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**ORIGINAL ARTICLE**

**The impact of postural and anthropometric properties of foot and ankle on physical performance and ambulation of patients with Duchenne muscular dystrophy**

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## Abstract

**Background:** Abnormal foot anthropometry and posture of patients with Duchenne Muscular Dystrophy (DMD) can be considered as possible risk factors for performance and ambulation. It was aimed to examine the effects of foot posture and anthropometric characteristics, which deteriorated from the early period, on ambulation and performance of patients with DMD.

**Methods:** The foot arch height (FAH), the metatarsal width (MW), subtalar pronation angle, and ankle limitation degree (ALD) were evaluated to determine the foot anthropometric characteristics of the patients. Foot Posture Index-6 (FPI-6) was used to evaluate foot posture. The performance of the patients was determined by the 6-minute walk test (6MWT), the 10-meter walk test (10MWT), and ascend/descend a standard 4-step test, and the ambulation was determined by the North Star Ambulatory Assessment (NSAA). Spearman's correlation coefficient was calculated to assess the relationship between foot anthropometric characteristics and posture, and performance and ambulation.

**Results:** The sample consisted of 48 patients with DMD aged 5.5 to 12 years. Both of the right and left foot FPI-6 scores were associated with all parameters, except descending 4-step. The left FAH was associated with 6MWT and NSAA, and the left MW was associated with 6MWT. The ALD of right foot was associated with 6MWT, ascending/descending 4 steps, and NSAA, and left ankle limitation was associated with NSAA ( $p < 0.05$ ). There was no relationship between other parameters.

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**Conclusions:** These findings suggest that postural disorders in the foot and ankle may contribute to the decrease in performance and ambulation in patients with DMD.

Clinical Trial Number: NCT05436210

**Keywords:** Duchenne Muscular Dystrophy, foot posture, foot deformities, performance, ambulation

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## 1. Introduction

Duchenne Muscular Dystrophy (DMD) which is caused by mutations in the dystrophin gene is a progressive and degenerative neuromuscular disease of childhood <sup>1</sup>. Deficiency of protein called “dystrophin” which originates from mutations in the dystrophin gene causes muscle weakness, which is prominent in the proximal muscles of the limbs, especially lower extremities, and then progresses distally. The secondary musculoskeletal complications caused by muscular weakness such as joint limitations, malalignment of the postural segments, and muscle shortness develop within the nature of the disease progression and cause a wide range of functional loss in patients <sup>1,2</sup>.

The ankle plantar flexion contractures are among the first musculoskeletal complications usually seen in patients with DMD. Contractures develop in the hamstrings, hip flexors, and tensor fascia latae muscles right after the impairment of the gastro-soleus complex <sup>2,3</sup>. Disturbances of foot structure and foot posture are typical and progress with the course of the disease, and affected boys predictably develop hindfoot varus deformity and progressive equinus <sup>4</sup>. It has been stated that the flat-foot position in patients with DMD may be caused by contracture of the triceps surae or weakness of the dorsiflexor muscles <sup>5</sup>. These problems of the feet also affect the gait characteristics and balance of the patients, and the patients begin to walk on their toes by increasing their support surface and lumbar lordosis <sup>6</sup>. Therefore, foot disorders are important and the patients are recommended stretching exercises and night

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splints to maintain foot posture and prevent limitations<sup>7</sup>. Besides, foot structure is among the factors affecting daily life of the patients with DMD due to pain, difficulty in fitting into shoes and cosmetic concerns after losing the ambulation<sup>4,8</sup>.

To the best of our knowledge, there are limited studies examining the effect of foot posture on patients with DMD. Considering the prominent foot anthropometric characteristics and foot posture disorders in DMD patients, abnormal foot anthropometry and posture can be considered as possible risk factors for performance and ambulation. Thus, the aim of current study was to investigate the effects of anthropometric characteristics and posture of foot on ambulatory status and performance of children with DMD.

## **2. Materials and Methods**

### **2.1. Participants**

The current study was conducted in line with the principles of the Helsinki Declaration and was approved by the ethics committee of the Hacettepe University (ethics code No: GO 22/414). All participants and their parents provided written informed consent forms. The study protocol was registered in the Clinical Trials Registry No. NCT05436210.

