Author’s response to reviews

Title: Prevalence and incidence of musculoskeletal extremity complaints in children and adolescents. A systematic review

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Author’s response to reviews:

Response letter

Dear Editor,

Thank you for the opportunity to submit a revised version of our manuscript; ID: BMSD-D-17-00271 about the prevalence and incidence of musculoskeletal extremity complaints in children and adolescents. We appreciate the constructive comments, and hope the reviewers’ concerns have now been addressed, and that you will find the changes to be adequate. Please, find below a response to each of the points raised by the peer reviewers.

Yours sincerely,

Signe Fuglkjær

Comments from Paul Campbell, PhD (Reviewer 1)

Background

There is a need to give greater justification for the focus on MEC, at present it seems an arbitrary selection, and perhaps more might be drawn from the relationship of MEC from physical activity, and the relevance to clinicians?

ANSWER: We made an attempt to give this justification by adding: ‘Recently, the Global Burden of Disease studies reported musculoskeletal pain as one of the leading causes of years
lived with disability [1] and this constitutes a substantial burden on society [2]. Therefore, it is important to design better prevention strategies and early effective treatment. To do that, more basic knowledge about the epidemiology of musculoskeletal complaints in children and adolescents must be obtained first.

The epidemiology of spinal pain in children is well-described [3-5], whereas less attention has been given to musculoskeletal extremity complaints (MEC) in children. Furthermore, musculoskeletal problems in childhood might not only lead to musculoskeletal complaints in adulthood, but could also be a barrier for physical activity and thus have a negative influence on general health [6].’ (p. 2, 1st paragraph).

There also needs to be more explanation for the distinction between traumatic and non-traumatic onset and how this might also be useful within a clinical context for example.

ANSWER: We have made an attempt to clarify this by adding the following paragraph ‘Various terms have been used in relation to MEC, starting from the less severe ache to injury or more severe musculoskeletal disorders. Commonly, MEC are divided into traumatic and non-traumatic complaints, where a traumatic complaint has been defined as an injury resulting from a specific identifiable event, whereas a non-traumatic complaint is not related to an identifiable event [10].’ (p. 2, 3rd paragraph).

However, we believe the real justification comes later in the same section: ‘While this type of research provides valid information about specific injuries, mainly injuries of a traumatic onset, they do not represent the full picture of MEC in the general population. Specifically, none of these methods collect valid information about non-traumatic complaints in the general population, which has been shown to represent a large part, close to two thirds, of MEC [6, 11].’ (p. 2, 3rd paragraph)

We hope, this is now more clear when the distinction between the two types of complaint is described first.

Further, it is described in the first paragraph of the method section, including clinical examples. (p. 3, 2nd paragraph)

Methods

Traditionally systematic reviews would normally search in more than 2 databases, can the authors justify this potential restriction.
ANSWER: We agree, that it would have been appropriate to repeat the literature search in other databases. However, it is our experience that the large majority of epidemiological articles can be found in the two databases that were used, PUBMED and EMBASE. Therefore, we do believe that the performed literature search can be justified, although we agree, that this is a weakness and has mentioned this in the discussion: ‘Finally, it would have been appropriate to repeat the literature search in other databases. However, it is our experience that epidemiological articles can be found in the two databases that were used, MEDLINE and EMBASE, and therefore we do believe that the performed literature search can be justified, although we realize that potential relevant articles can have be missed.’ (p. 14 4th paragraph)

Also I note that only one person conducted the title and abstract screening process, usually two or more would be involved?

ANSWER: Thank you for the comment. This part was not correctly explained in the method section, and has now been corrected: ’The first and third author (SF and LH) reviewed the titles and abstracts and identified relevant articles to be read in full text.’ (p. 4, 4th paragraph)

I wondered about the assessment of quality used in this review. The authors did not apply any weight to "study attrition", however they also report on “incidence” therefore study attrition would be an important component of this? The authors also stated that studies of a longitudinal design were considered as a series of cross sectional studies, can they offer support for this strategy?, an alternative for me would be to mark down cross sectional studies marked as "no" in an attrition category?

ANSWER: Good point and an illustration of the importance of peer review! We have now added the ‘Study Attrition’ domain from the QUIPS tool to the quality assessment, and performed the assessment on the incidence studies (Table 1). As you suggested the cross sectional studies have been marked as N/A.

In the evaluation of total risk of bias of the incidence studies, we have ranked the ‘Study Participation’ and ‘Study Attrition’ equally. Thus, both domains need to be considered ‘yes’ to be classified as low risk of bias. For further details, please see the method section (p. 5)

More information is needed on how trauma, and non-trauma were defined within this section, maybe some examples from the papers you included would be helpful here.

ANSWER: We made an attempt to clarify this in the method section by adding ‘When possible, the MEC were divided according to causation, and were categorised as either traumatic or non-
traumatic. A traumatic complaint was defined as an injury resulting from a specific identifiable event, whereas a non-traumatic episode was not related to an identifiable event [10]. In example, a traumatic complaint could be pain due to fall from a horse, and a non-traumatic complaint could be pain of unspecific origin developed over a longer period of time.’ (p. 3, 2nd paragraph).

Further, due to a suggestion of the other reviewer, this paragraph was moved to the beginning of the method section.

Discussion

I liked the discussion on the potential reasons for the differences reported (e.g. by age) and the authors highlight the need for more longitudinal studies to unpick these relationships further, however I would have thought of maturation and weight gain are important too and that maybe sports don't necessarily cause, but may also reveal issues in children?

ANSWER: We have tried to limit causative speculations, but we have changed that paragraph to read: ‘The dominance of ankle/foot and knee complaints was similar to what was found in a review of sports-related injuries in children and adolescents [11] indicating either that many of these complaints often are related to sport, or that sports participation actually reveals otherwise unnoticed injuries. A possible explanation for the changing complaint pattern with age could be the development of the musculoskeletal system due to pubertal growth including a general rapid physical growth [57]. One consequence of this could be that the calcaneal growth plate is stressed by the Achilles tendon (Sever’s disease) in younger children whereas, when the child matures, the growth of the immature skeleton more commonly leads to apophysitis around the anterior knee located to the tibial tubercle or the inferior patellar pool, leading to more knee complaints in older child.’ (p. 12, 1st paragraph in the discussion)

Minor point - on page 13, 3rd paragraph it states "...use of questionnaires is more than usually challenged in this age group..." , it might be better to say "there are challenges with the use of questionnaires within this age group"

ANSWER: Thank you for pointing this out. This have now been changed (p. 14 line last paragraph)

Comments from Helen French (Reviewer 2)

Title: The title does not really explain what the review is about - suggest revision

Description of musculoskeletal complaints in children and adolescents- a systematic review
ANSWER: Thank you for the comment. We recognize, that the word description implicates more than prevalence and incidence. We have changed the title to:

‘Prevalence and incidence of musculoskeletal extremity complaints in children and adolescents. A systematic review’

Abstract: In the background section, you refer to the 'occurrence'. I am unclear what this means. Do you mean prevalence?

ANSWER: Actually, we mean both prevalence and incidence and therefore tried to find a word, that could cover both. To be clear, we have now changed the abstract: 'The objectives of this systematic review were therefore to describe the prevalence and incidence of musculoskeletal extremity complaints…' (abstract, background section)

And in the background section as well (p. 2, 1st paragraph)

In the results section of the, can you give the age profile of the youngest age group and oldest age group.

ANSWER: Yes, good point. We have added this information to the abstract (result section): ‘For most of the analyses, a division between younger children aged 0-12 years, and older children aged 10-19 years was used.’ (abstract, result section).

Background;

I would suggest you outline here what you mean by 'musculoskeletal complaints' in this section

ANSWER: We made an attempt to explain it by adding the following paragraph ‘Various terms have been used in relation to musculoskeletal discomfort, starting from the less severe ache to injury or more severe musculoskeletal disorders. Commonly, musculoskeletal extremity injuries are divided into traumatic and non-traumatic complaints, where a traumatic complaint has been defined as an injury resulting from a specific identifiable event, whereas a non-traumatic complaint is not related to an identifiable event [10].’ (p. 2, 3rd paragraph).

