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MODIFIED DYNAMIC GAIT INDEX AND LIMITS OF STABILITY IN MYOTONIC DYSTROPHY TYPE 1

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ABSTRACT: *Introduction*: The purpose of this study was to describe and compare the performance of balance and walking tests in relation to self-reported fall history in adults with myotonic dystrophy type 1 (DM1). *Methods*: Twenty-two (13 male) participants with DM1 completed, a 6-month fall history questionnaire, the modified Dynamic Gait Index (mDGI), limits of stability (LoS) testing, and 10-m walking tests. *Results*: Mean (SD) falls in 6 months was 3.7 (3.1), and 19 (86%) participants reported at least 1 fall. Significant differences in mDGI scores ($P = 0.006$) and 10-m fast walking gait velocity ($P = 0.02$) were found between those who had been classified as "fallers" and those who had been classified as "nonfallers." Significant correlations were found between mDGI scores and 10-m walking time. *Discussion*: Falls are common in DM1, and the mDGI may have potential to distinguish fallers from nonfallers, whereas the LoS failed to detect such impairment. Future studies should further explore use of the mDGI in DM1.

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Myotonic dystrophy type 1 (DM1) is the most commonly inherited adult muscular dystrophy.[1,2] DM1 is characterized by progressive distal muscular weakness, myotonia, and early onset cataracts, although this multisystemic disorder affects many other organ systems.[3–8] Consequently, adults with DM1 often display alterations in gait kinematics, postural instability, balance impairment, and falls.[4–7,9–15] Falls, and even stumbles, are a major public health concern because of the resultant morbidity and mortality that often follows.[16–20] Furthermore, falls are a widespread problem in adults with neuromuscular conditions and may occur up to 10 times more often in persons with DM1.[21–23] Previous studies using 10-m walking tests have described the extent to which impairments in strength, balance, and gait may lead to falls in persons with DM1.[4,5,23,24] However, most measures of balance and falls in studies involving DM1 have been limited to select self-reported inventories or measures such as the timed up and go, step test, or the Berg balance scale.[4,5,12–15,25,26]

The modified Dynamic Gait Index (mDGI) and limits of stability (LoS) testing are two highly studied, reliable, and attractive balance measures for neurological populations.[27–41] The mDGI is a measure that can identify balance impairments that may lead to falls during locomotion and may possess the ability to detect such deficits in adults with DM1.[27,28,37,38] Similarly, LoS testing has been shown to identify impairments in postural stability and balance that can result in a fall and is also an attractive measure for persons with DM1.[25,29,33–36,41–45]

Prior studies have demonstrated the ability of the mDGI and LoS testing to distinguish those who have been classified as "fallers" from those who have been classified as "nonfallers" and to identify fall risk and balance impairment among healthy populations, older adults, and persons with neurological disorders.[25,32,39,41–45] Bachasson *et al.*[9] found strong relationships among postural instability, lower extremity weakness, and impaired locomotion in persons with DM1, and these could potentially be detected by the mDGI and LoS testing. Therefore, both the mDGI and LoS testing may have the potential to identify fallers in the DM1 population. However, currently, there are no published studies examining the use of either the mDGI or LoS testing in this population. Therefore, this study seeks to (1) further characterize adults with DM1 by describing performance on the mDGI, LoS testing, 10-m walking tests, and self-reported fall history; and (2) compare the results among outcome measures.

MATERIALS AND METHODS

The University of Utah Institutional Review Board approved all study procedures. Convenience sampling was used to select

Key words: balance, falls, limits of stability, modified dynamic gait index, myotonic dystrophy

Abbreviations: CoG, center of gravity; CTG, cytosine-thymine-guanine; DM1, myotonic dystrophy type 1; LoB, loss of balance; LoS, limits of stability; mDGI, modified Dynamic Gait Index; MXE, maximum excursion score

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Conflicts of Interest: N. E. Johnson has received research support from Valerion Therapeutics, Ionis Pharmaceuticals, and Biogen Idec. R. J. Butterfield serves on the scientific advisory boards for Bamboo Therapeutics, Sarepta Therapeutics, Marathon Pharmaceuticals, and Avexis. He is the site principal investigator for clinical trials sponsored by Marathon Pharmaceuticals, Sarepta Therapeutics, Pfizer, Eli Lilly, PTC Therapeutics, and aTyr Pharma. The remaining authors have no conflicts of interests to disclose.

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participants between the ages of 18 and 60 with genetic confirmation of DM1. Study participants were recruited through self-selection and direct recruitment options at the University of Utah Neuromuscular Clinic. All participants provided written informed consent prior to enrollment in the study.

Inclusion and Exclusion Criteria. Participants were included if they met the set criteria for adult-onset DM1 in this study: having reported onset of symptoms of DM1 after age 12 and possessing confirmed genetic testing of greater than 50 cytosine-thymine-guanine (CTG_n) repeats in the *DMPK* gene,[1] being ambulatory, and having no other major medical illnesses that would interfere with the study procedures. Participants were excluded from this study if they were taking mexiletine (an antimyotonia medication), had any other non-DM1 illness that could interfere with the ability to complete the study procedures, or had significant trauma within 1 month prior to the study.

Functional Measures and Assessments. Outcome measures for this study included the mDGI, LoS testing, and a 1- and 6-month participant-reported fall history. The 10-m fast walking test outcome measure was included to relate the results of this study to the prior literature involving balance and falls in DM1.

Modified Dynamic Gait Index. The mDGI measures a person's ability to adapt sensorimotor and cognitive systems to negotiate safely a variety of environments and tasks commonly required during household or community ambulation.[27,28,37,38] Prior studies have documented the ability of the mDGI to distinguish between fallers and nonfallers for other neurological diagnoses and highlight the discriminative power of the each of the 8 items of this tool.[27,37,38] The mDGI examines 3 major constructs of dynamic balance during locomotion: time to complete a task, gait pattern, and level of assistance required.[27,28,37,38] The 8 items of the mDGI vary task complexity and physical demand by changing speed and direction, walking over and around obstacles, and ascending stairs.[38] The 8 tasks in the mDGI were designed to evaluate the interactions between a participant's visual, vestibular, and sensory systems and the environment.[27,38] Prior studies have demonstrated excellent psychometric properties across a variety of populations.[27,38,40,44]

Participants in this study performed the mDGI in a quiet, 25-m long, noncarpeted, 2-m wide hallway at the direction of a trained clinical evaluator. Scoring consisted of the time required to complete each item (0–24), alteration in gait patterns (0–24), and required level of assistance (0–16) for each individual task. Thus, the mDGI scoring ranges from 0–64, where zero indicates the inability to complete any items successfully, and 64 indicates no observable impairment.[27]

100% LoS Testing. The NeuroCom SMART Balance Master (NATUS Medical, Seattle, Washington)[45] was used in this study. This is a computerized device that generates quantitative data on a participant's postural stability that is not easily ascertained by examination.[45] The 100% LoS maximum excursion (MXE) testing feature of this machine was used. Limits of stability MXE testing can identify abnormalities in a person's volitional motor control of balance and postural sway.[25,29,33–36,41,45] Limits of stability testing is a widely studied, valid, and reliable assessment of dynamic balance.[41,42,45] Results from LoS testing have proved useful in indicating when a person is at risk for fall and requires rehabilitative care.[41] There are several scores produced as a result of LoS testing, including reaction time, movement speed, endpoint excursion, directional control, and MXE.[45] For the purposes of this study, the LoS MXE score in each direction (anterior, posterior, right, and left) was used as a representation of dynamic balance for each participant. The LoS MXE is the maximum theoretical limit of voluntary excursion in an individual's center of gravity (CoG) away from their base of support in the anterior, posterior, lateral, and diagonal directions without loss of balance (LoB), taking a step, or falling.[42,45] Maximum excursion scores are calculated on the basis of a participant's height and each plane of movement through computerized posturography, whereby lower MXE scores represent decreased ability to control one's excursions in CoG.[41,42,45]

History of Falls Questionnaire. Each participant subjectively reported the number of falls they had experienced in the past 1 and 6 months during a baseline study visit. For the purposes of this study, falls were defined as any LoB that resulted in unintended contact with the ground. Participants were categorized as being either a faller (1 or more falls in the prior 6 months), or a nonfaller (no self-reported history of falls in 6 months).

10-Meter Walking Tests. Participants performed the 10-m walk and fast walking tests as described in prior studies in neuromuscular disorders.[4,5,46,47] The 10-m fast walking test was conducted by using a "cold start" and a "flying finish" method in a quiet, noncarpeted hallway along a 25-m track labeled at 1-m intervals.[48] Participants completed 3 consecutive trials of the test, and the best score and averages were calculated. Participants were given a maximum of 60 s to complete each test. If a subject was unable to complete the task, the time was not recorded.

Statistical Analysis. Descriptive and inferential statistics were performed. Mann–Whitney *U* tests were performed to compare the outcome measures between fallers and nonfallers because the data set did not conform to the assumptions of a normal distribution. Spearman's ρ correlation coefficients were calculated to determine the relationships between outcome measures for results that did not conform to normal distributions. Post hoc Bonferroni corrections to set levels of significance were performed for analyses with multiple comparisons. All analyses were conducted in SPSS 24 (IBM, Armonk, New York), and $P < 0.05$ was considered significant.

RESULTS

Demographics. This study enrolled a total of 22 adult participants (13 men, 9 women) with DM1. Mean (SD) age was 39.6 (8.4) years. Detailed participant demographic data including age, sex, fall history, and CTG_n repeat length are provided in Table 1. Significant differences were noted between men and women in CTG_n repeat length for this sample.

Falls and Balance. The mean (SD) number of falls reported in 6 months was 3.7 (3.1), and the number of falls reported in the prior 1 month was 1.2 (1.2). In this sample, in total, 19 (86.4%) participants met the definition of a faller and reported at least 1 occurrence of a fall in the previous 6 months (Table 1). No statistically significant associations were noted between 10-m walk/fast test and self-

Table 1. Participant characteristics

Variables	DM1	Men	Women	*P* value*
Sample, *n* (%)	22 (100)	13 (59.1)	9 (40.9)	n/a
Age, y, mean (SD)	39.6 (8.4)	38.0 (5.9)	41.9 (11.1)	0.36
CTG_n repeat length, mean (SD)	566.0 (237.6)	679.6 (193.8)	367.3 (173.8)	0.026†
Falls in prior 6 mo, mean (SD)	3.7 (3.1)	3.7 (3.7)	3.6 (2.2)	0.92
Falls in prior 6 mo, median (range)	3.0 (0–12)	3.0 (0–12)	3.0 (0–8)	
Falls in prior mo, mean (SD)	1.2 (1.2)	1.4 (1.4)	1.0 (0.9)	0.47
Falls in prior mo, median (range)	1.5 (0–4)	2.0 (0–4)	1.0 (0–2)	
Age of symptom onset, mean (SD)	18.9 (8.7)	17.1 (7.7)	21.4 (10.0)	0.33

CTG_n, cytosine-thymine-guanine; DM1, myotonic dystrophy type 1.

**Mann–Whitney U tests were performed between men and women.*

†$P < 0.05$.

reported fall history at either 1 or 6 months. However, gait velocity on the 10-m fast walking test revealed significant differences between fallers and nonfallers (Table 2). Results for the mDGI and LoS balance measures for adults with DM1, categorized as either fallers or nonfallers, are described by individual testing components in Table 2. The mDGI total score, time, and gait pattern scores were all significantly different between fallers and nonfallers with DM1. A correlation matrix comparing the results for each outcome measure can be found in Table 3. Strong significant associations were found between mDGI scores and 10-m walking test times. A modest, albeit insignificant, relationship was found between the mDGI total score and the LoS MXE composite score. A breakdown of the relationships between testing components of the mDGI and the LoS testing can be found in Table 4. Significant relationships were identified between the mDGI time, gait pattern, and total score with the LoS MXE anterior score and also between LoS MXE composite and the mDGI gait pattern scores. In addition, a significant relationship was noted between the LoS left lateral MXE scores and the mDGI gait pattern and total scores (Table 4). No additional significant relationships were detected between the two measures of balance. The relationships among 6-month fall history, mDGI total score, and LoS MXE composite score by age and sex is shown in Figure 1. The relationship between 10-m fast gait velocity and mDGI scores in relation to self-reported 6-month fall history is illustrated in Figure 2. A modest amount of variance can be seen in both Figure 1 and Figure 2 with respect to the 6-month fall history data superimposed on each data point.

