Author's response to reviews

Title: Inter-tester reproducibility and inter-method agreement of two variations of the Beighton test for determining Generalised Joint Hypermobility in primary school children

Authors:

Tina Junge (tjunge@health.sdu.dk)
Eva Jespersen (ejespersen@health.sdu.dk)
Niels Wedderkopp (nwedderkopp@health.sdu.dk)
Birgit Juul-Kristensen (bjuul-kristensen@health.sdu.dk)

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Author's response to reviews: see over
Dear Editor

We are pleased to have the opportunity to respond to the useful comments raised by Marietta van der Linden and Dariusz Czaprowski regarding the article ‘Inter-tester reproducibility and inter-method agreement of Beighton tests and criteria for Generalised Joint Hypermobility in primary school children. An evaluation of 2 test batteries for performing the Beighton tests’ by Tina Junge, Eva Jespersen, Niels Wedderkopp and Birgit Juul-Kristensen.

The comments and questions will clearly help to improve the presentation and the precision of the key points in the article.

We hereby address the comments giving a point-by-point response to the concerns as well as a revised manuscript.

For the question regarding the written consent, the following has been added to the manuscript:

‘The grades are representing the youngest and oldest children in the CHAMPS Denmark part 1- The Childhood Health, Activity and Motor Performance School Study Denmark, a longitudinal cohort study of 1300 children in the Municipality of Svendborg [14]. The Committee on Biomedical Research Ethics for Southern Denmark approved the experimental protocol (jnr. S-20080047 HJD/csf). For this sub study of the CHAMPS Denmark – part 1, The Regional Scientific Ethical Committee for Southern Denmark considered the experimental protocol as non-invasive. Therefore, the study was exempt from the obligation of ethical approval from the ethical committee. Parents of each participating child received written information according to the Declaration of Helsinki [15] and before examination each child gave oral consent to participate in the study. Parents were asked to react if they did not want their child to participate.’

Referee 1, Marietta van der Linden

Minor Essential Revision

1. Page 7, 3rd paragraph 'For the Beighton .... (Method B). In this table the authors refer to Table 3, which I think should be Table 2

Answer: We hope that the revised version you have, says: ‘In Phase 3, kappa values varied from 0.49-0.94 (Method A) and from 0.30-0.84 (Method B) for the nine single tests in the batteries (Table 2)’, as this was corrected from the former version.

Referee 3, Dariusz Czaprowski

Major Compulsory Revisions

1. Please add number of females and males included in the particular stages of the inter-tester reproducibility and inter-method agreement sections. In my opinion it is important to strictly repetition of the study.

Answer: The methods section has now been adjusted to the following: ‘A total of 38 children were included in Method A and 32 children in Method B distributed by 57% boys and 43% girls.
with an average age of 7.4 years. .. In total, the test phase consisted of 39 children, who were tested with both Methods A and B, and by all four testers. In the test phase there were 54% boys and 46% girls with an average age of 9.6 years (Table 1)’.

2. Phase 3: In the third sentence there is an information that ‘...1 children with, and 20 children without...were sent to the allocated testers (figure 2).’ Please explain why in each of diagrams in Figure 2 there exist 20 participants.

Answer: The Figure has now been corrected to ‘19 participants’.

3. In my last review I draw attention to the exclusion criteria. Did authors take into account the children with hypermobility syndrome?

Answer: As previously answered, the population was a healthy children population all attending a normal school, meaning that we anticipate that no children had systemic diseases. Since there is no consensus on criteria for Hypermobility syndrome for children, only for adults (Grahame, 2000), we only included pain as an important factor that could possibly influence reproducibility of these tests. This was our reason for excluding those children. This means that, if a child has had arthralgia for longer than 3 months in 4 or more joints, they would be excluded from the study, if they had pain in the involved joints on the day of testing.

Minor Essential Revisions

1. Methods, 1. sentence: I suggest to shift the citation at the end of the sentence or to remove the part after citation. In my opinion, if this sentence part is not from the reference [4] it is only the authors’ opinion without evidence.

Answer: We agree with that, and the text has been changed to: ‘The two methods of BT were both in accordance with the original text of Beighton et al [4]. The original article from Beighton et al has a rather imprecise description of the tests, with no description of the procedures for each test. This is among others the reason, why there is so much diversion regarding the BT, and very few of these methods have been tested for reproducibility’.

2. I have still problem with the validity of the assessment of range of motion based only on the visual judgment, but it worth noting that the discussion section is much better and the authors sufficiently discuss this problem. However, the authors focus only on knee, fifth finger and elbow. Therefore, I suggest adding similar information regarding visual judgment of the shoulder positions during evaluation of elbow hyperextension. Moreover please correct the word ‘hyperekstension’ in the Appendix 2.

Answer: The following has now been added to the discussion: ‘...This difference was obvious by both in-experienced and non-experienced physiotherapists. The visual judgment of the shoulder position during evaluation of elbow hyperextension could also be a potential source of violation, as the angle of the elbow may seem dissimilar, if the shoulder is not placed in the starting position instructed’.

The word ‘hyperekstension’ has now been corrected to ‘hyper extension’ in Appendix 2.

3. Paragraph 5, sentence 2: I suggest shifting the number of the citation at the end of the
The citation has now been placed at the end of the sentence: ‘In a child study, goniometry was used to measure the passive bilateral hyperextension of the knees [21].’

The authors included the title with second sentence. I agree with this addition. However, in new version the title seems to be too long. Therefore, I suggest putting together these two sentences. E.g. Inter-tester reproducibility and inter-method agreement of two variants of performing of Beighton test for evaluation of Generalised Joint Hypermobility in primary school children.

Answer: Thank you for that suggestion, we will change the title to ‘Inter-tester reproducibility and inter-method agreement of two variations of the Beighton test for determining Generalised Joint Hypermobility in primary school children’.

Methods, sentence 2: I suggest dividing this sentence. The first part should be finish after word ‘respectively’. The second part should be start from ‘13 children...’.

Answer: Thank you for this suggestion. The sentence is now divided into two as follows: ‘The number of participants in the three phases was 10, 70 and 39 respectively. For the inter-method study a total of 103 children participated’.

Conclusions: please check the English expression of the second sentence.

Answer: The conclusion has now been adjusted to: ‘The inter-tester reproducibility of Methods A and B was moderate to substantial, when following a standardised study protocol. The described Beighton tests and criteria for classification of GJH are reproducible for children and therefore suitable for comparative studies of children, when using a GJH criterion of ≥5/9.

However, both methods need to be tested for their predictive validity at a higher cut-off level, e.g. ≥6 and ≥7’.

Please check the English expression (e.g. Conclusion in the Abstract and the first sentence in the 2nd Paragraph in the Conclusions sections in main manuscript).

Answer: The sentence has now been adjusted to: ‘Both test batteries can be used in the same children population, as there was no difference in prevalence of GJH at cut point 5, when applying method A and B.

Paragraph 4: sentence 2, the references should be at the end of the sentence [4,11].

Answer: We hope that the revised version you have also says: ‘Possible methodological shortcomings arise, as the BT do not include detailed descriptions of the tests or definition of the criteria for GJI; the BT as a modification of Carter and Wilkinson’s test were simply describing the populations assessed in the studies [4, 11]’.

Sentence 3: first part of this sentence should to have a reference

Answer: The reference has now been added to the sentence: ‘Consequently, in none of the basic illustrations or descriptions for the BT [4] is it stated precisely how the tests should be performed.’
10. Sentence 5: Please add the reference. If the authors don’t have reference for this sentence, please be less declarative, as in this case the sentence is only the authors opinion.
Answer: The sentence has now been adjusted: ‘Different starting positions and benchmarks can hypothetically effect the prevalence of GJH, thereby influencing the inter study comparison, and possibly making it difficult to test the predictive validity of BT in a cohort of children’.

11. Appendix 2: please divide the words ‘shoulderflexion’ and ‘shoulderabduction’. I have also problem with the word ‘volarflexed’. Please rephrase it.
Answer: The words have now been divided and rephrased.

Discretionary Revisions

1. I still suggest to include in the Discussion section information what is the difference between generalized joint hypermobility and hypermobility syndrome. The authors used in the Reference list the position, which are focus on the hypermobility syndrome [3 and 20]. Therefore, I suggest including a comment to distinguish GJH and HS.
Answer: Thank you for this comment. We agree that the two conditions are very different, and in most studies it is important to distinguish between these two conditions. The following paragraph has now been added to the discussion: ‘We do not know whether the current results would be similar in a group with Hyper Mobility Syndrome (HMS), as the present study is a reproducibility study, where the aim is to test the reproducibility of only the BT in a normal and relevant population for our upcoming studies. A requirement of such study is to keep the testing conditions and the subject conditions as stable as possible for the test rounds. It could be anticipated that test results of BT in subjects with HMS would differ from first to second round due to increased pain, but this needs to be studied in a future study. Such considerations were bases for having pain as exclusion criteria in the present study’.

2. Please to add the phrase ‘et al’ where the cited paper has more than two authors (e.g. Hansen et al. [7] and Smits-Engelsman et al. in the Discussion section).
Answer: Et al has now been added to the following sentences: ‘This was in accordance with the study of Hansen et al [7], with kappa values of 0.68 for the elbows and only 0.44 for the knees.’ and ‘In the study of Smits-Engelsman et al. [19] the knee test was also applied in.’

References: