been reviewed and it could not be determined where this statement was made. The message being conveyed is that whilst adult tools may be appropriate for use with children to measure mechanical instability, tools such as the CAIT and FAOS are
inappropriate for use with children to measure perceived instability due to the personal, reflective nature of the construct.

45. Discussion (paragraph 7): Please cite the CAITY.

Authors’ response: The CAITY has now been cited.

46. Discussion (paragraph 8): limited number of existing papers that fit the inclusion and exclusion criteria of the systematic review is not a limitation; it is the nature of a systematic review

Authors’ response: As per the comments from both reviewers, this comment has been omitted from the manuscript.

47. Discussion (paragraph 9): I can agree that future research in CAI of the general pediatric population is needed, but why exclude the specific populations? That’s necessary as well. Please revise the paragraph.

Authors’ response: Thank you for your suggestion. It is important that more research in both general and specific populations is conducted. The conclusion has been revised to reflect this need and now reads:

Future research into perceived ankle instability in children is recommended to bridge this gap between clinical knowledge and evidence in the literature.

48. Conclusion: first sentence in unnecessary. The point of a systematic review is to systematically identify articles of interest and review the existing literature. The point of a SR is not to point out the lack of existing articles.

Authors’ response: The conclusion was altered to improve the flow of the manuscript and form stronger ties with the results and discussion sections. However, we felt that an important main finding of the review was that there is a very limited body of research available in the area of CAI in children. It is important to highlight this finding to drive future research in CAI in paediatric populations where it is truly needed. The conclusion now reads as:

The prevalence of CAI was high in specific groups of children and adolescents studied, comparable and often higher to that of adult populations. However, this systematic review found a limited volume of research about CAI in children. As no tool of high quality existed to measure perceptual components of CAI in children, the prevalence, distribution and impact of CAI on children was difficult to determine. Future research into CAI in children is recommended to bridge this gap between clinical knowledge and evidence in the literature.

49. Conclusion: the stated purpose of the SR was to examine the incidence of CAI in children, however, much of your conclusion talks about CAI in adults. Please revise the conclusion to fit your manuscript.

Authors’ response: The discussion of the systematic review highlighted the negative impact of CAI in adults. The conclusion reflected the focus on adults in the literature, highlighting the importance of the high prevalence of CAI in children. This may drive future research to explore the impact of CAI on children.

50. Table 1: a lot of information is lacking in this table. Some of the available information is confusing. Please organize the columns giving all of the same information. For example, under participants, start with type of participants, age (mean and SD), range of age. As it is organized now, some information is missing,
some numbers are included that don’t make sense to what it means. The table needs a little more organization to provide the necessary information.

Authors’ response: All relevant and available information from included papers was extracted and reported in the Table 1. Papers, such as those by Swenson et al. and Tyler et al., failed to provide detailed information regarding the participants included in their studies. Hence, the quality of their paper was assessed as low and information for all aspects of participant details is unavailable.

51. Table 1: Soderman et al and Hollwarth et al articles need to be excluded due to the age.

Authors’ response: Please refer to response to Comment 12 for the justification to include articles by Soderman et al. and Hollwarth et al.

52. Table 1: please explain the scored for the Steffen et al article

Authors’ response: Additional information has been included for Steffen et al.’s paper in Table 1.

53. Table 1: please provide actual number of ankle injuries for Weir & Watson article

Authors’ response: Information regarding injury numbers has been reworded to ensure the number of ankle injuries is clearly reported.

54. Figure 1: please report the number of articles found per search term, or at least how the search term was paired to find the relevant 9 articles.

Authors’ response: A clearer description of how the search terms were paired has now been included in Figure 1.

55. Figure 2: please specify how and why the 14,044 were excluded

Authors’ response: Figure 2 describes how and why papers were excluded. This is further explained within the Results section of the manuscript.

56. Figure 2: please specify the “other reasons, n=17” that those articles were excluded

Authors’ response: The ‘Other reasons’ number has been broken down further to justify the exclusion of papers.

Reviewer 2

1. Page 4, Inclusion criteria: The first sentence reads like a fragment, consider rewording to: “To be eligible for inclusion studies must have focused on…” The last sentence in this paragraph uses the term “author/s”, I believe the correct way to write this is “author(s)”, consider revising.

