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What influences changes in gait spatiotemporal parameters in boys with Duchenne Muscular Dystrophy? Focusing on contractures, muscle strength, and gait deviations

Anita Bagley¹ , Susan Sienko² , Cathleen E. Buckon², Kent Heberer³, Loretta Staudt³ , Eileen Fowler³ , Jason Newsom⁴ , Craig McDonald^{1,5} , Michael D. Sussman²

¹Shriners Children's Northern California, Sacramento, CA 95817, USA.

²Shriners Children's Portland, Portland, OR 97239, USA.

³Department of Orthopaedics, University of California, Los Angeles, CA 90025, USA.

⁴Department of Psychology, Portland State University, Portland, OR 97207, USA.

⁵Department of Physical Medicine, University of California Davis Medical Center, Sacramento, CA 95817, USA.

Correspondence to: Anita Bagley, Clinical Research, Shriners Children's Northern California, 2425 Stockton Blvd, Sacramento, CA 95817, USA. E-mail: abagley@shrinenet.org

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Abstract

Aim: The absence of dystrophin in patients with Duchenne Muscular Dystrophy (DMD) causes muscle fiber necrosis and fibrosis and eventually muscle weakness and contractures. This study aimed to investigate the timing and extent of lower limb contracture development and to examine how contractures, muscle strength, and kinematic (e.g., limb position or joint movement) deviations relate to longitudinal changes in gait spatiotemporal parameters.

Methods: Seventy-five ambulatory boys with DMD participated in this prospective longitudinal study of contractures, isometric muscle strength, and gait analysis. Nonlinear mixed modeling (NLMIXED) exponential growth curve models were developed to investigate the effects of contractures, muscle strength, and kinematic deviations on longitudinal changes in gait spatiotemporal parameters.



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Results: Ankle plantar flexion contractures were present as early as 4 years of age. Hip flexion contractures, assessed by the Thomas test, and knee flexion contractures, assessed by knee extension in supine, developed around 13 years of age and did not exceed ten degrees. Over time, gait speed, stride length, and cadence decreased. Statistically significant covariates for gait speed included hip flexor contracture (limitation in hip extension), hip extensor and ankle plantar flexor strength, and pelvic tilt Gait Variable Score (GVS). Plantar flexor strength was a significant covariate for stride length, while pelvic obliquity GVS significantly correlated with cadence.

Conclusion: Subtle changes in hip extensor and ankle plantar flexor strength, along with pelvic range of motion during gait, are indicators of DMD disease progression. Gait analysis, including easily accessible measures of gait speed and normalized stride length, may provide a more sensitive indicator of DMD disease progression than manual assessments of contractures and muscle strength.

Keywords: Duchenne muscular dystrophy, longitudinal lower limb contractures, gait spatiotemporal parameters

INTRODUCTION

Duchenne Muscular Dystrophy (DMD) is an X-linked muscle disorder caused by a mutation in the dystrophin gene, affecting approximately 1:3500 male births^[1]. The mutation in this gene compromises the myofibers of the muscle, making them susceptible to damage and leading to repeated bouts of myofiber necrosis and regeneration^[2]. Consequently, normal muscle contractions injure the sarcolemmal membrane and trigger recurrent inflammatory responses. Chronic inflammation causes muscle fiber necrosis and fibrosis, and eventually contractures^[3].

As DMD progresses in boys, muscle weakness and contractures develop in the first decade. Contractures are defined as limitations in the passive range of motion. To maintain walking, boys adopt compensatory gait strategies, such as reduced knee flexion in stance to lessen the demand on the quadriceps and lumbar hyperlordosis to shift the trunk mass over the hip joint center^[4]. By approximately eight years of age, boys with DMD typically have difficulty with independent ambulation and associated tasks such as rising from the floor and climbing stairs^[5]. Early initiation of corticosteroid treatment has been shown to extend ambulatory ability by two to three years^[6]. Furthermore, novel gene-based treatments^[7-9], including those utilizing CRISPR, are in development and, in some cases, have already entered clinical trials or approval. These therapies have the potential to mitigate disease manifestations and alter the clinical course of DMD.

