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| Abstract: | <p>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.</p> |

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

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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 **1. Introduction**

2 Individuals with Duchenne Muscular Dystrophy (DMD) present with gait deterioration
3 and progressive weakness starting in the hip and knee extensor muscles which overloads several
4 structures and tissues of the musculoskeletal system (Townsend et al., 2015). Postural kinematic
5 adaptations such as increased lumbar lordosis and anterior pelvic inclination, knee hyperextension
6 in the terminal support phase of gait, and increased hip and ankle flexion during the swing phase,
7 are necessary for these individuals to maintain their ability to walk even with the increase in the
8 muscle weakness (de Carvalho et al., 2015; Doglio et al., 2011). Thus, understanding the
9 kinematic adaptations of the gait of DMD can improve the knowledge of the disease progression
10 and guide health professionals into the choice of the best therapeutic interventions (Ropars et al.,
11 2016).

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

21 The clinical relevance of three-dimensional gait analysis motivated researchers to develop
22 indexes capable of synthesizing the data obtained using this very complex tool, facilitating its
23 comprehension. The most commonly used are the Gillette Gait Index (GGI), the Hip Flexor Index
24 (HFI), the Gait Deviation Index (GDI), the Gait Profile Score (GPS) and the Gait Variable Score
25 (GVS) (Beynon et al., 2010; Rasmussen et al., 2015). Among these indexes, GDI and GPS are
26 global measures of gait variability and are the most sensitive to detect the changes clinically
27 relevant in the gait deviations of children with neurological and orthopedic disorders (Celletti et

28 al., 2013). The GPS, proposed by Baker et al (2009) (Baker et al., 2009), is a compilation of the
29 root mean squares of the nine gait variable scores (GVS) which is capable of further describing
30 the joint specific measures of gait variability. The GPS and GVS (nine variables) can be
31 represented in a movement analysis profile (MAP) (Baker et al., 2009; Ropars et al., 2016). GPS
32 has already been used to assess the differences in kinematic parameters between boys and girls
33 with Down's syndrome (Zago et al., 2019). In patients with Charcot-Marie-Tooth, GPS was able
34 to quantify the degree of impairment of ambulation (Giancarlo Coghe, Massimiliano Pau, Elena
35 Mamusa, Cinzia Pisano, Federica Corona, Giuseppina Pilloni, Micaela Porta, Giovanni Marrosu,
36 Alessandro Vannelli, Jessica Frau, Lorena Lorefice, Giuseppe Fenu, 2018). However, GPS, GVS
37 or MAP has not been used in the evaluation of patients with DMD to determine whether
38 differences exist between subgroups of the disease.

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

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

55 **2. Methods**

56

57 2.1 Sample

58 Twenty walking male individuals with DMD, aged between 4 and 12 years, were included in
59 the study. The inclusion criteria were: 1) confirmed DMD diagnosis (by clinical history, genetic
60 testing and muscle biopsy), 2) independent walking and 3) no difficulty understanding the
61 instructions. Exclusion criteria consisted of presence of other disorders and previous surgical
62 procedures. This study was approved by the Ethics Committee of the Hospital das Clínicas da
63 Faculdade de Medicina de Ribeirão Preto da Universidade de São Paulo (process number
64 6017/2013). All patients were on treatment with corticosteroids. The parents or caregivers signed
65 the informed consent form consenting to participation in the study.

66 2.2 Evaluation procedures

67 Maximum isometric muscle strength of hip and knee flexor muscles, and extensors, were
68 tested using a hand-held dynamometer (Lafayette Instrument, Lafayette, US). The dynamometer
69 was held perpendicular and distally to the tested segment and three measurements, in kilogram
70 force, were taken for each tested muscle group and for each limb. The greatest of the three values
71 was used for statistical analysis. Additionally, the passive range of motion (PRoM)(Marques,
72 2003), 10-meter walking test (10MWT) with shoes, body composition by bioelectrical impedance
73 analyses and functional performance assessed by MFM (Iwabe et al., 2008).

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

81 We obtained fat-free mass, fat mass, body mass index (BMI) and phase angle (PA). PA depends
82 on resistance (opposition to the flow of electrical current) and reactance (effect of the capacitive
83 ability of cell membranes to resist the current). PA is used to quantify cell membrane integrity
84 (reactance; X_c) and the redistribution of fluid between intra- and extracellular fluid compartments
85 (resistance; R). $PA = \text{tangent arc } R/X_c$. (Berbigier MC, Pasinato VF, Rubin BA, Moraes RB,
86 2013). The MFM was applied according to the recommendations contained in the manual; it is
87 composed of three dimensions, totaling 32 items, which are scored from 0 to 3 according to the
88 performance of the patient in the execution of the tasks: Dimension 1 (D1): standing position and
89 transfers, with 13 items; Dimension 2 (D2): axial and proximal motor function, with 12 items;
90 Dimension 3 (D3): distal motor function, with 7 items (Iwabe et al., 2008).

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

98

99 2.3 Data processing and Statistical Analysis

100

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

109 Patients were grouped using cluster analysis, considering dimension 1 (standing and transfers)
110 of the MFM scale as a clustering factor. The clusters were obtained in two steps: in the first step
111 a hierarchical grouping method was used, specifically Ward's algorithm. This method uses a
112 variance analysis approach to evaluate the distances between clusters, that is, it minimizes the
113 sum of the squares of any two clusters that can be formed. In the second step the non-hierarchical
114 cluster was performed using the k-means method with the result of the Ward method as a starting
115 point (Johnson, Richard A.; Wichern, 2008), the objective is optimize the allocation in the
116 clusters. After this procedure, it was possible to divide the data into two distinct groups: group 1
117 (n = 10) with the highest MFM [D1 = 84.10% (11.06%)], and group 2 (n = 10) D1 = 62.64%
118 (9.58%).

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

125

126 **3 Results**

127

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

132

133 **Table 1**

134

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

139

140 **Table 2**

141

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

145

146 **Table 3**

147

148 **4 Discussion**

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

155 As DMD is a progressive disease, pelvic obliquity increases and it was higher in the Cluster
156 2 group. The Cluster 2 group was composed with children with lower motor abilities and it is
157 associated with evident gait changes. Even though there was no GPS difference between the
158 groups, GVS was able to detect the change in pelvic obliquity. In Gaudreault et al. (2010), the
159 weakness of the hip extensor muscles caused the greater pelvic obliquity facilitating pelvic
160 progression (Gaudreault et al., 2010) and Doglio et al (2011) observed greater pelvic obliquity in
161 the double support phase in patients with DMD when compared with a control group (Doglio et

162 al., 2011). This alteration can be explained considering that in healthy children, pelvic movement
163 is controlled by the eccentric contraction of the hip abductors which stabilize the hip in a “quasi-
164 static” position while for DMD children, the contralateral pelvis may be lifted by a concentric
165 contraction of the hip abductors (Gaudreault et al., 2010). In a clinical view, pelvic obliquity and
166 altered recruitment of pelvic girdle muscles can justify why patients of the Cluster 2 took a longer
167 time to perform 10 MWT than Cluster 1.

168 The gait deviations on DMD patients can be clustered as low, moderate or advanced,
169 following the study proposed by Thomas et al. (Thomas et al., 2010). Although the authors
170 described kinematic alterations using GDI scores, no difference was observed in 10 MWT [11].
171 Pizzato et al. (2016) reported that when the rates from the 10 MWT between 2 consecutive
172 sessions are greater than 1.25, it indicates the borderline between independent gait and to become
173 a wheelchair user (Maciel Pizzato et al., 2016). For McDonald et al. (2013) if the 10 MWT is
174 completed in less than 6 seconds it is associated with maintaining walking during the next 12
175 months (McDonald et al., 2013). Contrastingly, if this time is more than 10-12 seconds, it
176 represents a higher probability to loss of walking in 12 months (McDonald et al., 2013). [In this
177 context, our results demonstrated significant difference in the responses to 10MWT between the
178 groups and, Cluster 1 group presented almost half time of execution when compared with Cluster
179 2 although both groups presented time of execution lower than 10 seconds. Following this
180 reasoning, it could be expected that the patients of the Cluster 2 group will present a higher
181 probability of losing gait capacity, earlier than the patients of the Cluster 1 group. However, this
182 would need to be confirmed in a future study to determine whether alterations in the GVS seen in
183 the proximal segments, 10MWT responses and loss of ambulation are linked.](#)

184 Although we did not observe significant difference in muscle strength between the groups,
185 the individuals in Cluster 2 seemed to develop compensatory mechanisms, one of them being an
186 increase in pelvic obliquity. Unlike the pelvis adaptations, the hip joint showed kinematic
187 alteration in individuals with mild gait deviations (Thomas et al., 2010). The increase in hip
188 flexion an obvious kinematic alteration (Doglio et al., 2011), and we could observe greater hip
189 flexion-extension in the Cluster 2 than the Cluster 1 group. Attias et al. (2017) suggested that the

190 increased hip flexion during the gait cycle is associated with the shortening of the gastrocnemius
191 muscle (Attias et al., 2017). This information agreed with our data since a decreased range of the
192 knee extension was observed in the Cluster 2 group and we observed kinematics alterations of the
193 hip, knee and ankle during the gait cycle.

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

216 parameters to further understand longitudinal changes in these variables with
217 rehabilitation.

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

231

232

233 **6. Conclusion**

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

240

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252

253 **Declarations of interest**

254 None declared.

255

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344

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 (°) | 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 (°) | | |
| 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. °: degrees.

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

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