DISCUSSION

The purpose of this study was to describe and compare individual's performance on the mDGI, LoS, and 10-m walking tests in relation to self-reported fall history for adults with DM1. Prior literature has described balance impairment and a high risk for falls in persons with DM1. However, a tool to prospectively identify or measure these constructs in this population have yet to be reported. This study provides evidence that the mDGI may be a useful clinical outcome measure to identify falls and balance impairment in DM1. As such, the mDGI is worthy of additional investigation.

The mDGI and LoS measures have been highly studied in other populations and are well regarded for their ability to detect balance impairments. In this study, significant differences were found between fallers and nonfallers by the mDGI. Adults with DM1 who had reported a history of falls performed significantly lower on the mDGI time, gait pattern, and overall scoring components. This suggests that the individual scoring domains of the mDGI may be able to distinguish adequately fallers from nonfallers. In addition, an association was found between the

Table 2. Balance measures for DM1 fallers and nonfallers

Outcome measure	Fallers, *n* = 19, mean (SD)	Nonfallers, *n* = 3 mean (SD)	*P* value
mDGI total score, 0–64	43.2 (9.7)	59.8 (5.3)	0.006*
mDGI time score, 0–24	12.1 (4.5)	21.0 (4.1)	0.010*
mDGI gait pattern score, 0–24	16.7 (4.2)	22.8 (1.5)	0.016*
mDGI level of assistance score, 0–16	15.0 (2.6)	16.0 (0.0)	0.392
LoS MXE composite score	55.7 (14.5)	66.3 (24.6)	0.406
LoS MXE anterior score	45.6 (27.1)	65.0 (31.1)	0.209
LoS MXE posterior score	27.0 (18.8)	45.7 (30.4)	0.314
10-m fast gait velocity, m/s	0.9 (0.3)	0.6 (0.1)	0.020*
10-m walk gait velocity, m/s	0.9 (0.2)	1.2 (0.3)	0.148

DM1, myotonic dystrophy type 1; LoS, limits of stability; mDGI, modified dynamic gait index; MXE, maximum excursion.

**P < 0.05, Mann–Whitney U test.*

Table 3. Spearman correlation matrix of outcome measures

Measure	mDGI total score	LoS MXE composite score	10-m fast time, s	10-m walk time, s	Falls in prior 1 mo	Falls in prior 6 mo
mDGI total score	1.0					
LoS MXE composite score	0.6	1.0				
10-m fast time, s	−0.9*	−0.6	1.0			
10-m walk time, s	−0.8*	−0.6	0.9*	1.0		
Falls in prior mo	−0.2	−0.1	0.4	0.3	1.0	
Falls in 6 mo	−0.4	0.01	0.5	0.3	0.7*	1.0

LoS, limits of stability; mDGI, modified dynamic gait index. MXE, maximum excursion

**P < 0.025, displayed with Spearman's ρ values after post hoc Bonferroni corrections.*

individual scoring domains of the mDGI and the 10-m walk/fast time; this could be the result of an overlap in the functional constructs being measured. For example, many of the items on the mDGI include a forward propulsion component of gait, such as the initiation of stepping and acceleration, that could explain the strong correlations noted in this study. The 10-m fast walking test was similarly able to distinguish fallers from nonfallers in this sample; this supports findings from the prior literature that implicate gait speed as a strong predictor of falls.[17,18] However, the results of the 10-m walk test in this sample of adults with DM1 do not concur, yet our sample did perform poorly on both 10-m walking tests compared with previously established norms.[17,18,48]