Boys with DMD generally exhibit reduced gait speed compared to typically developing peers, primarily due to reduced stride length^[10]. The progression of both muscle weakness and lower extremity joint contractures contribute to the deterioration of walking spatiotemporal parameters, specifically gait speed (meters per second), stride length (meters), and cadence (steps per minute). However, limited data exist on concomitant longitudinal changes in lower extremity contractures in a large cohort of boys with DMD assessed using a standardized methodology. This paper aims to address this gap by providing data on lower extremity contracture progression and linking these changes to alterations in gait parameters. Additionally, volitional muscle strength measures^[11] from the same cohort were analyzed to explain the variability in longitudinal changes in gait speed, stride length, and cadence. Finally, changes in Gait Variable Scores (GVSSs)^[12] were examined to relate kinematic changes (e.g., limb position or joint movement) to variance in gait spatiotemporal parameters. Parsing out the contribution of these factors to changes in walking speed, stride length, and cadence may inform targeted treatment strategies.

The specific goals of this study were to: (1) investigate longitudinal changes in lower extremity contractures, determining which joints are affected earliest and most severely; (2) determine which contractures explain variance in longitudinal changes in gait speed, cadence, and stride length; (3) determine which muscle strength measures account for variability in these gait parameters; and (4) determine which GVS measures explain variance in longitudinal changes in gait speed, cadence, and stride length.

METHODS

Subjects

In this eight-year prospective longitudinal study, 85 boys with DMD aged 4-16 years at enrollment were recruited from clinics at three participating hospitals: 32 boys from Shriners Children's (SC) Portland, 33 from SC Northern California, and 20 from the University of California, Los Angeles (UCLA). Between 2006-2010, only SC-Portland and SC-Northern California participated, with subjects evaluated annually. In 2010, UCLA joined the study, and all remaining participants were then assessed every 6 months until study completion in 2014.

Inclusion criteria included a typical clinical presentation of DMD and at least one of the following: documentation of a disease-causing mutation in the dystrophin gene, elevated serum creatine kinase levels, or a family history of an affected relative with a disease-causing dystrophin mutation and/or complete dystrophin deficiency confirmed by muscle biopsy immunostaining. Clinical notes were reviewed for evidence of typical DMD features, such as delayed walking or abnormal gait, calf hypertrophy, and a positive Gower's sign. Exclusion criteria were inability to walk independently, cognitive inability to follow testing instructions, and age under four years.

All participants and their parents or guardians provided informed consent and/or assent as required by each Institutional Review Board.

Of the 85 boys enrolled, nine were excluded due to inability to complete gait analysis, and one was excluded because genetic testing and clinical manifestation confirmed Becker muscular dystrophy. Among the 75 subjects retained in the study, the mean baseline age was 88.8 months (SD 30.1 months), the mean height was 117.4 cm (SD 14.6 cm), and the mean weight was 26.6 kg (SD 12.7 kg).

For contracture assessment, three additional boys were excluded due to missing data resulting from a shortage of physical therapy staff at one site, yielding a total of 72 subjects for these analyses. For muscle strength assessment, 18 boys were further excluded due to non-compliance or insufficient strength to generate the minimum torque required for the Biodex system, resulting in 57 subjects for these analyses. All 75 boys were included in the GVS assessment.

Contractures

Contracture development was assessed by measuring the passive range of motion in the hips, knees, and ankles. A manual detailing the passive range of motion protocols was developed, and evaluators received in-person training with the lead investigators before data collection. Bilateral measurements included: hip abduction, hip extension (Thomas test^[13]), knee extension, popliteal angle, ankle dorsiflexion with the knee flexed and extended, and ankle plantar flexion. Measurements were performed by trained personnel using a goniometer, following the guidelines in *Measurement of Joint Motion*^[13]. Left side data were used for analysis, as no significant asymmetry was found between sides. Longitudinal contracture data were calculated relative to age-matched (but not sex-matched) normative data from Mudge et al.^[14] for hip abduction and hamstring/popliteal measures, and from Liyanarachi et al.^[15] for ankle range of motion. Hip

extension (Thomas test) and knee extension were referenced to baseline measurements of zero degrees. The popliteal angle was defined as the tibial angle in degrees from vertical.

Volitional muscle strength

Volitional muscle strength was assessed using a Biodex System 3 Pro isokinetic dynamometer, which measures force output at different speeds and ranges of motion. Subjects were seated and positioned appropriately for each test. For example, during knee testing, the dynamometer's center of rotation was aligned with the knee joint, and the dynamometer arm was aligned parallel to the shank. Selected muscle groups were assessed unilaterally (based on dominant handedness), as previous studies reported comparable strength bilaterally in boys with DMD^[16,17]. Isometric hip extensor strength was assessed in supine with the hip at 85° flexion. Isometric knee flexor strength was assessed in sitting with the knee at 30° flexion, while knee extensor strength was assessed at 90° flexion. The isometric hip and knee protocol consisted of three consecutive five-second contractions per muscle group, with 10-second rests in between. Isokinetic concentric knee extensor strength was assessed at 60°/sec, with three consecutive arcs of extension and flexion through the full voluntary range. Isometric ankle plantar flexion was assessed in a semi-reclined position with the ankle at 5° plantar flexion, using three five-second contractions into plantar flexion. The testing order was standardized: ankle first, followed by knee, then hip. Peak torque output for each muscle group and contraction type was used in analyses, and all torque values (Nm) were normalized by body weight (kg). Longitudinal muscle strength data have been published previously^[11].

Gait variable scores and spatiotemporal parameters

Joint kinematics were collected using either a VICON 612 system (SC-Portland) or a Motion Analysis Corporation (SC-Northern California, UCLA) 3-D system. Kinematic data included pelvic tilt (sagittal plane), pelvis obliquity (coronal plane), pelvic rotation (transverse plane), hip flexion/extension (sagittal), hip abduction/adduction (coronal), hip rotation (transverse), knee flexion/extension (sagittal), ankle plantar/dorsiflexion (sagittal), and foot progression angle (transverse). All sites applied passive retroreflective markers bilaterally per the Plug-in Gait model (Oxford Metrics Group, Oxford, England^[18]). Testing was performed barefoot at each child's self-selected walking speed. Data were processed and analyzed centrally using Plug-in Gait.

The Movement Analysis Profile (MAP) contains individual GVSs, which quantify deviations from normal gait at each joint and plane of motion. Baker *et al.*^[19] identified nine key GVSs: pelvic tilt, pelvic obliquity, pelvic rotation, hip flexion, hip abduction, hip rotation, knee flexion, ankle plantar flexion, and foot progression. GVSs are calculated as the root mean square difference between a specific kinematic variable and the mean from a reference population across the gait cycle. Reference data were established using 90 typically developing children at SC-Portland. Spatiotemporal parameters included walking speed, stride length normalized to height, and cadence. Representative trials (mean of 3, range 2-6) were used to compute GVSs and spatiotemporal parameters. No significant differences were found between the right and left GVSs; right-side data were used for analysis. GVSs were not adjusted for walking speed because, in typically developing children, joint kinematics do not vary once gait matures around age 4^[20]. Longitudinal GVS data have previously been published in relation to age and steroid use^[12], but not in relation to contracture.

Statistical analysis

Exponential growth curve models were tested using SAS PROC NLMIXED Version 9.4 for nonlinear mixed modeling^[21]. Growth curve modeling is a powerful approach for longitudinal data analysis, accommodating complexities such as missing data, uneven time points, and non-normal distributions^[22]. This approach enhances understanding of developmental change and responsiveness to therapeutic interventions^[22,23]. Disease heterogeneity was partly addressed by including random effects for model intercepts. For practical

reasons, random effects were not modeled for slopes. Given the small sample size, multiple predictors, and exponential trajectories, estimating slope variances across individuals was not feasible. Such estimation can cause convergence issues and biased variance estimates with small samples^[24]. Analyses included data collected at both 12- and 6-month intervals, as implemented during different study phases. Exact months of visits were used as the time variable in models to account for unequal and imprecise intervals.

In the base model, trends were examined across the full sample. Age (in months) was centered around the baseline mean, making the intercept interpretable as the predicted value at baseline. Each mixed model included a random intercept (varying by participant), but fixed growth parameters. Estimates of residual and intercept variance were obtained, representing within- and between-person variance, respectively.

RESULTS

The mean age at baseline for the study cohort was 88.8 months (7.4 years), with a standard deviation of 30.1 months (2.5 years). Boys contributed an average of 4.3 longitudinal visits (range: 1 to 11 visits), resulting in a total of 359 visits included in the analysis. The primary results were the documentation of hamstring contractures (popliteal angle data) and ankle plantar flexion contractures. In longitudinal models examining gait spatiotemporal parameters, hip flexion contracture, hip extension and ankle plantar flexion strength, as well as pelvic tilt GVS, were found to influence gait speed. Ankle plantar flexion strength affected stride length, and pelvic obliquity GVS influenced cadence.