Further, it is described in the first paragraph of the method section, including clinical examples. (p. 3, 3rd paragraph)

Pg 2, line 31 change 'risk factor of' to 'risk factor for'
ANSWER: Thank you for pointing this out. Due to the changes in the background section this paragraph have now been erased.

Pg 2 line 35 am unclear what you mean by 'minor distortion'. Suggest rewording for clarity.

ANSWER: We made an attempt to clarify this point by changing it into ‘unspecific minor complaints’ (p.2, at the bottom).

Methods

Was the review protocol registered in the PROSPERO database?

ANSWER: Yes it was. And it is now registered as: completed, but not published.

Was the review conducted according to PRISMA guidelines? Suggest submitting a completed copy of the PRISMA checklist

ANSWER: Yes, it was. And the prisma checklist is now completed, and submitted as an additional file (Additional file 3). Further it is now mentioned in the method section: ‘The review was conducted according to the PRISMA guidelines, and the PRISMA checklist can be seen in Additional file 3.’ (p. 6, last part of the method section)

Pg 3, line 54. Only 2 databases were searched, which is a limitation of this review. Was any grey literature searched?

ANSWER: We agree, that it would have been appropriate to repeat the literature search in other databases. However, it is our experience that the large majority of epidemiological articles can be found in the two databases that were used, PUBMED and EMBASE. Therefore, we do believe that the performed literature search can be justified, although we agree, that this is a weakness and has mentioned this in the discussion: ‘Finally, it would have been appropriate to repeat the literature search in other databases. However, it is our experience that epidemiological articles can be found in the two databases that were used, MEDLINE and EMBASE, and therefore we do believe that the performed literature search can be justified, although we realize that potential relevant articles can have be missed.’ (p. 14, 4th paragraph). We did not consider gray literature.

Pg 3, line 70. You refer to excluding athletes from this review- how did you define 'athletes'
The aim of this systematic review was to describe the prevalence and incidence of all musculoskeletal extremity complaints in children and adolescents in both general and clinical populations, and not complaints limited to specific types of sport. We decided to exclude sports-setting studies, in example studies investigating the frequency of injuries among football players. This was decided because the pattern of injuries in these settings is not representative of the pattern in the general population.

To answer your question: Our definition of an ‘athlete’: children or adolescents participating in sport.

Obviously, our data contain complaints that the children had during sport, and this was not well formulated. Therefore, we have amended the following:

‘Exclusion criteria

Special settings or groups, e.g. children with other diseases such as diabetes or other chronic diseases, or children from a specific sport setting (e.g. football players) were excluded, because their pattern of injuries might not be comparable to rest of the population.’ (p. 4, 3rd paragraph)

Pg 3, line 79. I suggest you move the section on case terminology further up into the methods section- before the search strategy so that it is at the beginning of the methods section.

ANSWER: We agree with you. This have been changed (p. 3, 2nd paragraph)

Pg 4, line 86. You refer to studies of prevalence or incidence here- is this a study of prevalence/incidence but it is not clear until now that is what this review is studying. There are assessment checklists for prevalence studies.


This contains extra items in addition to the ones you included to assess study quality including: response rate. Valid methods for measuring the condition.

ANSWER: Thank you for your comment. The Joanna Briggs checklist looks very useful. As we wrote in the manuscript, we were not aware of quality assessment tools designed for studies of prevalence and/or incidence, and we therefore decided to use a modified version of the QUIPS tool. Fortunately, we believe, that the nine questions in the Joanna Briggs checklist are fully covered in our quality assessment, including response rate and assessment of valid methods. We have tried to clarify by adding the form we used as additional file 2. We believe, that:
The ‘Study Participation’ Domain covers question 1-5 + 9 in the checklist by Briggs.

The ‘Outcome Measurement’ Domain covers question 6-7 in the checklist by Briggs.

The ‘Statistically Analysis and Reporting’ Domain covers question 8 in the checklist by Briggs.

The response rate was assessed in the ‘Study participation’ domain (question 1a in additional file 2), and the outcome measurements were in the ‘Outcome measurements’ domain (question 3b in additional file 2).

In addition, due to a suggesting from the other reviewer, the ‘Attrition Domain’ was added to quality assessment. The presence of attrition bias is important in incidence studies, therefore the ‘Study Attrition’ domain was included in for assessment of quality of the studies reporting incidence. Please, see table 1 and our response to the other reviewer.

Pg 4, line 104: In relation to the response options, there appears to be no 'unclear' option.

ANSWER: There was an unclear option. The checklist have now been submitted as an additional file, please see additional file 2.

Pg 4, line 105: 'full-filled' should read 'fulfilled'

ANSWER: Thank you for pointing this out. This have now been changed (p. 5, 2nd paragraph).

Pg 5, lines 109-115. Whilst the authors have attempted to assess methodological quality and devise a scoring system, this is not validated and should be noted in the discussion section as a limitation of the review.

ANSWER: We have added the following to the discussion: ‘Although the QUIPS tool has been validated [25] the modified version tool used in this review has not, which might be considered as a limitation of this review.’ (s. 14, 4th paragraph).

Pg 5, lines 118 and 133 'Data was' should read as 'data were'

ANSWER: Thank you for pointing this out. This have now been changed (p. 6, 1st and 3rd paragraph).
Pg 6, line135. I wonder about the rationale of considering the longitudinal studies as two cross-sectional studies. Why do some of the prospective studies report only one set of data (e.g. Mikkelson, Shier, Verhagen).

ANSWER: We have tried to explain this part further in the description of the included articles:

‘Of the 19 general population studies, seven were prospective. Four with one follow up evaluation [37, 43, 51, 52], and three studies reporting incidence over time.’ (p. 9, 1st paragraph)

Thus, Verhagen et al. - injuries were collected over a 1-year period, therefore reported one set of incidence data. Same thing for the two articles of Jespersen et al.

Mikkelsson et al. we only reported baseline data, since follow-up data was only reported in relation to persistent pain. The study design of this study has been changed to a cross-sectional study.

Results

Table 1: please add the citation number for each study in this table.

ANSWER: We have added the references to table 1.

Table 2 which provides the study characteristics is an important table due to the heterogeneity of the included studies and should be included in the main paper, rather than as a supplementary file.

ANSWER: We agree and have now included the table in the main paper (Table 2).

Pg 8, line 171. This section could provide more detail e.g. countries where studies took place, populations studies, methods of determining the prevalence of MSK complaints.

ANSWER: We agree, and have added some changes to the section:

‘The search resulted in 19 general population studies and three clinical population studies (Table 2). All the included studies covered children and adolescents of both sexes. Most of the studies were conducted in the northern part of Europe, but also other parts of the world were represented with three studies from North America [35, 43, 56], one from Australia [46] and one from India [50] were represented. Of the 19 general population studies, seven were prospective. Four with
one follow up evaluation [37, 43, 51, 52], and three studies reporting incidence over time [14, 39, 44]. In most of the studies, data was collected via questionnaires, either self-reported or filled in by parents, but telephone interviews, mobile phone text messages and diagnoses from general practice were also used. The 22 studies used 14 different outcome measurements as different as point prevalence, twelve month prevalence and incidence per 1000 exposure hours.’ (p. 8, last paragraph)

Pg 8, line 188. Can you clarify the age groups of the 'younger children' and 'older children' in this line. Are you including the adolescents here?

ANSWER: We have tried to clarify this, by adding the age in brackets (p. 9, 2nd paragraph). Furthermore, the description of the included children has now changed because of the corrections above, therefore the age groups are described as the last thing in the paragraph above.

Why are tables 2a and 2b labelled as such? Why not call them tables 3 and 4 and label the others accordingly.

ANSWER: They were both labelled as Table 2 (now table 3, due to the change of supplementary file 2 to Table 2) to indicate that they report the same outcomes, only in different age groups, and therefore essentially constitute one table, just divided into two age groups. This is equivalent to Table 3 (now Table 4), which has fewer data, and therefore can be included in one table.

Overall, I find the results tables confusing to navigate. What are the units of measure for incidence and prevalence.

Without knowing the actual complaints in each region, it is hard to see the value of these results.

ANSWER: We agree, that the results are not straight forward. However, this is primarily due to the heterogeneity of the outcomes, and we find it difficult to simplify/standardize further. We have tried to further the understanding by adding ‘%’ in the headlines where relevant, and explaining the brackets in the table legends. Furthermore, we have changed the footnotes to numbers instead of asterisks, not to be confused with indicators of significance.

Discussion

The authors attempt to explore reasons for presence of MSK complaints in different body regions, which is quite difficult without knowing the actual pathologies involved. There are
potentially lots of factors which would contribute to pain in these populations. For example, on pg 11, line 260, the authors report posture as potential causes of neck/shoulder pain which of course is possible. But what about heavy schoolbags?

ANSWER: We agree that there is a plethora of factors attributing to the reported pain, of which school bags might be one. However, the discussion about neck vs. shoulder pain does not relate to etiology but rather a potential source of misclassification.

In relation to the clinical population studies, the sources of this data are important to note as they are all from general practice, where one would assume less severe complaints present. As the authors point out, more severe injuries would present in an Emergency department setting. The authors should make this more explicit.

ANSWER: We wanted to describe the prevalence and incidence of all musculoskeletal extremity complaints. We recognize that the proportion of traumatic complaints most likely will be larger in an emergency department setting, compared to a general practice setting. But the traumatic/non-traumatic ratios were conducted from the population-based studies, which we believe contain both severe and less severe MEC, and describe the pattern of MEC within the general population.

Pg 12, lines 285-290 would sit better if it were placed straight after the discussion on upper limb results.

ANSWER: This could have been placed differently, and we have considered changing the order of the paragraphs, but we have decided to leave it as it is.

Pg 13, line 292 this should have been reported in the results section (in relation, to location of studies)

Pg 13, line 300: again this information should have been presented more explicitly in the results section and also in the methods section (that you would include studies which measured different types of prevalence)

ANSWER: As mentioned earlier, the description of the included studies were insufficient and we have amended the description of the included articles ‘Most of the studies were conducted in the northern part of Europe, but also other parts of the world were represented with three studies from North America [35, 43, 56], one from Australia [46] and one from India [50] were represented. Of the 19 general population studies, seven were prospective. Four with one follow up evaluation [37, 43, 51, 52], and three studies reporting incidence over time [14, 39, 44].
most of the studies, data was collected via questionnaires, either self-reported or filled in by parents, but telephone interviews, mobile phone text messages and diagnoses from general practice were also used. The 22 studies used 14 different outcome measurements as different as point prevalence, twelve month prevalence and incidence per 1000 exposure hours.’ (p. 8, last paragraph)

Further, in the method section we have added:

‘All levels of prevalence and incidence rates were included, and could be both parental reported or self-reported values’ (p. 4, 2nd paragraph)

Pg 13, line 305- in relation to the questionnaire used in the original studies, were they validated questionnaires or originally designed by the original authors. Again, this is important information not provided before now.

ANSWER: This was not always mentioned in the original articles, and thus the information was not available.

The authors do not outline the limitations of their review which I have outlined above. The lack of clarity around exclusion of ‘athletes’ is also a major limitation as it is unclear what their definition of athlete is.

ANSWER: Hopefully, this has been clarified in the explanation earlier.

Conclusion:

Pg 14, line 335. I don't think the fact that a meta-analysis was not possible should not be mentioned first in the conclusion.

ANSWER: This a good point. We have restructured the conclusion (p. 16)

The conclusion about traumatic vs non-traumatic may be misrepresented as traumatic complaints may present in emergency departments rather than general practice.

ANSWER: We wanted to describe the prevalence and incidence of all musculoskeletal extremity complaints. We recognize that the proportion of traumatic complaints most likely will be larger in an emergency department setting, compared to a general practice setting. But the traumatic/non-traumatic ratios were conducted from the population-based studies, which we
believe contain both severe and less severe MEC, and is a representative pattern of musculoskeletal extremity complaints in the general population.