Author’s response to reviews

Title: Motor and cognitive functioning in children treated for idiopathic clubfoot at the age of 3 years

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Author’s response to reviews:

Dear Professor Santosh,

Attached please find the revision of our manuscript and below our detailed replies to each of the concerns raised by the reviewers. We think that with the help of the reviewers' comments our manuscript really improved and now provides a more detailed picture about the motor and cognitive functioning in children treated for idiopathic clubfoot at the age of 3 years. As you will see, we responded to all of the very helpful suggestions.

We like to thank you and all reviewers for their so thoughtful comments that helped us to reflect our study in detail and to definitely improve the manuscript.

We hope that our revision satisfactorily addresses all of the comments and look forward to your decision.

Sincerely,

Dr. Julia Dillmann
Replies to Reviewer 2 (Susan Mahan):

This paper investigates the motor and cognitive skills of 3 years old with idiopathic clubfoot and compares to a cohort of similar aged normal children.

This is an interesting idea, but has some significant procedural flaws and inappropriate conclusions.

In the methods section, they note that they matched each of the 10 children with clubfoot with a randomly chosen age and gender match child from the "address list of the department of Developmental Psychology". It's not clear how these "normals" came to be on the address list of this department.

We would like to thank the reviewer and have included some further information about the control group in the method section:

Line 111-113: The typically developing children came to be on the address list of our department because they have already participated in an earlier and different study of our department and their parents have agreed to be contacted again. Originally, we got the addresses from the city administration.

Most concerning, it was not mentioned in the methods whether or not the experimenters who assessed the children (half with clubfoot, half "normals") were blinded to the clubfoot diagnosis; we find in the discussion that they were not. This is, to me, a major weakness of the project because of the subjective nature of the assessment.

We wrote it in the procedure section as well as in the discussion section that the experimenters were non-blinded with regard to the clubfoot diagnosis. As suggested, we now included some further information.

Line 131-136: Written informed consent was obtained from all parents in advance. The two experimenters were non-blinded with regard to the clubfoot diagnosis of the children. However, they were carefully trained and supervised by experienced researchers regarding the administration and interpretation of the Bayley-III-Scales. This should ensure that they were able to run and to analyze the Bayley-III-Scales objectively, regardless of the children’s diagnosis.
Line 221-226: Another limitation was that the two experimenters were not blinded with regard to the diagnosis of the children. However, the Bayley III-Scales are objective, reliable, valid and applied measurement tools for assessing early motor and cognitive development both in clinical practice and research settings [27]. Therefore, we consider it unlikely that the data were considerably influenced by subjective factors of the experimenters.

In the results the discussion of the cognitive assessment was not clear what in fact the difference were and what they were measuring.

Thank you for this suggestion. We now included some further information in the results as well as in the discussion section, for example:

Line 178-179: As can be seen from Figure 2, children treated for idiopathic clubfoot were less able to select object pairs correctly, a task quantifying visual-spatial-memory performance.

Line 197 – 202: Third, we found some slight cognitive impairments in children treated for idiopathic clubfoot. These children had difficulties selecting object pairs correctly, a task quantifying visual-spatial-memory performance. Particularly, the children were allowed to view 10 seconds a display of six cards (three object pairs: tops, flowers and cars). Then, the cards were turned over and the children had to identify the correct pairs of cards for the first two objects (tops and flowers) [27].

In the discussion, the authors initially claim that since the 40 month old children have decreased gross motor limitations, that this would lead to low self esteem. This is a tremendous leap. First, the references articles linking decreased motor skill with low self esteem were in a much older age group (youngest age 8). There is no evidence in this paper to suggest that these 40 month old children will continue to have decreased motor function. And there is no evidence that there is low self esteem in 40 month old children. So that is an incorrect leap of faith.

We like to thank the reviewer very much and agree that our conclusion was inappropriate. Therefore, we deleted this paragraph. Instead, we wrote a short paragraph at the end of the paper (future directions).

Line 233-236: Finally, previous studies revealed that motor competence seems to be important for school children’s self-esteem [34, 35]. Hence, future studies with school-aged children treated for idiopathic clubfoot should measure self-esteem additionally.
More discussion on how motor development and "visual-spatial-memory" cognitive performance is warranted, including what is meant by "visual-spatial-memory" cognitive performance. It is also not clear if this is a lasting deficit or if this difference resolves by school age years.

We agree that we do not know if it is lasting deficit or not, we have now discussed this aspect in more detail in the discussion section, for example:

Line 197-210: Third, we found some slight cognitive impairments in children treated for idiopathic clubfoot. These children had difficulties selecting object pairs correctly, a task quantifying visual-spatial-memory performance. Particularly, the children were allowed to view 10 seconds a display of six cards (three object pairs: tops, flowers and cars). Then, the cards were turned over and the children had to identify the correct pairs of cards for the first two objects (tops and flowers) [27]. Notably, the children treated for idiopathic clubfoot had difficulties selecting these object-pairs correctly. However, on all other cognitive items, the two groups achieved comparable results. Hence, it seems that gross motor development does not have a strong impact on cognitive development per se, but it seems to facilitate the development of specific cognitive skills, for example visual spatial memory performance. These results confirmed the findings of current studies in typically developing children [18–21], spina bifida children [22] and infants treated for idiopathic clubfoot [14]. Overall, it seems that gross motor abilities can influence specific spatial cognitive skills, emphasizing the need for a motor intervention program for children with strong motor disabilities.

Line 230- 232: Hence, future studies in older children treated for idiopathic clubfoot are needed to examine whether the observed gross motor and spatial cognitive deficits persist through childhood.

While this paper has some interesting ideas, the lack of blinding by the assessor is a major procedural flaw. And inappropriate conclusions in the discussion further weakens this study.

Thank you so much for all of the very helpful comments!