Authors’ response: These suggestions have been included in the manuscript. The section now reads: To be eligible for inclusion studies must have focused on CAI, commonly defined as experiencing perceived instability, mechanical instability or recurrent sprain [4,14], although papers reporting any long-term problems following ankle sprain were included.

2. Page 5, paragraph 2, sentence 1: This is insufficient information for another researcher to recreate your methods. Please give the references for the 3 manuscripts for which you solicited additional information from the author. Also, it’s not clear why these 3 were selected (e.g. did you request this information only from articles that appeared to report the variables of interest but did not separate children from adults in their
analysis?). Please tell the reader what about these articles triggered these requests.

Authors’ response: References and additional information regarding the reasons for seeking contact with authors has now been included in the manuscript in Page 5, Paragraph 2. The section now reads:

Three authors [15-17] were contacted to provide original data sets for analysis. Authors were contacted to obtain additional data for a variety of reasons including; combined scores for foot and ankle problems were reported [15], data for children and adults were reported together [16] or additional baseline data for ankle instability was not provided [17]. One author responded, providing original datasets for the study by Hiller et al. [17] regarding the laxity and sprain history of participants.

3. Page 5, paragraph 2: In numbered citation formats, the citation for “Hiller et al. (2008)” should read “Hiller et al. [16]” so that the reader is able to identify the correct reference from the reference list. This same citation error occurs a few times throughout the manuscript, including: Page 5 Downs and Black’s; Page 11 for Hiller et al. and Hollwarth

Authors’ response: Citations have been adjusted throughout the text.

Page 5, Paragraph 3: The writing of “Two independent examiners (MM and AS/FP) screened…” is unclear because you’ve stated 2 and then listed 3 examiners. If what is meant is “(MM and either AS or FP)” then it may be more clear to use this revised wording. Revise in the final paragraph on page 5 as well.

Authors’ response: This wording has been revised throughout the manuscript.

5. Page 5, final paragraph: The authors state that 2 independent examiners rated the studies, then jump to what happened when consensus could not be reached. Between these 2 final sentences, it should be added that “After independent review, discrepancies were settled by consensus” (or some other wording to the same effect).

Authors’ response: This sentence has been revised and now reads:

After independent review, discrepancies were settled by consensus. When consensus could not be reached, an additional examiner evaluated the quality to reach a final decision.

6. Page 6, Results: The reporting of the results of the search strategy should be improved. Reporting in Figure 2 is adequate, but the written paragraph feels incomplete. Specifically, it is written, “14,263 papers were screened”. It appears from figure 2 that this would be more appropriately stated, “the title and/or abstract of 14,263 papers were screened for potential eligibility.” Then the intermediate process (narrowing to 219) isn’t described at all. Consider something to the effect of, “Following initial screening, 219 articles were identified as potentially eligible and full text sought. Following full text review, nine articles were included in the review.”

Authors’ response: These suggestions have been incorporated into the manuscript and the section reads:

Initial searching resulted in 31,299 papers. Following the removal of duplicates, the titles and abstracts of 14,263 papers were screened for potential eligibility. After initial screening, 219 articles were identified as potentially eligible and full texts were sought. Succeeding full text review and the translation of a German paper [19], nine full papers were included in the review (Figure 2) [17,19-
26].

7. Page 6, Quality paragraph: Grammatically the final sentence “and loss of number of participants at follow up” reads awkwardly and should be revised. Is there a word missing? Consider, “and the number of participants lost to follow up”.

Authors’ response: This sentence has been revised to:

Other criterion commonly unfulfilled was the reporting of exact $p$ values [19,21,23,25,26] and the number of participants lost to follow up [22,25,26].

8. Page 7, paragraph 2, final sentence: the superscript for degrees is incorrect. Please use the degree symbol rather than the number 0.

Authors’ response: This superscript has been corrected.

9. Page 8, paragraph 2: The phrase “…with re-injuries of the ankle ranging from 16-100%” is not clear. Do you mean, of the injuries reported, 16-100% were re-injuries? Or, was the prevalence of re-injury following an acute sprain between 16-100%? Please add detail to clarify.

Authors’ response: This suggestion was incorporated into the manuscript to provide further clarification. Please refer to our response to comment 29 for the first reviewer.

10. Page 9, paragraph 1: The sentence, “The reported prevalence in adults is lower than results of children in this review” needs a citation. I’m assuming the citation is the same as is given in the next sentence. Consider rewording to combine the sentences, or add a citation here.