In future studies, it may be more clinically meaningful to analyze fall risk in place of dichotomous "faller" and "nonfaller" categories. Normative values and fall-risk cutoff scores for gait velocity on the 10-m walk and fast walking tests have been previously established in the literature for other neurological diagnoses.[17,18,21,44] However, minimal clinically important differences, standard error of measurement, fall-risk cutoff scores, and floor or ceiling effects for the mDGI are largely unknown across neuromuscular disorders.[38] Therefore, a better understanding of the psychometric properties and measures of responsiveness for the mDGI will contribute to the understanding of the mDGI's usefulness in the DM1 population and should be a subject of future inquiry.

Although large differences existed in our sample, no statistically significant findings were noted in the LoS MXE scores between fallers and nonfallers. This could be the result of compensatory maneuvers or strategies to avoid a fall during testing, such as approaching the LoS with decreased velocity and/or with increased path variation, as previously reported in other neurological populations.[25,31,33,34] It is unclear why significant relationships were found between LoS left lateral MXE and both the mDGI gait pattern and total scores in our sample; these findings could be the result of measurement error. However, a strong significant association was found between the anterior LoS MXE and the performance of the mDGI time, gait pattern, and total score components despite a lack of a correlation to fall history. Anticipatory actions of forward propulsion could be signals on the anterior LoS MXE for sway and postural control. Previous studies have documented that anterior ankle strength is greatly affected in DM1 and, as a result, contributes to gait abnormalities and increased fall risk.[5,9,49] This may suggest that ankle dorsiflexion strength and the lack of an adequate ankle righting reaction contribute to posterior LoB and falls and perhaps have less to do with the tasks assessed on the mDGI. However, it is unclear which factors can explain such differences among the LoS MXE scores in this sample, and we caution against drawing conclusions.

Limitations to this study include, but are not limited to, a small sample, a cross-sectional nature, and the potential for the inaccuracy in self-reported participant fall history. Therefore, we can neither draw conclusions regarding the broader population of DM1 nor fully elucidate any true relationships because of the narrow scope of this study. A large majority of this sample reported a history of falls, and this could have had an effect on the outcomes by entering a bias toward fallers in the results and analysis. Limitations inherent to the phenotype of DM1, such as ankle dorsiflexion weakness or apathy, could have also played a role in confounding the measurement and collection of outcomes and, in turn, the validity of results.

Table 4. Comparison of the individual scoring components of the mDGI and LoS

Balance measure	LoS anterior MXE	LoS posterior MXE	LoS right lateral MXE	LoS left lateral MXE	LoS MXE composite score
mDGI time score, 0–24	0.7*	0.3	0.3	0.5	0.6
mDGI gait pattern, 0–24	0.8*	0.3	0.6	0.7*	0.7
mDGI level of assist, 0–16	0.4	−0.1	−0.1	0.2	0.2
mDGI total score, 0–64	0.7*	0.2	0.3	0.6*	0.6*

LoS, limits of stability; mDGI, modified dynamic gait index; MXE, maximum excursion.

