Author's response to reviews

Title: Correlates of functional ankle instability in children and adolescents with Charcot-Marie-Tooth disease

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Version: 2
Date: 15 October 2015

Author's response to reviews: see over
15th October 2015

Editors-In-Chief, *Journal of Foot and Ankle Research*

Dear Prof Hylton Menz and Mr Mike Potter,

Manuscript ID: 2600035998412681
Title: Correlates of functional ankle instability in children and adolescents with Charcot-Marie-Tooth disease

We thank the Reviewers for their comments regarding our manuscript and for considering our study for publication in *Journal of Foot and Ankle Research*. We have addressed the issues raised by the reviewers and now resubmit our revised manuscript.

We have resubmitted two copies of the manuscript (clean and track changes version). In addition, we have provided a separate outline below listing the comments made by the editorial team and reviewers verbatim and our replies, including the page numbers of any changes implemented.

We thank you once again for your patience and willingness to review and consider this manuscript.

Yours sincerely,

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On behalf of the co-authors: Joshua Burns PhD, Claire Hiller PhD, Melissa Mandarakas, Jacqueline Raymond PhD, Kathryn Refshauge PhD
REVIEWER 1

Major Compulsory Revision *or* clarification/justification

Reviewers’ comment: The authors report in the statistical methods that they applied my previous recommendation of analysing the gender data using Chi square rather than Pearson’s r. Throughout the manuscript however there are still repeated references to Pearson’s:

Abstract results – “female gender (r = -.469, P < .001) and impaired balance (r = .495, P < .001).”

Foot and ankle variables – “No significant gender differences (P <.05) were observed for age.”

Variables associated with ankle instability – “Factors significantly associated with right FAI included female gender (r = -.469, P < .001),”

“Factors associated with left FAI included female gender (r = .396, P < .05),”

Table 2. Col 3
These should be corrected throughout.

Authors’ reply: We have now used point biserial correlation to correlate with FAI with male/female gender. As the procedure for point biserial correlation in SPSS is the same as that for Pearson’s correlation these results remain unchanged.

Minor essential revisions

Reviewers’ comment: BACKGROUND – 2nd para. The assertion that the cavus foot is cardinal manifestation and may be another cause of FAI is overstated unless referenced to a definitive study. It may be associated but causality is being assumed here, rather than having been established.

Authors’ reply: The second paragraph has now been amended so as not to overstate pes cavus as a potential cause of FAI in CMT. A reference has been included which is supportive of foot deformities such as pes cavus, predisposing an individual to FAI.

Para 1, Pg 6: “High rates of cavovarus deformity have been reported in individuals undergoing surgery for chronic FAI (8). Pes cavus is a frequently reported manifestation of CMT. The excessively supinated foot structure associated with the cavus foot type might predispose the ankle to recurrent episodes of plantarflexion and inversion injury, particularly when it is coupled with foot and ankle strength imbalance”

METHODS

Reviewers’ comment: Functional measures. Could the authors provide some clarification on the definition of falls used? Young adults are relatively unlikely to fall completely to the ground even when significantly unbalanced, was some threshold used and/or guidance given.

Authors’ reply: Falls in this study were defined as the number of falls to the ground over the course of a day. This has been amended for greater clarity in the methods section of the manuscript. It has been our clinical experience that children and young adults with CMT lose their balance very regularly during the day. Because this happens so regularly we questioned the reliability of children reported “episodes of loss of balance” Falls are a less regular but more significant event. We felt the reporting of falls would have greater reliability in this population as they happen much regularly and are of greater significance to this population.

Para 1, Pg 10: “Falls are commonly reported in CMT and may be a result of instability at the ankle joint (3). To observe any relationship between falls and FAI, participants were asked to keep a diary the week preceding the study visit where they recorded the number of falls to the ground they experienced each day. The number of falls recorded in the diary were added and used for analysis.”

Reviewers’ comment: Discussion p13 typo “As this tool was unavailable AT for the present study,” remove redundant “at”

Authors’ reply: The typo has been corrected.

REVIEWER 2

Minor Essential Revisions

Reviewers’ comment: The authors reported that the CAIT has been validated in children (see discussion) yet in the conclusion suggest that development and validation of a paediatric version is required. This seems contradictory.

Authors’ reply: We have amended this section of the manuscript to reduce confusion. We have also included the reference of the journal article that discusses the construction and validation of the CAITY.

Para 2, Pg 13: “Although the assessor felt most children understood the questions and had good insight into the extent of their FAI they found some of the younger children required clarification and assistance with some items. As a response to this limitation, authors have since validated a paediatric version of the CAIT – the Cumberland Ankle Instability Tool – Youth, or the CAITY, to allow children to
answer the questionnaire alone, without influence from a third party (parent or clinician)"

Reviewers’ comment: The choice of statistical analysis measures linear correlation between two variables and quantifies the direction and strength of the relationship between variables. Causality, or the direction of causality, cannot be assumed as the analysis doesn’t take into account any other variables (measured or unmeasured). It is important that the authors are cognisant of the limitations of the analysis when interpreting the results and cautious about the translation of the findings to practice.

Reviewers’ comment: The choice of chi-squared test, as opposed to kendall tau, warrants brief justification.

Authors’ reply: Point biserial correlation has now been employed to correlate gender with FAI.

Reviewers’ comment: Review p values on pages 11-12. Some are reported incorrectly. Coefficients should be reported to two decimal places.

Authors’ reply: The p values have been reviewed and corrected. Coefficients have been reported to 2 decimal places.

Reviewers’ comment: I may be missing this but the r-values for the contralateral FAI do not appear in Table 2.

Authors’ reply: The r-values have been reported for both right and left feet in Table 2.


Authors’ reply: The spelling error in this reference has been amended.