Forty-eight patients with DMD volunteered to participate in this study from June 2022 to November 2022. The main inclusion criteria of the subjects were listed as follows; having a diagnosis of DMD with genetic confirmation; being able to walk without assistance; having no difficulty in understanding and following the instructions of the evaluator. Patients under 5

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years old who had a trauma or surgery history within the last 6 months, and who had concomitant disease affecting their motor functions were excluded.

## **2.2. Assessments**

Age (year), weight (kg), height (cm), body mass index (BMI), functional level of the lower limbs according to Vignos Lower Extremity Rating Scale (VLERS), information about the use of steroids and orthotics were recorded. VLERS is a 10-level lower extremity functional rating scale which grade 1 indicates that the child can walk and climb stairs without assistance and grade 10 indicates that the child is bedridden<sup>9</sup>.

### **2.2.1. Foot Anthropometry**

*The foot arch height (FAH)* was determined by the *navicular drop test (NDT)*. For the measurement, after the patients were seated on a chair and the feet were placed on the floor, the distance between the navicular tuberosity and the floor was measured. The measurement was then repeated while the patients were in a relaxed standing position. The difference between the two measurements was recorded and a difference over 10 mm was considered as flat feet<sup>10</sup>.

*The metatarsal width (MW)*, the transverse distance between the 1<sup>st</sup> and 5<sup>th</sup> metatarsal heads, was measured with a calliper. Weighted (standing) and unweighted (sitting) measurements were performed, and a difference more than 6mm between the measures

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meant that forefoot width has increased which demonstrates high pressure on the metatarsal heads <sup>11,12</sup>.

*The subtalar pronation angle (SPA)* was determined by measuring the angle between the long line of the Achilles tendon and the midpoint of the calcaneus with a goniometer while standing <sup>13,14</sup>. Approximately 3° to 8° pronation is considered as normal whereas greater degrees indicate increased pronation of foot which cause greater load on the subtalar joint <sup>15</sup>.

### **2.2.2. Foot Posture**

*Foot Posture Index-6 (FPI-6)* was used to evaluate the foot posture of the patients. FPI-6 is a valid and reliable assessment tool for pediatrics <sup>16</sup>. The items are scored separately for the right and left foot while the patient is in an upright position with a relaxed posture where their body weight evenly distributed on both feet. Each item is scored as -2, -1, 0, 1, 2 and the total score is categorized as follows: 0 to +5 normal; +6 to +9 pronated; 10+ highly pronated; -1 to -4 supinated; and -5 to -12 highly supinated <sup>17,18</sup>.

The ankle range of motion of the patients was also evaluated by a goniometer and the ankle limitation degree (ALD) of both feet was also recorded. Goniometric measurements with high intra- and interobserver subjectivity have been reported in the literature <sup>19</sup>.

### **2.2.3. Performance**

*The 6-minute walk test (6MWT)* which was accepted as the primary outcome measure in clinical trials for patients with DMD <sup>20</sup> was performed according to the instructions of the



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American Thoracic Society <sup>21</sup>, by placing a cone at the start and end points, and asking the patient to walk as fast as he could for 6 minutes. Patients paused and rested if necessary, and continued. Also, for safety, the physiotherapist walked behind the patient and gave positive verbal support <sup>20</sup>. The distance taken during the 6 minute was recorded as meters. The other timed performance tests such as *10 meters walk test (10MWT)* and *ascending-descending a standard 4-step* were also performed. For 10 meters walk test, patients were asked to walk 10 meters as fast as possible without shoes and for *ascending-descending a standard 4-step*, patients were asked to ascend and descend a standardized 4-step as fast as they could by holding or not holding the handrails. The duration of the performances was recorded in seconds <sup>22</sup>.

#### **2.2.4. Ambulation**

The ambulatory status of patients was evaluated by using the *North Star Ambulatory Assessment (NSAA)*, a DMD-specific ambulation scale. NSAA is a 17-item scale that evaluates ambulatory activities such as walking, climbing stairs, hopping, getting up from a chair, and running. The scale is scored between 0 and 34 which the higher scores indicate better ambulatory status of the children. <sup>23</sup>.