Longitudinal contracture development

Figure 1 shows the age at assessment (in years) versus the magnitude of contracture (in degrees). Sample sizes were fewer than 10 at ages 14, 15, and 16 years due to loss of ambulation.

Hip flexion contracture did not exceed 8° [**Figure 1A**]. Adductor tightness was typically less than 6° and was not observed in boys aged 8 to 13 years [**Figure 1B**]. Knee flexion contracture greater than 5° did not occur until age 13 and did not exceed 10° [**Figure 1C**]. Hamstring tightness, as indicated by popliteal angle, was present from age 4 and increased to over 20° by age 13 [**Figure 1D**]. Ankle plantar flexion contractures were present as early as age 4. Loss of dorsiflexion was greater when the knee was flexed (approximately 20°) than when extended (approximately 10°) [**Figure 1E and F**].

Figure 2 shows longitudinal changes in gait spatiotemporal parameters (speed, stride length, and cadence) for this cohort.

Influence of contractures, muscle strength and gait pathology on spatiotemporal gait parameters

In longitudinal models investigating the effect of contractures on gait spatiotemporal parameters [**Table 1**], gait speed decreased at a statistically significant rate, with hip flexion contracture being a significant covariate ($P = 0.0468$). The rate of gait speed decline was slower in subjects with a greater range of hip extension. Both normalized stride length and cadence also decreased significantly over time, but no contracture demonstrated a statistically significant effect on these rates of change.

In models investigating the effect of muscle strength [**Table 2**], gait speed decreased at a significant rate. Both isometric hip extension ($P = 0.0337$) and ankle plantar flexion strength were significant covariates. The rate of gait speed decline was slower in subjects with greater hip extension and ankle plantar flexion strength. Normalized stride length also decreased significantly over time, with ankle plantar flexion strength as a significant covariate. The decline in stride length was slower in subjects with greater ankle plantar flexion strength. Cadence also decreased significantly; however, no muscle group showed a significant effect

Table 1. Significant model results for spatiotemporal gait parameters as a function of contractures

	Estimate	P value	Confidence interval
Gait speed			
Average intercept	0.9415	< 0.0001	0.8390, 1.0440
Average rate	-0.0022	< 0.0001	-0.0031, -0.0012
Hip flexion contracture	0.0039	0.0468	0.0001, 0.0078
Residual variance	0.0202	< 0.0001	0.0164, 0.0239
Internal variance	0.0144	0.0007	0.0062, 0.0225
Normalized stride length			
Average intercept	0.7396	< 0.0001	0.6801, 0.7992
Average rate	-0.0015	< 0.0001	-0.0022, -0.0008
Residual variance	0.0066	< 0.001	0.0053, 0.0079
Internal variance	0.0049	0.0012	0.0020, 0.0078
Cadence			
Average intercept	132.76	< 0.0001	124.09, 141.42
Average rate	-0.0033	< 0.0001	-0.0039, -0.0027
Residual variance	121.64	< 0.0001	99.33, 143.96
Internal variance	161.03	< 0.0001	89.02, 233.04

Table 2. Significant model results for spatiotemporal parameters as a function of muscle strength