Authors’ response: This suggestion has been incorporated into the manuscript and the sentences are now combined. The section now reads:

The prevalence of perceived instability was as high as 71% of children following ankle injury across the populations studied, with many participants reporting symptoms lasting up to 12 years [20]... A systematic review of ankle sprains in adults reports a prevalence of perceived instability following acute ankle sprain ranging between 7% and 53% [27].

11. Page 9, paragraph 2, sentence 2: Has “AP” been previously defined? If not, please define.

Authors’ response: The acronym for anterior-posterior laxity has now been defined and included in the text.

12. Page 9, paragraph 2, sentence 3: This sentence is not well connected to the ideas above or below. At a minimum consider adding some joining words such as, “In previous reports, 25% of adults.....with an anterior drawer test [26], whereas symptoms of …”.

Authors’ response: This paragraph has been reviewed and adjusted in the manuscript and now reads as:

The prevalence of mechanical instability was reported to be as high as 47% in children who were dancers using the anterior drawer test [17] and in 42% of children using stress x-ray [19]. In previous reports, 25% of adults who had experienced lateral ankle sprain within six months prior to the study were found to be ‘moderately lax’ with an anterior drawer test [28]. The higher occurrence of mechanical instability in dancers may be due to the increased general joint laxity in this population [29].
13. Page 10, paragraph 2, sentence 1: There appears to be a typo here in the word “alike”. Do you mean unlike adults, or like adults? Sentence 2: please add an “and” prior to “11% are unstable…”

Authors’ response: These errors have been corrected in the manuscript.

14. Page 10, paragraph 2: Have you thought about the age of first sprain of the adults recorded in the literature? The age at first sprain is rarely recorded in studies of adults with CAI. But these adults with long term disability or increased re-sprain rate may have incurred their initial sprain as children, or as adults (it’s unknown). So the consequences we see in adults may be unique to adults, or they may be the exact same as these adolescents will experience if they were followed forward into adulthood. When comparing and contrasting the prevalence of long term symptoms or re-injury between these 2 groups, it would strengthen the discussion to include this possible connection and how that might affect our understanding of CAI.

Authors’ response: Thank you for your input. This is an interesting interpretation of the results and has been incorporated in the discussion of the manuscript within the Discussion (Page 9, Paragraph 3). A new section has been included to raise this interpretation:

Alternatively, the experiences of adults compared to children with CAI may differ depending on the age of the first ankle sprain encountered and the onset of CAI. The age of the first ankle sprain endured by adults is rarely reported in the literature. Hence, it is unknown if adults with long-term symptoms of perceived instability incurred their first sprain as a child or as an adult. The lower prevalence of perceived instability observed in adults might be unique to this older age group if their first ankle sprain leading to CAI was recent, during adulthood.

15. Page 11, paragraph 1, sentence 2: As written this sentence is conflicting, it appears to give a published reference for unpublished data. From your methods, it sounds like you were able to get additional data from this study. This could be made more clear here, consider rewording to the effect of, “An analysis of additional, unpublished data collected as part of a study by Hiller et al. [16]…” Also, sentence 3 states, “This finding highlights a relationship between perceived and mechanical aspects of ankle instability”…however the proceeding sentence talks about a relationship among 3 factors, not between these 2 only.

Authors’ response: These sentences have been edited within the manuscript to enhance clarity and now reads:

An analysis of additional, unpublished data collected as part of a study by Hiller et al. [17] showed that an increased number of ankle sprains and increased laxity were found to be significantly associated with decreased CAIT scores (indicating greater instability).

16. Reference list #17: Was this the article obtained in German? I believe it’s most appropriate to cite the title in German, with the translation in brackets. An example I found: Janzen, G., & Hawlik, M. (2005). Orientierung im Raum: Befunde zu Entscheidungspunkten [Orientation in space: Findings about decision points]. Zeitschrift für Psychologie, 213(4), 179–186.

Authors’ response: The German title “Spätfolgen nach Supinationstrau ma des kindlichen Sprunggelenkes” has been included in reference list.

17. Table 1: Overall, a very helpful table, well-structured and efficient in displaying information, however there are several small inconsistencies and formatting errors which need revision. Column header row: the
abbreviation CAI is not defined at the end of the table.

*Authors’ response: This abbreviation is now included in the key at the end of the table.*

18. Table 1, Row for Hiller et al.: (1) it might be helpful to the reader to include the reference # in the Author & Year column, for example ‘Hiller et al. 2008 [16]’. (2) There is an asterisk after 13 months, yet I don’t see this defined at the end of the table.

*Authors’ response: References for the papers listed have now been included in the table.*

19. Table 1, Row for Hollwarth et al.: The age range is 9-21, which exceeds the limit of 18 that appears to be set in your methods. It’s not clear in your inclusion criteria that a study which self-defines as of adolescents but includes youth up to 21 would still be allowable.

*Authors’ response: The first paragraph of the methods section describes that participants were included if they were below the age of 18 years old, or described/classified as ‘children’ or ‘adolescents’ by the author(s):

Participants aged up to 18 years old were included along with studies including participants classified by the author(s) as children or adolescents.

In the exclusion criteria, it states that papers with a “mixed sample” would be excluded. What was your boundary line?

*Authors’ response: Papers including a mixed sample of participants classified as a mixture of adults and children or those including the combined reporting of results for participants aged above and below 18 years of age were excluded.*

Also, ligaments avulsions are reported. Perhaps I’m not clear, but with a ligament avulsion I’m envisioning an avulsion fracture. Fractures were to be excluded based on your methodology. The two additional variables reported are also unclear, “pathologic clinical findings” and “abnormal” are very broad terms, can any additional detail be added to their description here.

*Authors’ response: We sought to investigate CAI following injuries to the ligaments of the ankle. Hence, papers involving participants who experienced traumatic fractures to the bones were excluded from the review. Papers including those with sprain or ligament injuries (who may have also suffered an avulsion from the inversion trauma) were excluded. The paper by Hollwarth was of a very low quality. No further information was provided for their definition of the terms ‘pathological clinical findings’ and ‘abnormal’. Quotation marks have been included in the table surrounding these terms to highlight the uncertainty with their definition.*

20. Table 1, Row for Marchi et al.: It’d be helpful for the reader if you could report whether the percentages given here were at the 3 year or 12 year follow-up point.

*Authors’ response: The percentages provided have now been more explicitly labeled as either the 3 or 12 year follow-up time points.*

21. Table 1, Row for Soderman et al.: If this is a prospective cohort, why is the length of time the cohort was tracked not reported? This information would be helpful, as 56% of sprainers had recurrent sprain within 1 year would have very different implications than if this were a lifetime recurrence rate.

*Authors’ response: The length of time for this study was one season long (length undefined by
This information has been incorporated into the table.

22. Table 1, Row for Steffen et al.: Abbreviations PI & NH are defined in the results column, rather than the end of the table. Additionally, the abbreviation “mean diff” is used but not defined at the table end. Since each line wraps around due to length anyways, consider eliminating the abbreviation entirely. Please check spacing as well and maintain consistency, for 95%CI is written both with and without a space between 95% and CI. Why is the last variable reported (ankle-related quality of life) formatted differently than all the others? Personally, I prefer this formatting to the formatting of the previous lines but either way revise to maintain consistency. Also, maintain consistency with abbreviations (here “mean difference” is spelled out).

Authors’ response: The consistency of the table formatting has been addressed as per the suggestions above.

23. Table 1, Row for Tyler et al.: In all the previous studies reported in this table, variables are reported as percentages or percentage plus n. Here only the n is reported. It’s aid comparison to convert to report n plus percent. Also, the sentence “9 sprained the same ankle (incidence 2.1)” is unclear. Does this mean, “9 of the 15 were recurrent ankle sprains”? What is being compared to come up with the incidence, first time sprains to recurrent sprains?

Authors’ response: Additional information has been entered to the table for clarification of these questions.

24. Table 1, Row for Weir & Watson: Again, why is the length the cohort was tracked not reported?

Authors’ response: This information has been added to the table.

25. Table 2: Author column: consider including the reference number, e.g. Hiller et al. [16], for additional clarity.

Authors’ response: Reference numbers have been included in Table 2 for additional clarity.

26. Table 2: Outcome column: Row for Hollwarth et al. in mechanical instability section: is there any way to further describe the outcome terms “total abnormal” or “pathologic clinical findings”?

Authors’ response: Further description to clarify the meaning of “total abnormal” was included.