#### **2.3. Statistical Analysis**

The IBM SPSS Statistics software version 26.0 was used to analyse the data. Descriptive characteristics were defined by mean and standard deviation ( $X \pm SD$ ) for quantitative data and

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number/percentage (n/%) for qualitative data. Spearman's correlation coefficient was calculated to assess the relation between foot anthropometric characteristics and posture, and performance and ambulation. The strength of the correlations were classified as follows; (r)=0.05-0.30 weak correlation, 0.30-0.40 weak-to-moderate correlation, 0.40-0.60 moderate correlation, 0.60-0.70 good correlation, 0.70-0.75 strong correlation, and 0.75-1.00 very strong correlation<sup>24</sup>. Statistically significance level (p) was accepted as <0.05.

### 3. Results

The sample consisted of 48 patients with DMD aged 5.5 to 12 years (mean age  $\pm$  SD, 8.04 $\pm$ 1.8 years). It was observed that most of the patients had decreased arch height (flat feet) in both feet (Right Foot: 60.4% Left Foot: 73.5%). The characteristics of the study population were shown in Table 1.

The results related to the measurements of foot characteristics and posture, and the performance and ambulatory tests were given in Table 2.

Table 3 shows the associations between the foot posture and anthropometric characteristics and performance and ambulation tests scores. Posture index of both of the right and left foot were moderately associated with all parameters, except for the duration in descending 4-step. Among the anthropometric characteristics, left FAH was weak-to-moderately associated with 6MWT and NSAA, and MW-L had weak correlation with 6MWT. The weak-to-moderate relations were determined between ALD of right foot and 6MWT,

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ascending/descending 4 steps, and NSAA. The ALD of left ankle had weak-to-moderate correlation with NSAA.

#### **4. Discussion**

Prolonged ambulation is the main focus of the medical and other therapeutic approaches for patients with DMD <sup>25</sup>. Thus, it is essential for patients with DMD to elucidate the factors which may affect physical ability to maintain ambulation. This study revealed that impairment of foot posture and foot characteristics as metatarsal width and arch height negatively affected performance and ambulation in patients with DMD.

The feet of the patients in the present study were in pronation according to the FPI-6. The more impaired foot parts were talar head palpation, talo-navicular congruence, medial arch height and calcaneal eversion according to the FPI-6 which indicated impaired foot posture in sagittal, transverse, and longitudinal planes of DMD population in this study. In a previous study, foot posture was shown to affect gait characteristics and balance negatively in patients with DMD <sup>6</sup>. In another study, it was reported that as the foot dorsiflexion range of motion decreased in DMD, worse results obtained in NSAA, 6MWT and 10MWT <sup>26</sup>. Besides foot posture, ankle limitation was also determined to be related to the performance and ambulation in current study. In terms of muscular weakness, although the proximal involvement was more prominent than the distal in DMD <sup>1</sup>, the present study showed that, besides proximal weakness, distal body involvement including joint limitations and muscle shortness also contributes to the

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performance and ambulation in line with the previous literature. The findings also revealed the importance flexibility and posture of foot and ankle in order to achieve functional activities of daily life in patients with DMD.

It has been known that patients with DMD may present flat foot in their physical examination<sup>2,5</sup>. Similarly, it was observed that most of the patients included in current study had flat feet (FAH>10mm). In addition, the current findings showed that as the medial longitudinal arch height decreased, that is, as the flatfoot severity increased, the physical performance and ambulation status of the patients with DMD deteriorated. Chiang et al. showed that repetitive and demanding motor activities such as 6-minute fast walking required more extrinsic muscle activation as a compensation in adult individuals with flatfoot compared to non-flatfoot,<sup>27</sup>. Similarly, another study revealed that individuals with flatfoot showed greater muscle activation during postural performance such as standing on one leg, and their center of pressure displacements were larger<sup>28</sup>. In a study by Nakhostin-Roohi et al., in which they compared the physical fitness factors including static and dynamic balance, agility and speed of children with flatfoot and non-flatfooted children aged 14-17 years, they showed that individuals with flatfoot had worse static balance and agility scores, and had similar speed and dynamic balance compared to non-flatfoot children<sup>29</sup>. In contrast, Tudor et al. stated that flatfoot does not affect performance in healthy children aged 11-15 years<sup>29</sup>. In addition to these contradictory results in the literature, in this study, considering the negative correlation