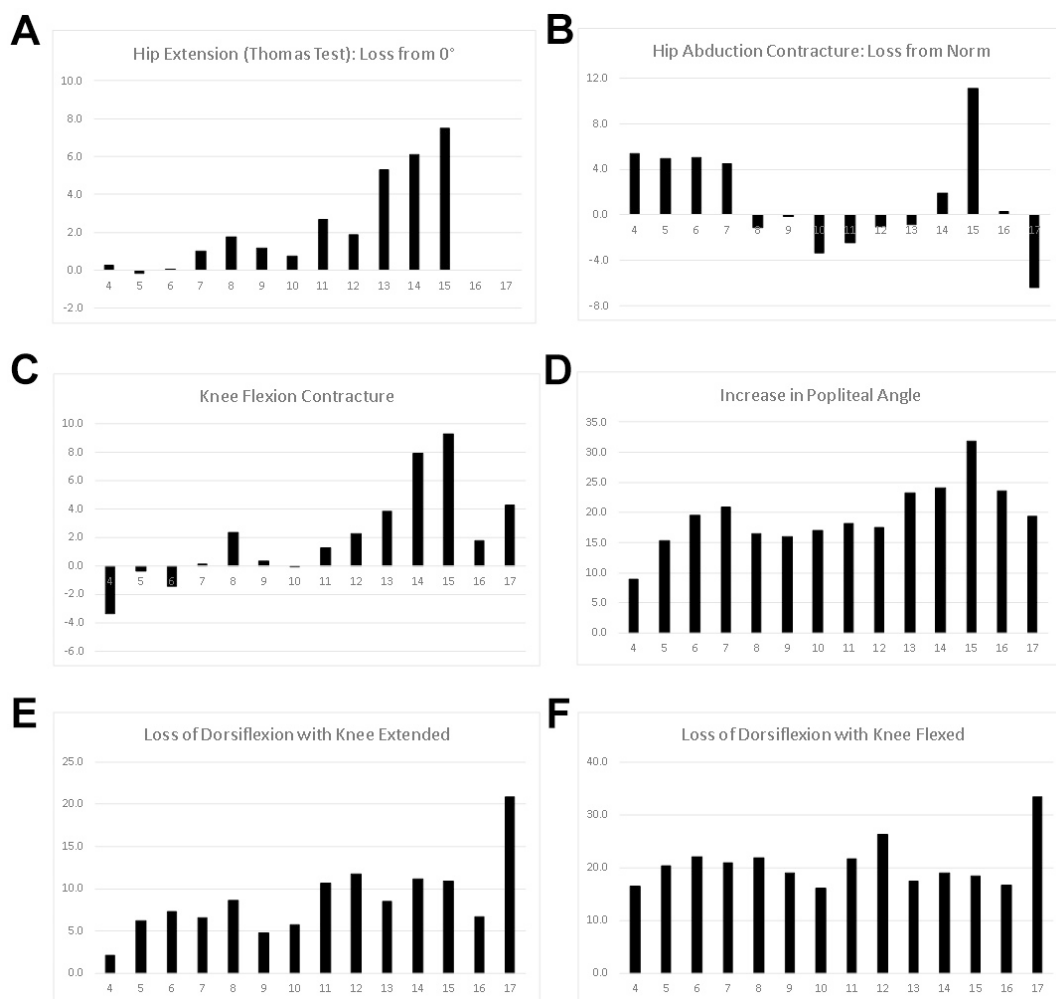
	Estimate	P value	Confidence interval
Gait speed			
Average intercept	0.9413	< 0.0001	0.9144, 0.9682
Average rate	-0.0014	0.0012	-0.0022, -0.0006
Isometric hip extension	0.0821	0.0337	0.0065, 0.1576
Isometric ankle plantar flexion	0.1193	0.0004	0.0563, 0.1823
Residual variance	0.0165	< 0.0001	0.0128, 0.0203
Internal variance	0.0035	0.0381	0.0002, 0.0067
Normalized stride length			
Average intercept	0.0698	< 0.0001	0.06803, 0.0715
Average rate	-0.0016	< 0.0001	-0.0022, -0.0009
Isometric ankle plantar flexion	0.0073	0.0002	0.0037, 0.0109
Residual variance	4.5 E-5	< 0.0001	3.5 E-5, 5.6 E-5
Internal variance	2.1 E-5	0.0049	6.7 E-6, 0.0000
Cadence			
Average intercept	125.78	< 0.0001	122.53, 129.02
Average rate	-0.0026	< 0.0001	-0.0032, -0.0020
Residual variance	97.11	< 0.0001	74.68, 119.55
Internal variance	96.33	0.0003	46.15, 146.51

on the rate of cadence change.

In models evaluating the effect of gait pathology, as measured by GVS [Table 3], gait speed decreased at a significant rate, with pelvic tilt GVS being a significant covariate. The decline in gait speed was slower in subjects with less anterior pelvic tilt. Cadence also decreased significantly, with pelvic obliquity GVS as a significant covariate. The decline in cadence was slower in subjects with less pelvic obliquity. Normalized stride length also showed a significant change over time in this model.