Row for Hiller et al. in recurrent sprain section: typo writing greater than 2 sprains.

Authors’ response: This error has been corrected.

Row for Tyler et al. under recurrent sprain section: Again consider adding a percent or incidence rather than purely raw numbers.

Authors’ response: A percentage has been included in the results for Tyler et al.’s paper.

27. Table 2: Abbreviations: Add CAI to the abbreviation key, as well as “mean diff”.

Authors’ response: These abbreviations have now been included in the key for Table 2.

28. Figure 1: The formatting of this figure is not sufficiently clear. Please revise. Specifically, consider reformating to a table that more closely mimics a Medline search (at least as conducted in Medline/Pubmed). For example, interpreting from your current figure I believe the search strategy you used was as shown below:

#4 #1 AND #2 AND #3
#3 Children OR Child OR Paediatric OR…
#2 Instability OR “Ankle Instability” OR “Chronic instability” OR…
#1 “The ankle” OR ankle OR “ankle joint” OR …

The specific format example above is a personal preference, if the authors decide to leave the figure in the current format altered borders and more description would help increase clarity. For an example of this table format, see "Systematic Review of Eversion Force Sense Characteristics in Individuals with Functional Ankle Instability" by Wright & Arnold, Athletic Training and Sports Health Care, 2010.

Authors’ response: Figure 1 has been modified to more clearly describe the search strategy employed.

29. Figure 2: Very nice figure. Under “records screened”, please add further description by stating (here and/or in the methods) what they were screened for. If the authors mean “Title and/or abstracts screened for preliminary eligibility” that might be a more precise way to report what actions were taken at this step.

Authors’ response: This information has been incorporated into the figure.

30. Figure 2: Under reasons for exclusion of full-text articles, one reason is “abstract only”. Since it’s impossible to have a full-text article which only has an abstract, it’d be more clear to rephrase. If the box to the left stated “Full text articles retrieved and assessed for eligibility”, the first bullet in the box giving reasons for exclusion could be Abstract only, followed by description of the reasons for exclusion of articles with full text. Also, 17 is a fairly large number under the “other reasons” category—can any more description be given?

Authors’ response: This figure has now been adjusted according to above suggestions.

Discretionary Revisions: 31-35

31. Page 5, final paragraph: consider adding a comma after “…the representativeness and groupings of participants” to enhance readability. With multiple uses of the word “and” in this sentence it would help the reader.

Authors’ response: A comma was added in this sentence to aid readability.

32. Page 6, results paragraph final sentence: This statement about no pediatric-specific measures of CAI seems a little out of nowhere—the background or rationale for looking at this isn’t included in the introduction. The discussion section about pediatric-specific measures is very interesting and well connected. I’m not sure how this first mention of the topic could be better connected, but it might be worth considering.

Authors’ response: This sentence has been deleted, in the results section, as it does not clearly answer an aim of the review.

33. Page 8, paragraph 1: Consider adding a “0” before the decimal place so the correlation reads, “r= -0.484”. Especially with a negative sign it’s hard to read a partial number without the 0 to hold the space prior to the decimal point. This could also be done on page 7, paragraph one for the “p=.01”.

Authors’ response: This suggestion has been incorporated into the text.

34. Page 12, paragraph 1: Sentence 2 reads awkwardly, consider revising. Sentence 3 doesn’t appear to flow well with the previous sentence. Consider connecting to the rest of your limitations by rewording to “Additionally, numerous studies…” Or consider eliminating this limitation entirely—I’m not sure it’s a limitation that acute instability (an entirely different phenomenon) has been studied but had to be excluded
from this review because the scope was only chronic pathology.

Authors’ response: Sentence 2 has been adjusted to enhance readability. The third sentence has been eliminated in light of the reviewer’s suggestion. The section now reads:

A limitation of this review was that many papers that did investigate CAI were excluded due to the grouping of the results of children and adults together. Therefore, more information on CAI in children may be available than could be extracted for review due to the inclusion criteria of utilised for the present study.

35. Table 3: The definition for “1” could be revised to increase clarity. It appears that the best label for “1” that fits with all columns might be simply “criteria met”. The other descriptors which are currently used do not apply to all columns, and thus have a potential to confuse the reader.

Authors’ response: The definition included in the key for Table 3 has been adjusted.