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between flatfoot and demanding and repetitive performance task (6 min walk test) and an ambulation assessment (NSAA) that includes many functional motor activities such as standing up from the ground, jumping, standing on one leg, and standing up from a chair, the necessity of emphasizing the importance of foot structure in patients with DMD has been revealed. Thus, it was concluded that strengthening of foot-ankle muscles, especially the tibialis posterior muscle, which is considered to play an important role in stabilising MLA <sup>30,31</sup>, should be included in the exercise programs of patients with DMD.

In this study, SPA of the feet of the patients was higher than normal <sup>15</sup>, which indicated pronated feet. However, the pronation of the feet did not affect ambulation or performance. Toe-standing position and toe-walking are common in patients with DMD because of the forward shift of the gravity center <sup>32,33</sup>. Therefore, the load on the forefoot is higher during dynamic activities such as walking in patients with DMD. This may be due to the fact that SPA, which is a rearfoot deformity, was not associated with the functional tests involving dynamic activities such as 6MWT, NSAA, and 10MWT. Said et al. showed correlation between functional reach distance (static activity) and the rearfoot area of the pronated foot in the older persons <sup>34</sup>. Hargrave et al. showed that subjects with pronated feet did not produce different peak vertical forces or rate of loading when compared to subjects with neutral feet during a single-leg landing (dynamic activity) <sup>35</sup>. These results of the studies mentioned can support our thoughts that there was no relationship between SPA and dynamic activities, since patients

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with DMD cannot transfer weight on the hind foot due to toe-standing position and toe-walking. However, since excessive pronation is thought to play a very important role in lower limb injuries and shock absorption<sup>36</sup>, although it is not associated with ambulation and performance, the subtalar pronation angle should be protected with orthoses and exercises in patients with DMD.

Metatarsal width of the foot was also within normal limits in most of the subjects in the study population and it was not associated with the ambulation and performance parameters except for the left foot. The higher number of patients with increased left foot metatarsal width in comparison to the right foot might be the rationale for the relation between left foot metatarsal width and the 6MWT distance in this study. This finding suggested that the increase in metatarsal width might cause excessive load on the metatarsal heads and adversely affect performance.

There are some limitations in this study. One of them was that the anthropometric characteristics of foot could not be evaluated with an objective assessment method. The other was that the underlying factors for foot impairments, such as the architectural features of foot muscles in patients with DMD, were still unclear, thus, this caused limited statements on the association with foot disorders. Another limitation was that, to the best of our knowledge, the limited number of studies including values such as FAH, MW and SPA in patients with DMD limited us to discuss with similar studies.

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As a conclusion, changes in posture and anthropometric properties of foot occur because of muscle weakness, joint contractures, and postural adaptations, and these changes may contribute to the decrease in performance and ambulation in patients with DMD. This study revealed that patients with DMD have pronated and flat feet and that these disorders are associated with performance and ambulation parameters of patients with DMD. In addition, ankle limitation and metatarsal width were also shown to affect performance and ambulation. Since it has been known that foot structure is important for the spinal alignment of patients with DMD, even after the loss of ambulation<sup>37</sup>, the timely detection of any possible foot impairment with regular evaluation of foot may contribute timely interventions such as positioning, strengthening and flexibility exercises in rehabilitation programs to prolong ambulation and preserve functional performance in DMD.