**P < 0.025, significant results displayed with Spearman's ρ values after post hoc Holm–Bonferroni corrections.*

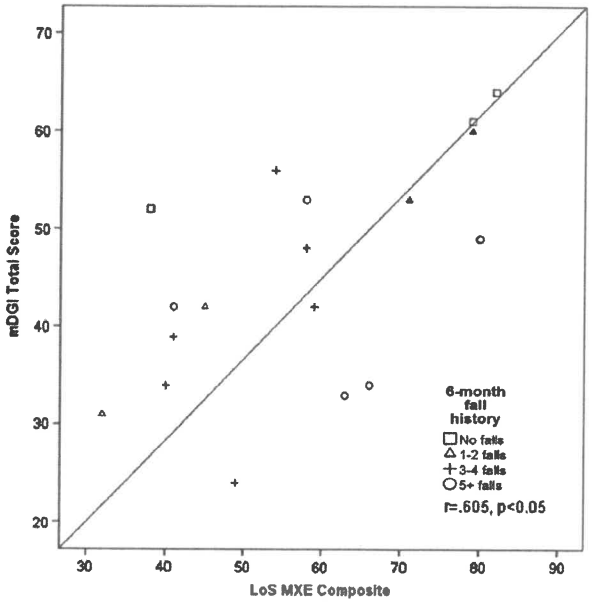

FIGURE 1. mDGI and LoS composite scores in DM1. This figure demonstrates the relationship between the mean scores of the mDGI and the LoS MXE composite scores for our sample of persons with DM1. There appears to be agreement between the mDGI and the LoS MXE measures. This figure also overlays 6-month history of falls in relationship to mDGI and LoS MXE scores. This indicates that lower mDGI and LoS MXE scores were generally associated with higher self-reported frequency of falls. Geometric symbols (boxes, triangles, plus signs, circles) represent the individual self-reported fall history for each participant. DM1, myotonic dystrophy type 1; LoS MXE, limits of stability maximum excursion scores; mDGI, modified Dynamic Gait Index (score 0–64).

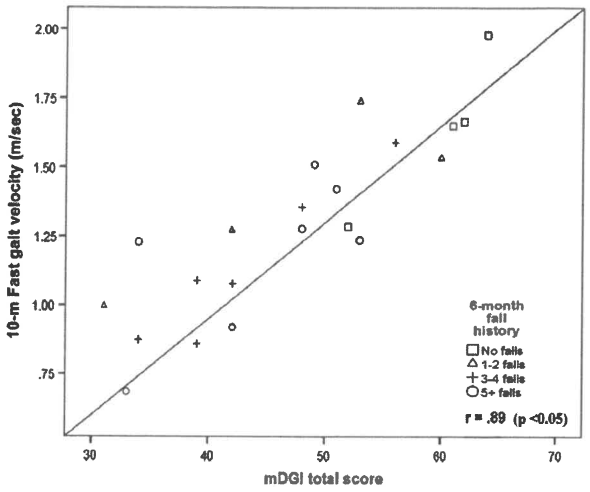

FIGURE 2. mDGI total scores and gait velocity on the 10-m fast test for adults with DM1. This figure illustrates the strong relationship between a decreasing gait velocity and decreasing balance as measured by the mDGI score. In our sample, there was a significant correlation between decreased performance on the mDGI and 10-m timed walking test with respect to 6-month fall history. This figure also overlays 6-month history of falls per individual data point to highlight the relationship between balance impairment and gait speed that results in falls. Geometric symbols (boxes, triangles, plus signs, circles) represent the individual self-reported fall history for each participant. DM1, myotonic dystrophy type 1; mDGI, modified Dynamic Gait Index.

Furthermore, comparisons in our study are greatly limited by the small number of nonfallers noted in this sample. Because of the limited size and scope of this study, the full scope of the mDGI and LoS tests cannot yet be fully understood. Therefore, future studies should seek to validate these outcome measures in the adult DM1 population.

Furthermore, the accurate capture of falls data has historically been a challenge for both practitioners and researchers across age, sex, and diagnosis.[16,19] Prior studies indicate that only a small percentage of falls are accurately reported by participants regardless of the population; this may have interacted with our results.[16,19] Future research could improve methodology by providing participant and practitioner education to establish a consistent definition of falls that is supported by the current literature by encouraging participant fall journals, requiring more in-depth qualitative inquiry on fall incidence and characteristics, and incorporating meaningful use of technology to record fall data.

Overall, the mDGI appeared to be a feasible outcome measure in the adult DM1 population and demonstrated an adequate ability to distinguish between fallers and nonfallers across multiple scoring domains. A history of falls was associated with decreased performance on the mDGI and 10-m fast gait velocity. Balance impairments that were identified by the mDGI and the 10-m fast walking tests went undetected by the LoS measures. This indicates that, for adults with DM1, an important relationship may exist between fall risk and balance as measured by the mDGI, and this relationship should be the subject of future inquiry.

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Ethical Publication Statement: We confirm that we have read the Journal's position on issues involving the ethical reporting and affirm that this report is consistent with those guidelines.

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