TEST-RETEST RELIABILITY OF GAIT TESTS IN A DANISH RETT SYNDROME POPULATION

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INTRODUCTION
Rett Syndrome (RTT) is a rare innate neurologic development deficit, which mainly affects females. It occurs worldwide in 1 of every 10,000 female births. In the majority of cases, Rett syndrome is caused by a mutation in the MECP2 gene. In Denmark, 110 individuals aged 2-61 years are diagnosed with Rett syndrome and 95 of them have a MECP2 mutation. The syndrome causes physical and mental disabilities. The girls apparently develop normally during the first 6-18 months of life. After a period of regression, the girls lose both fine and gross motor skills along with communicative skills\cite{1,2}. Due to their physical and mental deficits, they have difficulties with initiation of movements and to follow instructions. In the literature and in practice a 10-meter walk test is frequently used to evaluate gait speed in a broad range of patient groups. However, the usability of this test to evaluate effects of interventions is questionable. Especially in patients who are easily exhausted, a 4-meter walk test may be more feasible and useful.

CLINICAL SIGNIFICANCE
For the RTT population very few reliable and valid test exist. Because it is important to be able to evaluate the efforts to maintain function, particularly tests that can evaluate changes in condition over time are warranted. Therefore, the purpose of this study was to evaluate the reliability of the 10- and 4-meter walk test. Furthermore, we wanted to investigate if the 4-meter walk test was an ample replacement for the 10-meter walk test.

METHODS
Sixteen participants (all female and 5-45 years) with RTT and a MECP2 mutation were included in the study. 8 were children (median age: 7.5 years) and 8 were adults (median age: 34 years). All participants or guardians signed informed consent prior to inclusion in the study. Participants were excluded if they had received botox injections within three months prior to the tests, leg surgery within six months, and back surgery within 12 months. Three trials of both 4-meter and 10-meter walk tests were performed on two test occasions one week apart. One participant used a walking aid. Three participants walked without support from another person. All tests were performed in the participants school or activity center to make the participants feel more comfortable.
For assessment of intra- and interday reliability Intraclass Correlation Coefficients $2,k$ (ICC) were applied along with inspection of Bland-Altman plots. The agreement between 10-meter and 4-meter tests was investigated with Pearson correlation coefficient.

RESULTS
The average intraday ICC for the 4-meter walk test ranged from .77-.88 and .88-.92 for the 10-meter walk test. The interday ICC was .91 for the 4-meter walk test (Fig. 1) and .94 for the 10-meter walk test. All ICCs were significantly different from 0 at $p<0.0001$

The agreement between the two tests was .93 ($p<0.0001$) (Fig. 2).

DISCUSSION
We found high reliability in both the intra- and interday setting for both walk tests. These findings support the use of both the 10-meter walk test and 4-meter walk test to assess walking velocity in patients with RTT. The results need to be verified in a larger sample, which will allow further analysis of the standard error of measurement and minimal detectable change. We recommend the use of a practice attempt before the actual testing to account for a possible learning effect.

Furthermore, the 4-meter walk test can replace the 10-meter walk test in a RTT-population. Therefore, if the condition of the surroundings or the patients do not allow for a 10-meter walk test to be performed, a 4-meter walk test could be used instead.

REFERENCES

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DISCLOSURE STATEMENT
There have been no conflicts of interest.