Table 3. Significant model results for spatiotemporal parameters as a function of GVS

	Estimate	P Value	Confidence interval
Gait speed			
Average intercept	0.9849	< 0.0001	0.9346, 1.0351
Average rate	-0.0021	< 0.0001	-0.0029, -0.0013
Pelvic tilt	-0.0113	0.0008	-0.0177, -0.0049
Residual variance	0.0197	< 0.0001	0.0162, 0.0233
Internal variance	0.0111	0.0008	0.0047, 0.0174
Cadence			
Average intercept	139.19	< 0.0001	134.77, 143.60
Average rate	-0.0029	< 0.0001	-0.00343, -0.00243
Pelvic obliquity	-1.6680	0.0096	-2.9180, -0.4180
Residual variance	121.53	< 0.0001	99.91, 143.15
Internal variance	132.32	< 0.0001	73.50, 191.14

**Figure 1.** Mean Hip Flexion Contracture versus Age (A), Mean Hip Adduction Tightness versus Age (B), Mean Knee Flexion Contracture (negative values are hyperextension) versus Age (C), Mean Increase in Popliteal Angle versus Age (D), Mean Plantar Flexion Contracture measured as Dorsiflexion with Knee Extended versus Age (E), Mean Plantar Flexion Contracture measured as Dorsiflexion with Knee Flexed versus Age (F).

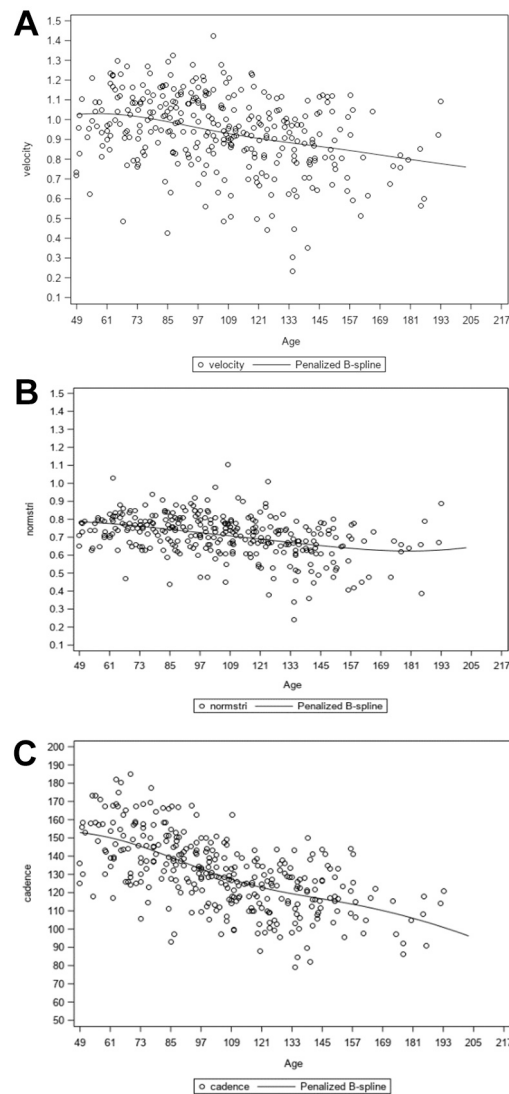


Figure 2. Longitudinal Change in Gait Speed (meters/second) versus Age in Months (A), Longitudinal Change in Stride Length Normalized to Leg Length versus Age in Months (B), Longitudinal Change in Cadence (steps/minute) versus Age in Months (C).

DISCUSSION

The goal of this work was to examine the effect of progressively increasing impairments (e.g., contractures and muscle weakness) due to DMD on gait spatiotemporal parameters. This study is novel for its longitudinal and multifaceted assessments. The primary findings indicate that ankle plantar flexor and hamstring contractures are present as early as 4 years of age. Gait speed, stride length, and cadence declined as ankle and hip impairments worsened, since the hip extensors and ankle plantar flexors are the primary torque generators responsible for propelling gait.

Contractures

Contractures were the earliest to occur and the most pronounced at the ankle, consistent with previous publications^[25,26]. Loss of dorsiflexion was particularly evident with the knee flexed, implicating the soleus muscle. In typically developing children, dorsiflexion is reduced (3° to 8°) with the knee extended compared to when the knee is flexed (18° to 28°) (12). The latter position minimizes the influence of gastrocnemius

tightness. In boys with DMD, both the gastrocnemius and soleus are shortened, leading to a greater discrepancy in ankle dorsiflexion range when the knee is flexed than when extended.

Hamstring tightness, which affects both hip and knee motion, was noted at 5 years of age (15°). Knee flexion contracture did not appear until 13 years of age and was less than 10° . Choi *et al.*^[25] reported that only 5.1% of their cohort had knee flexion contractures of 15° or greater.

Even during adolescence, hip flexor and adductor contractures remained mild ($< 10^\circ$) in this ambulatory cohort. Willcocks *et al.*^[26] found that passive range of motion significantly decreased at the ankle starting five years before loss of ambulation, at the knee starting two years before, and at the hip starting four years before. Sutherland *et al.*^[4] reported lack of hip extension at the end of the stance phase of gait for boys with DMD in the transitional stage of gait impairment. Hip contractures typically worsen after individuals with DMD become non-ambulatory and spend extended periods seated in a wheelchair^[27].

Gait spatiotemporal parameters

The models demonstrated that the greatest rate of walking speed decline was associated with increased hip flexion contracture, increased pelvic tilt GVS, decreased hip extensor strength, and decreased ankle plantar flexor strength. As Perry noted^[28], the hip extensors and ankle plantar flexors are the primary torque producers for forward propulsion during gait. The sagittal plane changes in the hip and pelvis reflect the progressive loss of pelvic girdle muscle strength as DMD advances.

The greatest rate of stride length decline was associated with decreased ankle plantar flexor strength, consistent with their role in gait propulsion. No statistically significant effects of contractures were found, and in the GVS model, normalized stride length did not demonstrate a statistically significant change over time.

The greatest rate of cadence decline was associated with increased pelvic obliquity GVS, without significant effects of contracture or muscle strength. Increased pelvic obliquity results from weak abductors, causing pelvic instability. This instability may lead to cautious forward movement of the swing leg (thus reduced cadence) and increased double support time. In a subgroup of 16 boys from this cohort, double support time increased by an average of 88% from the first to the last gait evaluation.

Gait pathomechanics

In DMD, weakness begins with pelvic muscles and progresses from proximal to distal muscle groups in the legs. In boys aged 6-10 years, gait pathomechanics reflect this pattern of weakness through increased lumbar lordosis, increased anterior pelvic tilt, reduced initial knee flexion at weight acceptance, and increased foot drop during swing^[4,29]. This alignment shifts the center of mass closer to the hip joints and positions the ground reaction force posterior to the knee joint centers, reducing the muscular effort required by the hip and knee extensors^[10,30].

This study provides detailed insight into how pelvic positioning during gait indicates disease progression in DMD. Pelvic tilt GVS was associated with reduced walking speed, while pelvic obliquity GVS was linked to reduced cadence. As reported by Heberer *et al.*^[31], hip joint kinetics may serve as an early and sensitive marker of muscle weakness and disease progression in boys with DMD. Similar to children with low-level spina bifida (L4 and below), increased anterior pelvic tilt results from hip extensor weakness, while increased pelvic obliquity arises from hip abductor weakness^[32,33].

None of the knee parameters were statistically significant covariates in the model, likely due to the limited knee flexion contracture observed in this cohort. The popliteal angle, reflecting hamstring tightness affecting both hip and knee extension, increased to a 20° deficit by 13 years of age. In typically developing boys, the popliteal angle reaches a mean value of 26° by age 5 and beyond^[34]. Although knee strength is impacted by DMD progression^[35], compensatory strategies - such as positioning the ground reaction force anterior to the knee joint - help mitigate the effects of decreased knee strength. Duong *et al.*^[36] found that isometric knee extensor strength correlated with speed in the 10-meter walk/run test but noted that torque, rather than strength alone, is functionally more relevant. They also cautioned that their testing did not include hip or ankle strength assessments.

At the ankle, as plantar flexor strength declined, both gait speed and stride length decreased. An earlier publication reported ankle plantar flexion contracture greater than 30° in 27% of boys aged 9-13 years and in 53% of those older than 13 years^[17]. The stiffness associated with plantar flexor contractures may partially compensate for muscle weakness by restraining forward tibial progression over the foot during stance, thereby contributing to knee stability^[37].

A systematic review of gait deviations in boys with DMD^[38] consistently showed decreased walking speed and stride length relative to typically developing peers. In this cohort, gait speed, stride length, and cadence all declined over time, in contrast to typically developing children, whose gait speed and stride length increase with age primarily due to leg length growth^[39]. Between-subject variability indicates that boys decline in gait function at different rates, while within-subject variability suggests that a single boy may experience periods of more rapid decline. This variability may reflect individual genetic variants, as disease severity has been linked to specific mutations^[40].

