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The use of the gait profile score and gait variable score in individuals with Duchenne Muscular Dystropy.

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Title Page

The use of the gait profile score and gait variable score in individuals with Duchenne Muscular Dystropy.

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Abstract

Therapeutic gait interventions for individuals with Duchenne Muscular Dystrophy (DMD) should be based on understanding how movement of the individual is affected and whether different clusters of individuals, determined by clinical severity, differ. Gait indexes have been developed to synthesize the data provided by the three-dimensional (3D) gait analysis such as the Gait Deviation Index (GDI) and the Gait Profile Score (GPS) where the gait variable score (GVS) can be calculated. The objective this study was to evaluate the potential use of the GDI and GPS and MAP using data from 3D gait analysis of DMD patients. The dimension 1 score of the Motor Function Measurement defined the groups that composed the cluster analysis. Twenty patients with DMD composed 2 groups according to the cluster analysis (Cluster 1, n=10; Cluster 2, n=10). Three-dimensional gait analysis was conducted where GDI, GPS and GVS (pelvic tilt/obliquity; hip flexion-extension/ adduction-abduction/ rotation; knee flexion-extension; ankle dorsiflexion-plantarflexion, foot progression angle) were calculated. Cluster 1 group presented lower hip flexion-extension and lower pelvic obliquity when compared with Cluster 2 group ($p<0.05$). There was no difference between groups for GDI, GPS total and maximum isometric muscle strength of the lower limbs ($p>0.05$). This study showed that GVS could detect alterations on the parameters obtained using three-dimensional gait analysis for those DMD patients separated according to motor function regarding pelvic and hip kinematic patterns. The rehabilitation of patients with DMD is recommended from the early stages of the disease (as Cluster 1, with > MFM) with the hip joint being the therapeutic target.

Keywords: Duchenne muscular dystrophy, gait, gait disorders, gait profile score

1. Introduction

Individuals with Duchenne Muscular Dystrophy (DMD) present with gait deterioration and progressive weakness starting in the hip and knee extensor muscles which overloads several structures and tissues of the musculoskeletal system (Townsend et al., 2015). Postural kinematic adaptations such as increased lumbar lordosis and anterior pelvic inclination, knee hyperextension in the terminal support phase of gait, and increased hip and ankle flexion during the swing phase, are necessary for these individuals to maintain their ability to walk even with the increase in the muscle weakness (de Carvalho et al., 2015; Doglio et al., 2011). Thus, understanding the kinematic adaptations of the gait of DMD can improve the knowledge of the disease progression and guide health professionals into the choice of the best therapeutic interventions (Ropars et al., 2016).

Three-dimensional gait analysis is a tool that provides specific quantitative data on gait patterns through the integration of kinematic and kinetic parameters and is a widely used tool in clinical research for the evaluation and follow-up of individuals with DMD (Celletti et al., 2013; Ferreira et al., 2014). In clinical practice, the interpretation of the data obtained by three-dimensional analysis allows the quantification of the functional limitations related with the disease, guiding decisions such as surgical interventions and / or indication of orthoses. Souza et al. (2016), for example, found that an ankle foot orthosis (AFO) improved the kinematic and kinetics parameters of the gait of patients with DMD recommending them for aiding gait in these individuals (De Souza et al., 2016).

The clinical relevance of three-dimensional gait analysis motivated researchers to develop indexes capable of synthesizing the data obtained using this very complex tool, facilitating its comprehension. The most commonly used are the Gillette Gait Index (GGI), the Hip Flexor Index (HFI), the Gait Deviation Index (GDI), the Gait Profile Score (GPS) and the Gait Variable Score (GVS) (Beynon et al., 2010; Rasmussen et al., 2015). Among these indexes, GDI and GPS are global measures of gait variability and are the most sensitive to detect the changes clinically relevant in the gait deviations of children with neurological and orthopedic disorders (Celletti et al., 2013). The GPS, proposed by Baker et al (2009) (Baker et al., 2009), is a compilation of the root mean squares of the nine gait variable scores (GVS) which is capable of further describing the joint specific measures of gait variability. The GPS and GVS (nine variables) can be represented in a movement analysis profile (MAP) (Baker et al., 2009; Ropars et al., 2016). GPS has already been used to assess the differences in kinematic parameters between boys and girls with Down's syndrome (Zago et al., 2019). In patients with Charcot-Marie-Tooth, GPS was able to quantify the degree of impairment of ambulation (Giancarlo Coghe, Massimiliano Pau, Elena Mamusa, Cinzia Pisano, Federica Corona, Giuseppina Pilloni, Micaela Porta, Giovanni Marrosu, Alessandro Vannelli, Jessica Frau, Lorena Lorefice, Giuseppe Fenu, 2018). However, GPS, GVS or MAP has not been used in the evaluation of patients with DMD to determine whether differences exist between subgroups of the disease.

Earlier work has utilized the GDI in individuals with DMD and good correlations with functional alterations were seen when motor function was evaluated by the Gross Motor Function Measure (GMFM) (Thomas et al., 2010). Although the latter authors have obtained positive results between GDI and GMFM, GMFM is not a scale that was developed or used for individuals with DMD.

One of the scales for DMD is the Motor Function Measure (MFM) (Iwabe et al., 2008) and thus one would expect to see gait profile differences between the different clusters within the MFM. Since DMD is a progressive disease, it is well known that muscle strength and abilities, obtained through MFM, decrease day by day (Mcdonald et al., 2013; Pizzato et al., 2014). In this context, we seeked to understand how these variables influence the kinematic parameters of gait and vice-versa. Thus, this study aimed to evaluate the applicability of the GDI and the GPS in patients with DMD, using the MFM score levels as a clustering factor. In our hypotheses, GDI and GPS will be considered a good index to use in the clinical practice if they can answer both questions: a) if we have 2 DMD groups with different motor abilities, GDI and GPS should detect deviations of the gait parameters between them; b) if differences between the groups can be detected, the GVS will be able to show which kinematics of the joints are different.

2. Methods

2.1 Sample

Twenty walking male individuals with DMD, aged between 4 and 12 years, were included in the study. The inclusion criteria were: 1) confirmed DMD diagnosis (by clinical history, genetic testing and muscle biopsy), 2) independent walking and 3) no difficulty understanding the instructions. Exclusion criteria consisted of presence of other disorders and previous surgical procedures. This study was approved by the Ethics Committee of the Hospital das Clínicas da Faculdade de Medicina de Ribeirão Preto da Universidade de São Paulo (process number 6017/2013). All patients were on treatment with corticosteroids. The parents or caregivers signed the informed consent form consenting to participation in the study.

2.2 Evaluation procedures

Maximum isometric muscle strength of hip and knee flexor muscles, and extensors, were tested using a hand-held dynamometer (Lafayette Instrument, Lafayette, US). The dynamometer was held perpendicular and distally to the tested segment and three measurements, in kilogram force, were taken for each tested muscle group and for each limb. The greatest of the three values was used for statistical analysis. Additionally, the passive range of motion (PRoM)(Marques, 2003), 10-meter walking test (10MWT) with shoes, body composition by bioelectrical impedance analyses and functional performance assessed by MFM (Iwabe et al., 2008).

PRoM was evaluated in the sagittal plane by means of a conventional goniometer following the methodology proposed by Marques, 2003 (Marques, 2003) Total body composition was obtained by bioelectrical impedance analysis (BIA), following manufacturer's recommendations (Biodynamics-450, São Paulo, Brazil). Prior to the test each participant was instructed to intake minimal liquid, empty their bladders, avoid alcohol consumption and intense exercise (24 hours prior to the test), caffeine or food 4 hours prior to the test. Two pairs of sensor pads were placed on the participants - one on the right wrist and hand and the other one on the right foot and ankle.

We obtained fat-free mass, fat mass, body mass index (BMI) and phase angle (PA). PA depends on resistance (opposition to the flow of electrical current) and reactance (effect of the capacitive ability of cell membranes to resist the current). PA is used to quantify cell membrane integrity (reactance; Xc) and the redistribution of fluid between intra- and extracellular fluid compartments (resistance; R). PA = tangent arc R/Xc. (Berbigier MC, Pasinato VF, Rubin BA, Moraes RB, 2013). The MFM was applied according to the recommendations contained in the manual; it is composed of three dimensions, totaling 32 items, which are scored from 0 to 3 according to the performance of the patient in the execution of the tasks: Dimension 1 (D1): standing position and transfers, with 13 items; Dimension 2 (D2): axial and proximal motor function, with 12 items; Dimension 3 (D3): distal motor function, with 7 items (Iwabe et al., 2008).

Following this, three-dimensional gait was assessed using an eight-camera motion analysis Qualisys Oqus 300 system sampling at 120Hz. Bony prominences were marked using the Conventional gait model (Helen Hayes marker set) (Kadaba et al., 1990). The reflective markers were placed on antero-superior iliac crest, mid-point between postero-superior iliac crest, medial and lateral epicondyle, medial and lateral malleolus, calcaneal tuberosity and mid-point between second and third metatarsals. Additionally markers on wands over the thigh and leg for were utilized.

2.3 Data processing and Statistical Analysis

Kinematic parameters were computed using Visual3D® (2007) software. The joint's angles were calculated by a coordinate system according to the Cardan sequence (X, Y, Z), being considered two body segments. X represents sagittal plane, Y frontal plane and Z transverse plane. Each individual completed a minimum of eight trials. Mean maximum and minimum gait kinematic parameter peaks were obtained for each individual considering the entire gait cycle, normalized to 100%. Subsequently, the GDI, GPS and GVS for each individual were calculated by GDI-GPS calculator version 3.2 (Baker, n.d.) using the control data provided by Dr Baker in the calculator.

Patients were grouped using cluster analysis, considering dimension 1 (standing and transfers) of the MFM scale as a clustering factor. The clusters were obtained in two steps: in the first step a hierarchical grouping method was used, specifically Ward's algorithm. This method uses a variance analysis approach to evaluate the distances between clusters, that is, it minimizes the sum of the squares of any two clusters that can be formed. In the second step the non-hierarchical cluster was performed using the k-means method with the result of the Ward method as a starting point (Johnson, Richard A.; Wichern, 2008), the objective is optimize the allocation in the clusters. After this procedure, it was possible to divide the data into two distinct groups: group 1 (n = 10) with the highest MFM [D1 = 84.10% (11.06%)], and group 2 (n = 10) D1 = 62.64% (9.58%)].

After the groups were divided, statistical analysis was performed using linear regression models for comparison between the groups. This model assumes that its residuals have normal distribution with mean 0 and constant variance. Comparisons between the groups were obtained by orthogonal contrasts. The results were obtained with the aid of SAS software (Version 9.2), using PROC GLM. In these analyses a level of significance of 5% was considered and the adjustments were obtained in SAS software (Version 9.2).

3 Results

The groups Cluster 1 and Cluster 2 were obtained according to the MFM score, dimension 1. As seen in Table 1, the Cluster 1 group (n=10), with the greater MFM, showed: greater phase angle, smaller execution time in 10-meter walk test and greater total score of MFM ($p<0.05$) than the Cluster 2 group (n=10), with the smallest MFM.

Table 1

The Cluster 1 group showed greater knee extension passive range of motion than the Cluster 2 group ($p<0.05$). For the other values of passive amplitude of joint movement and maximum isometric muscular strength, there were no significant differences between Cluster 1 and Cluster 2 groups ($p> 0.05$) (Table 2).

Table 2

The Table 3 indicates that Cluster 1 group showed reduced hip flexion-extension and reduced pelvic obliquity GVS scores than the Cluster 2 group ($p<0.05$). For the other variables there were no significant differences between groups ($p> 0.05$).

Table 3

4 Discussion

In patients with DMD, the progressive decline of musculoskeletal function is associated with gait deviations (Thomas et al., 2010). Our study showed that GVS gave further information of gait deviations and showed to be a more sensible approach rather than global gait measures as the GDI and GPS cannot distinguish any differences considering motor abilities (Cluster 1 vs Cluster 2). GVS allowed the identification of kinematic differences in the pelvis and in the hip joint between the different clusters which would help to inform rehabilitative targets.

As DMD is a progressive disease, pelvic obliquity increases and it was higher in the Cluster 2 group. The Cluster 2 group was composed with children with lower motor abilities and it is associated with evident gait changes. Even though there was no GPS difference between the groups, GVS was able to detect the change in pelvic obliquity. In Gaudreault et al. (2010), the weakness of the hip extensor muscles caused the greater pelvic obliquity facilitating pelvic progression (Gaudreault et al., 2010) and Doglio et al (2011) observed greater pelvic obliquity in the double support phase in patients with DMD when compared with a control group (Doglio et al., 2011). This alteration can be explained considering that in healthy children, pelvic movement is controlled by the eccentric contraction of the hip abductors which stabilize the hip in a "quasi-static" position while for DMD children, the contralateral pelvis may be lifted by a concentric contraction of the hip abductors (Gaudreault et al., 2010). In a clinical view, pelvic obliquity and altered recruitment of pelvic girdle muscles can justify why patients of the Cluster 2 took a longer time to perform10 MWT than Cluster 1.