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**Table 1.** The characteristics of the study population

	Mean (SD)	Range (Min -Max)
Age (year)	8.04 (1.8)	5.5 – 12
Height (cm)	125.91 (9.71)	109 – 158
Weight (kg)	28.55 (7.71)	18 – 49
Body Mass Index (kg/m <sup>2</sup> )	17.76 (3.05)	11.97 – 28.34
	<b>N (R – L)</b>	<b>% (R – L)</b>
<b>Foot Arch Height</b>		
> 10mm	29 – 25	60.4 – 73.5
< 10mm	19 – 9	39.6 – 26.5
<b>Metatarsal Width</b>		
> 6 mm	11 - 15	22.9 – 31.3
< 6 mm	37 - 33	77.1 – 68.8
	<b>N</b>	<b>%</b>
<b>VLERS</b>		
Level I	27	56.3
Level II	17	35.4
Level III	4	8.3
<b>Steroid usage</b>		
Yes	42	87.5
No	6	12.5
<b>Orthotic Device</b>		
Yes	38	79.2
No	10	20.8

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VLERS : Vignos Lower Extremity Rating Scale, SD: Standart Deviation, N: Participation number)

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**Table 2.** Descriptive analysis for each of the variables measured.

Foot Posture - Anthropometric Tests	Mean (SD)	Range (Min-Max)
FAH -R (cm)	1.01 (0.38)	0.3 – 1.9
FAH -L (cm)	1.14 (0.41)	0.4 – 2.2
MW -R (cm)	0.41 (0.36)	0.1– 1.9
MW -L (cm)	0.44 (0.37)	0.1 – 1.5
SPA-R (°)	11.72 (4.75)	2 – 23
SPA-L (°)	11.43 (4.67)	2 – 21
FPI-R	5.35 (2.16)	1 – 10
FPI-L	5.25 (2.04)	1 – 9
ALD (R)	15.68 (7.78)	0 – 30
ALD (L)	14.00 (7.91)	0 – 30
Performance Tests		
6MWT (m)	392.87 (85.89)	170 – 630
10MWT (sec)	8.82 (2.41)	2.67 – 8.85
Ascend 4-step (sec)	5.53 (4.72)	1.27 – 26.15
Descend 4-step (sec)	4.13 (2.84)	1.50 – 19.10
Ambulation Test		
NSAA	24.17 (6.64)	10 – 34

(FAH: Foot Arch Height; MW: Metatarsal Weight; SPA: subtalar pronation angle; FPI: Foot Posture Index; R: Right; L: Left; 6MWT: 6-Meter Walk Test; 10MWT: 10-Meter Walk Test; NSAA: Nort Star Ambulatory Assessment)

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**Table 3.** Associations Between Foot Posture and Anthropometric Characteristics and Performance and Ambulation Tests (Spearman’s *r*)

	<b>6MWT</b>	<b>10MWT</b>	<b>Ascending 4-step</b>	<b>Descending 4- step</b>	<b>NSAA</b>
<b>FPI-R</b>	<b>-0.55**</b>	<b>0.45**</b>	<b>0.47**</b>	0.26	<b>-0.47**</b>
<b>FPI-L</b>	<b>-0.47**</b>	<b>0.45**</b>	<b>0.35*</b>	0.25	-0.37
<b>FAH -R</b>	-0.09	-0.06	-0.04	-0.02	-0.16
<b>FAH -L</b>	<b>-0.31*</b>	0.09	0.20	0.09	<b>-0.33*</b>
<b>MW -R</b>	-0.05	0.12	-0.06	-0.07	0.2
<b>MW -L</b>	<b>-0.29*</b>	0.26	0.07	-0.06	-0.01
<b>SPA-R</b>	-0.15	0.13	0.13	0.01	-0.18
<b>SPA-L</b>	0.08	0.07	-0.03	0.05	-0.02
<b>ALD-R</b>	<b>-0.36*</b>	0.28	<b>0.39**</b>	<b>0.34*</b>	<b>-0.40**</b>
<b>ALD-L</b>	-0.21	0.13	0.27	0.16	<b>-0.32*</b>
* p<0.05					
** p<0.01					

(FPI: Foot Posture Index; FAH: Foot Arch Height; MW: Metatarsal Weight; SPA: subtalar pronation angle; ALD: Ankle Limitation Degree; R: Right; L: Left; 6MWT: 6-Meter Walk Test; 10MWT: 10-Meter Walk Test; NSAA: Nort Star Ambulatory Assessment)