The cessation of walking in boys with DMD is multifactorial. Besides contractures, loss of ambulation may result from decreased muscle strength^[11], increased energy cost of walking^[41], progressive gait pathology^[12], or an inability to keep up with peers. Decreased strength and increased pelvic instability may impair a boy's ability to control his center of mass during gait and challenge stability during single-limb stance^[4]. Slower walking may also arise from a fear of falling (5), compounded by an inability to rise from the floor.

Limitations

The sample size for muscle strength analysis was limited, as only 57 of 75 subjects were able to complete these assessments. The use of the Thomas test may have underestimated hip contracture, as the baseline measurement assumes a neutral femur position relative to the pelvis in supine. Mean hip extension measured in prone has been reported as 19° in boys aged 6-10 and 15° in those aged 11-17^[42]. Inter-rater reliability for passive range of motion measures has been reported to be $\pm 6^\circ$ ^[43].

At the start of the study, assessments were conducted annually. For some boys, this one-year interval included their loss of ambulation, limiting their contribution to longitudinal gait data. To increase data collection during the ambulatory period, the protocol was adjusted midway to include assessments every 6 months. Due to variations in age at baseline and the heterogeneity of disease progression in DMD, the number of data points per subject varied from one to eleven. These unequal time points were accounted for in the statistical models.

In nonlinear modeling, it is important to note that the effects of multiple predictors are not additive. In bivariate analyses (one predictor, one outcome), a predictor may be significant, but it may not remain significant in a full model due to interactions among predictors. Suppression effects can also occur; for

example, if two predictors each have a positive relationship with the outcome but are negatively related to each other, their combined effect in the full model may be reduced.

Due to sample size constraints, it was not possible to estimate random effects for model slopes (slope variances) or test their significance. While we expect individual differences in changes over time, fixed effects for slopes are generally stable, as the average rate of change is calculated across individuals. The inability to estimate random effects for model slopes is a limitation of this study and should be addressed in future research.

Conclusion

The decline in walking ability in boys with DMD is multifactorial. Besides contractures, gait deficits can result from decreased muscle strength and increased energy cost of walking. Additionally, slower walking may stem from a fear of falling and the resulting difficulty in rising from the floor.

Muscle weakness drives compensatory strategies that impact kinematic and spatiotemporal gait parameters over time. Decreased strength of plantar flexors, which are primary contributors to forward propulsion, may reduce gait speed. Increased pelvic instability may impair the ability to control the center of mass during gait and challenge single-limb stance stability, leading to reduced cadence and stride length. The significance of longitudinal changes in pelvic GVS in models of gait speed and cadence supports findings from previous studies^[31], suggesting that motion analysis may serve as a sensitive marker of subtle muscle weakness at an early age and may help detect the efficacy of new therapeutics. Pharmacologic treatments to maintain or enhance strength, along with interventions such as nighttime ankle orthotics and daytime straight-knee stretching to limit ankle contracture development, may help prolong ambulation in boys with DMD.

DECLARATIONS

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Authors' contributions

Made substantial contributions to the conception and design of the study: Sussman MD, McDonald, C, Sienko S, Buckon CE, Fowler E, Staudt L, Herberer K, Bagley A

Performed data acquisition: Sienko S, Buckon CE, Fowler E, Staudt L, Herberer K, Bagley A

Performed data analysis: Newsom J, Sienko S, Buckon CE, Bagley A

Performed data interpretation: Sussman MD, McDonald, C, Sienko S, Buckon CE, Fowler E, Staudt L, Herberer K, Bagley A, Newsom J

Availability of data and materials

According to Shriners Children's policy, data for this work cannot be shared.

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Conflicts of interest

All authors declared that there are no conflicts of interest.

Ethical approval and consent to participate

This study was approved by the following institutional review boards: University of California, Davis (Protocol No. 219294-3), Oregon Health & Science University (IRB No. IRB00001988), University of California, Los Angeles (IRB No. 11-000530-CR-00009). Written informed consent was obtained from all participants and/or their parents or legal guardians, in accordance with the requirements of the respective institutional review boards. Ethical approvals were maintained and updated as required throughout the study period.

Consent for publication

Not applicable.

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