The gait deviations on DMD patients can be clustered as low, moderate or advanced, following the study proposed by Thomas et al. (Thomas et al., 2010). Although the authors described kinematic alterations using GDI scores, no difference was observed in 10 MWT [11]. Pizzato et al. (2016) reported that when the rates from the 10 MWT between 2 consecutive sessions are greater than 1.25, it indicates the borderline between independent gait and to become a wheelchair user (Maciel Pizzato et al., 2016). For McDonald et al. (2013) if the 10 MWT is completed in less than 6 seconds it is associated with maintaining walking during the next 12 months (Mcdonald et al., 2013). Contrastingly, if this time is more than 10-12 seconds, it represents a higher probability to loss of walking in 12 months (Mcdonald et al., 2013). In this context, our results demonstrated significant difference in the responses to 10MWT between the groups and, Cluster 1 group presented almost half time of execution when compared with Cluster 2 although both groups presented time of execution lower than 10 seconds. Following this reasoning, it could be expected that the patients of the Cluster 2 group will present a higher probability of losing gait capacity, earlier than the patients of the Cluster 1 group. However, this would need to be confirmed in a future study to determine whether alterations in the GVS seen in the proximal segments, 10MWT responses and loss of ambulation are linked.

Although we did not observe significant difference in muscle strength between the groups, the individuals in Cluster 2 seemed to develop compensatory mechanisms, one of them being an increase in pelvic obliquity. Unlike the pelvis adaptations, the hip joint showed kinematic alteration in individuals with mild gait deviations (Thomas et al., 2010). The increase in hip flexion an obvious kinematic alteration (Doglio et al., 2011), and we could observe greater hip flexion-extension in the Cluster 2 than the Cluster 1 group. Attias et al. (2017) suggested that the increased hip flexion during the gait cycle is associated with the shortening of the gastrocnemius muscle (Attias et al., 2017). This information agreed with our data since a decreased range of the knee extension was observed in the Cluster 2 group and we observed kinematics alterations of the hip, knee and ankle during the gait cycle.

The body composition, mainly phase angle showed differences between Cluster 1 vs Cluster 2. The phase angle has been used as an indicator of cell membrane integrity, so the higher the phase angle, the greater the cell membrane integrity (reactance) and the lower the phase angle, the greater redistribution of fluid between intra- and extracellular (Marino et al., 2017). The review published by Llameset al. (2013) points out that lower phase angle values are associated with higher risk of postoperative complications, greater severity of congestive heart failure, and shorter survival in cancer patients (Llames et al., 2013). Despite its clinical relevance, the phase angle has not been reported yet for patients with DMD. Patients with lower functional score (Cluster 2 group) had lower phase angle than patients with better functional score (Cluster 1 group) and this result can be explained by the greater cellular degeneration and it characterizes a more advanced stage of the disease of Cluster 2 group. The data indicate that phase angle may be useful in assessing the clinical progression and prognosis of DMD, as already reported for other diseases (Llames et al., 2013). The main limitation of this study is the sample size although equal groups were in the clusters. Future work should identify whether clustering by gait pathology would classify the individuals into different groups than the MFM. We also do not have longitudinal follow-up of these individuals so determining whether these changes are predictive is a future direction. We also should describe that the individuals did not wear ankle foot-orthoses during the tests and this may have caused some individuals to walk worse. However, we assessed whether the gait indexes would be able to distinguish between groups and thus this should also be repeated in users of AFOs. There was no control group assed in the paper for the primary reason of distinguishing between DMD individuals. However, future studies should have a control group range of parameters to further understand longitudinal changes in these variables with rehabilitation.

In summary, individuals with DMD with different motor abilities presented with important alterations in body composition, timed performance tests and kinematic alterations of the pelvic girdle during gait. The biomechanical changes observed in the hip joint of patients with DMD may suggest therapeutic strategies as maintain the hip extension function and the flexibility of the hip flexors. The kinematic findings help the therapeutic direction, as the therapeutic interventions can affect the kinetics findings. For example, corticosteroid intervention improved kinematics of the hip joint [28]. These authors suggested that hip joint kinetics, which represents an initial marker of proximal weakness and it is sensitive to interventions, should be studied for its reliability, feasibility and applicability as outcome measures for new therapies in DMD. In this sense, the results of the present study indicate that gait variable score allows the detection of proximal kinematic changes in pelvis and hip even in the patients with more preserved motor function, assessed by MFM and would be a useful measure to consider for longitudinal assessments of individuals with DMD

6. Conclusion

This study showed that global measures of gait such as the GDI and GPS did not detect alterations on the parameters obtained using three-dimensional gait analysis for those individuals with DMD separated according to motor function. However, the gait variable score highlighted where those changes were and would be a useful tool to be used for longitudinal assessments in these individuals. The impairments identified the hip joint as a target for rehabilitation in individuals with DMD in the early stages of the disease.

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Declarations of interest

None declared.

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Table 1

Table 1: Sample characterization, according Cluster 1 group and Cluster 2 group.

	Cluster 1 (>MFM; n=10)	**Cluster 2 (<MFM; n=10)**
Age (years)	6.7 (1.9)	8.4 (2.5)
Mass (kg)	24.6 (7.3)	33.2 (16.0)
Height (m)	1.2 (1.1)	1.3 (1.1)
BMI (kg/m2)	17.8 (2.0)	19.8 (5.7)
Fat free mass (kg)	20.8 (3.5)	23.3 (7.7)
Fat mass (kg)	6.2 (2.4)	11.4 (8.7)
Phase Angle (º)	3.7 (0.6)*	3.0 (0.7)
10-MWT (s)	5.1 (1.2)*	9.3 (3.6)
D1- MFM (%)	87.2 (5.6)*	57.3 (16.2)
MFM total score (%)	94.2* (2.7)	80.6 (7.9)

*p<0.05 when compared to Cluster 2. BMI: body mass index. 10-MWT: 10 meter walk test. D1-MFM: dimension 1 of motor function measure scale.

Table 2: Passive range of motion (PRoM) and maximum isometric muscle strength, to Cluster 1 group and Cluster 2 group.

	Cluster 1 (>MFM; n=10)	Cluster 2 (<MFM; n=10)
PRoM (º)		
Hip extension	10.8 (4.2)	9.5 (3.4)
Hip flexion	125.4 (10.9)	121.0 (12.8)
Knee extension	1.0 (1.7)*	-2.2 (4.1)
Knee flexion	136.6 (8.4)	131.8 (11.7)
Dorsiflexion (KF)	5.3 (8.3)	1.2 (9.6)
Dorsiflexion (KE)	2.1 (10.1)	0.6 (12.5)
Plantar flexion	64.8 (13.9)	54.8 (14.7)
Maximum isometric muscle strength (Kgf)		
Hip flexors	6.5 (3.3)	5.0 (1.8)
Knee flexors	8.5 (4.3)	7.5 (3.3)
Knee Extensors	8.4 (6.7)	5.4 (2.4)

*$p<0.05$ when compared to Cluster 2. KF – Knee in flexion. KE – knee in extension. MFM: Motor function measure scale. n= number of patients. Kgf: kilogram force. º: degrees.

Table 3

Table 3: Gait profile index (GDI) and Gait profile score (GPS)/ Gait variable scores (GVS) to groups Cluster 1 and Cluster 2.

	Cluster 1 (>MFM; n=10)	**Cluster 2 (<MFM; n=10)**
GDI	81.5 (5.0)	76.1 (8.8)
GPS total (º)	8.8 (1.4)	10.7 (2.9)
Pelvic tilt (º)	6.2 (3.4)	10.4 (6.9)
Pelvic obliquity (º)	2.2 (1.3)*	3.7 (1.7)
Pelvic rotation (º)	4.4 (1.7)	5.0 (3.8)
Hip Flexion-extension (º)	6.7 (1.4) *	12.2 (5.4)
Hip Adduction-Abduction (º)	3.2 (1.1)	4.7 (3.2)
Hip rotation (º)	13.5 (5.9)	11.9 (10.8)
Knee Flexion-extension (º)	12.3 (1.6)	11.3 (4.0)
Ankle Dorsiflexion-plantarflexion (º)	14.2 (4.7)	11.8 (11.2)
Foot progression angle (º)	11.8 (6.0)	8.5 (5.3)

* $p<0.05$ when compared with Cluster 2 group. GDI, GPS and GVS for each individual were calculated by GDI-GPS calculator version 3.2 [20].

Declarations of interest: none