

Spatiotemporal gait and fatigue do not change when using

common at-home gait tasks in patients with Facioscapulohumeral Muscular

Dystrophy: a pilot study

Nicholas G. Murray*, Marie Kelly, Vipul Lugade, Ryan Wuebbles, Madison Taylor, Douglas Powell, Takako Jones, Peter Jones

*Corresponding Author Nicholas Murray Neuromechanics Laboratory, School of Public Health, University Of Nevada, Reno, 1664 N. Virginia Street m/s 0274, Reno, NV 89557, United States of America.

Handling editor: Michal Heger Department of Pharmaceutics, Utrecht University, the Netherlands Department of Chemistry, Utrecht University, Utrecht, the Netherlands Department of Pathology, Erasmus Medical Center, the Netherlands Department of Pharmaceutics, Jiaxing University Medical College, Zhejiang, China

Review timeline:

Received: 20 December, 2022 Editorial decision: 13 January, 2023 Revision received: 23 January, 2023 Editorial decision: 23 January, 2023 Published online: 7 February, 2023

1st Editorial decision 13-Jan-2023

Ref.: Ms. No. JCTRes-D-22-00223 Spatiotemporal gait and fatigue do not change when using common at-home gait tasks in patients with Facioscapulohumeral Muscular Dystrophy - A pilot study Journal of Clinical and Translational Research

Dear author(s),

Reviewers have submitted their critical appraisal of your paper. The reviewers' comments are attached to this decision letter. Based on their comments and evaluation by the editorial board, your work was FOUND SUITABLE FOR PUBLICATION AFTER MINOR REVISION.

If you decide to revise the work, please itemize the reviewers' comments and provide a point-bypoint response to every comment. An exemplary rebuttal letter can be found on at http://www.jctres.com/en/author-guidelines/ under "Manuscript preparation." Also, please use the track changes function in the original document so that the reviewers can easily verify your responses.

Your revision is due by Feb 12, 2023.

Journal of Clinical and Translational Research Peer review process file 09.202302.004



To submit a revision, go to https://www.editorialmanager.com/jctres/ and log in as an Author. You will see a menu item call Submission Needing Revision. You will find your submission record there.

Yours sincerely,

Michal Heger Editor-in-Chief Journal of Clinical and Translational Research

Reviewers' comments:

Reviewer #1: I gave all my comments as a pdf attachment

There is additional documentation related to this decision letter. To access the file(s), please click the link below. You may also login to the system and click the 'View Attachments' link in the Action column.

Authors' response

Spatiotemporal gait and fatigue do not change when using common at-home

gait tasks in patients with Facioscapulohumeral Muscular Dystrophy -

A pilot study

The authors describe an interesting study about the use of a phone app to measure longitudinal gait decline in FSHD. In order to create this app they first study three commonly used gait tasks in order to identify which one is the most appropriate for a at-home assessment of gait. This is a very interesting and well-written study with clear novelty and impact. Besides some unclarities I do not see any major issues.

Dear reviewer – thank you for time and effort in reviewing our manuscript. My co-authors and I greatly appreciate the comments and the opportunity to amend the manuscript.

General Comments

- 1) This excerpt from the manuscript:
 - a. Modern smartphones with appropriate accelerometers are used by nearly 45% of the world's population21 and can be carried in numerous positions on the body to accurately track gait features.22,23 One particular smartphone application, Gait Analyzer24 created by Control One LLC, has been used with moderate success to analyze the gait of pathological and older adult populations.24–26 This application has high intrarater and interrater reliability,22 sufficient validity when compared to motion capture22,



is almost identical with the same excerpt from this paper:

b. A novel smartphone application is reliable for repeat administration and comparable to the Tekscan Strideway for spatiotemporal gait

Please modify to avoid self-plagiarism or cite the paper where you have got this excerpt from.

Thank you for this comment, this has been addressed and now reads:

Modern smartphones that are used by approximately 45% of the world's population²¹ and are fairly accurately, depending on the internal accelerometers, in tracking gait features.^{22,23} The Gait Analyzer²⁴ created by Control One LLC, has gained popularity as published research indicates it can accurately track gait of pathological and older adult populations.^{24–26} This application was recently validated against a gold standard gait mat²⁶, acceptable levels of intrarater and interrater reliability,²² and sufficient validity when compared to motion capture²².

Abstract

1) Please mention in the abstract briefly what the motivation was behind those 3-tasks (are those some sort of golden standard for gait analysis in FSHD?). I have the same comment for the introduction where the authors could elaborate further and give some references.

This had been further clarified within the abstract and now reads:

"These included the most common gait tasks reported in the FSHD literature 1) 12 consecutive walking trials over a ten-meter level surface, 2) 6 consecutive walking trials across a ten-meter level surface in the morning and afternoon (a minimum of 4 hours between testing) and 3) ambulating for as long as they can for 6-minutes."

This has been further clarified within the introduction and now reads:

"The purpose of this study was to analyze three commonly used gait tasks reported in the FSHD literature for use in an at-home setting for FSHD patients"

2) Any indication as to why patients found the 6-minute walking task difficult, however, this was not reflected in the fatigue analysis? Especially since this was one of the hypotheses of the study could you also please discuss it more comprehensively in the discussion section?

This has been expanded upon in the abstract and now reads:

"FSHD patients self-reported that the 6-minute walk test was the most difficult, however, the delta fatigue score was not different between the gait tasks but had a moderate effect size compared to the 12 meter conservative walking. This is most likely due to the small and heterogeneous sample size but indicates the 6-minute walk test may be more physically demanding."

This has been expanded upon in the discussion and it now reads:



It is the author's expectation that with a larger sample size and less heterogeneous population, the 6-min walk test self-reported delta fatigue score effect size would reduce. However, the moderate effect size should not be ignored, as it is a warning to clinicians that the 6-min walk test is physically demanding and could be unsafe for more affected FSHD patients. This study included individuals who could complete the 6-min walk test safely unsupervised by the study team. Future research should carefully consider if this test is necessary and appropriate due to the potential fall risk when not conducted in a highly controlled environment.

Methods

 The authors mention: Each gait task trials across the 5 days were ensemble averaged by each week for an overall velocity, cadence, and delta fatigue score (post-pre).
Descriptive statistics were calculated for **both** variables to create a direct comparison of mean and standard deviation along with assessment of the **normality** of the data.



- a. Are there 2 or 3 variables (since you say both)? maybe specify here that you only mean cadence and velocity (as you in the previous sentence you also mention fatigue)
- b. Could you please mention which test was used to assess normality?

Thank you for this very important comment. We have amended the methods per your recommendations and they now read:

"Descriptive statistics were calculated for all three variables (velocity, cadence and delta fatigue score) to create a direct comparison of mean and standard deviation along with assessment of the normality of the data using skewness and kurtosis. If any variable was ± 2.0 for skewness or kurtosis, it was considered abnormally distributed. From the descriptives, the data velocity and cadence data were considered parametric and without influential skewness while the delta fatigue score was skewed."

2) Could you please mention the sample size for each variable used for the statistical comparison? Essentially how many data points compose the boxplots you present in your figures?

Thank you for this comment and we are happy to clarify the sample size for each variable. For the variable, there were 5 days of measurements which resulted in:

T1 = 60 total trials, 60 data points that were ensemble averaged

T2 = 60 total trials, 60 data points that were ensemble averaged (this is due to the morning and afternoon testing)

T3 = 5 total trials, 5 data points that were ensemble averaged

This numbers are reflected within the methods and it now reads:

"Each gait task trials across the 5 days were ensemble averaged by each week (T1=60 trials, T2=60 trials and T3=5 trials that were ensemble averaged) for an overall velocity, cadence, and delta fatigue score (post-pre)."

Discussion

1) Sometimes the authors say slower and sometimes lower when referring to velocity. Maybe pick one for consistency?

Thank you for this comment. The term "lower" has been adjusted to "slower" and is reflected throughout the manuscript.

- 2) The authors mention: *We had no ability to control the patient's current athletic ability including weekly exercise programs and it is possible that some individuals lead healthier lifestyles.*
 - a. This is absolutely reasonable. However, do you have any suggestions of study protocols that may overcome this limitation. And if yes could you please link them to your future work?

Thank you for this comment. We have amended the limitations to expand on this limitation and it now reads:

Additionally, if the participants were not actively exercising this at-home walking protocol may have promoted physical activity and increased their gait capacity over time. The use of numerous trials for each protocol may have washed out any increase in gait capacity. Our

Journal of Clinical and Translational Research Peer review process file 09.202302.004



future research will pursue a standard exercise protocol and/or an exercise log to track exercise capacity in order to control for exercise and reduce it as potential confounder in statistical analysis.

2nd Editorial decision 23-Feb-2023

Ref.: Ms. No. JCTRes-D-22-00223R1 Spatiotemporal gait and fatigue do not change when using common at-home gait tasks in patients with Facioscapulohumeral Muscular Dystrophy - A pilot study Journal of Clinical and Translational Research

Dear authors,

I am pleased to inform you that your manuscript has been accepted for publication in the Journal of Clinical and Translational Research.

You will receive the proofs of your article shortly, which we kindly ask you to thoroughly review for any errors.

Please notify our assistant editor/production editor when you receive the proofs if your article should belong to a special issue specifying the issue's title.

Thank you for submitting your work to JCTR.

Kindest regards,

Michal Heger Editor-in-Chief Journal of Clinical and Translational Research

Comments from the editors and reviewers: