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RESEARCH ARTICLE

THE ASSOCIATION BETWEEN LUMBAR LORDOSIS AND FUNCTIONAL PERFORMANCE IN CHILDREN WITH DUCHENNE MUSCULAR DYSTROPHY

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ABSTRACT

Purpose: The aims of this study were to compare the lumbar lordosis values measured with flexible ruler between healthy peers and children with Duchenne Muscular Dystrophy (DMD) and to investigate the relationship between lumbar lordosis values and functional performance in children with DMD.
Material and Methods: The study included 20 children with DMD (median age: 8.29 years) and 10 healthy peers (median age: 7.96 years). Lumbar lordosis values and postural alignment were assessed using a flexible ruler and New York Posture Rating (NYPR). Functional performance was evaluated with the 32-item Motor Function Measure and timed performance tests.
Results: The demographic characteristics such as age and body mass index were similar for both the children with DMD and the healthy peer group ($p>0.05$). A higher degree of lumbar lordosis was determined in the children with DMD compared to the healthy peers (median angle: 46.58° vs. 36.10°) ($p<0.001$). The children with DMD had greater disturbance according to the NYPR ($p=0.002$). Both the degree of lumbar lordosis and NYPR scores were moderately to strongly related to the functional performance parameters ($p<0.05$, rs:0.5 - 0.8 and -0.5 - -0.8).
Conclusion: Assessment of lumbar lordosis using a flexible ruler in DMD may be an alternative method to differentiate between children with DMD and healthy peers, and between children with DMD at different functional levels. This study may provide additional evidence regarding the possible relationships between postural alignment and motor function parameters.

Keywords: Duchenne muscular dystrophy, spine, lordosis, posture, physical functional performance

INTRODUCTION

Duchenne muscular dystrophy (DMD) is one of the most prevalent (7.1 per 100000 males) fatal diseases of childhood with X-linked recessive inheritance (1). The disease is caused by a mutation in the DMD gene responsible for the production of dystrophin protein (2). Although DMD is characterized by more prominent muscle weakness in proximal muscles, the disease affects all muscles in the body (3, 4). The first symptoms of the disease generally emerge around the age of 3 years, and include delay in motor milestones, frequent falls, fatigue, and difficulty in getting up from the floor. Functional performance parameters tend to improve until the age of 7 years (5, 6) while symptoms become more pronounced from this age onwards and children lose their walking ability around the ages of 10-12 years (7). Cardiac

and respiratory complications can be the cause of death for these children (8, 9).

Muscle weakness in DMD results in not only decreased physical performance but also joint contractures and postural changes. Ankle contractures, which usually appear as a primary deformity, have serious side effects on functional performance (9). In a study on this subject, increased ankle contractures were found to be associated with decreased functional performance in DMD (10). However, postural alignment disturbances such as scoliosis, kyphosis and lordosis are observed at different frequencies in neuromuscular diseases (11). Increased lumbar lordosis is a compensatory mechanism in response to the pelvic girdle muscle weakness. This weakness leads to deterioration in pelvis stability, and the compensatory shortness of the iliotibial band often forces the pelvis into anterior tilt. The lumbar lordosis automatically increases to balance the compensations in the pelvis and lower limbs (12). Baptista et al. evaluated the postural alignment of children with DMD using digital photogrammetry and found a relationship between forward displacement of the centre of mass and pelvic anteversion with balance deficit (3). Increased lumbar lordosis has also been reported to negatively affect gait quality in addition to balance (12).

There is currently no cure for DMD. Disease management generally consists of symptomatic treatments such as rehabilitation, and cardiac and respiratory drugs (8, 9). It is well known that to increase the efficacy of these treatments, the symptoms should be detected at an early age while physical performance is still preserved (13). Therefore, to be able to detect symptoms as early as possible, it is crucial to select outcome measures that are practical, low-cost, provide rapid results and are relevant to the disease (14). Thus, early detection of postural changes including lumbar lordosis due to muscle weakness in the early period of DMD is one of the factors that increase the effectiveness of the treatment. In a recent study in which lumbar lordosis values were determined using a digital inclinometer, it was found that the lumbar lordosis of children with DMD was high and these values were associated with gait and balance parameters (12). However, it remains unclear whether the flexible ruler, which is valid and reliable in the measurement of lumbar lordosis in children, is an alternative to a digital inclinometer and whether lumbar lordosis is related to functional performance other than gait and balance.

The aims of this study were to compare the lumbar lordosis values assessed with a flexible ruler between healthy peers and children with DMD and to investigate the relationship between the severity of lumbar lordosis and functional performance.

MATERIAL AND METHODS.

Design and Participants

This cross-sectional observational study was conducted at Hacettepe University, Faculty of Physical Therapy and Rehabilitation between October 2022 and May 2023. Approval for the study was obtained from the Hacettepe University Non-Interventional Clinical Studies Ethics Board (Decision Date: 18.10.2022, Number: 2022/16-27). Written informed consent was obtained from children and their families, and the study was conducted in accordance with the Declaration of Helsinki.

The study included children aged 5-12 years, who were genetically confirmed, Level 1-3 on the Vignos scale, and using steroids regularly for at least six months. Children with an additional chronic disease, severe joint contractures and shortness, inadequate co-operation, or history of an injury or surgery in the last 6 months were excluded from the study. The control group was formed of healthy peers with no history of any chronic diseases and injuries or surgery in the last 6 months. A total of 22 children with DMD and 11 healthy peers were initially enrolled in the study. Two children with DMD were excluded because of a history of lower extremity fracture and insufficient co-operation, and one of the healthy children was excluded because of regular use of immunosuppressive medication. The study was completed with a total of 30 children (20 DMD and 10 healthy subjects).

Outcome measures

The demographic characteristics of all the children such as age, height, weight, and dominant side were recorded.

Functional Level

The Vignos Scale was used to determine the functional level of the children with DMD (15). The Vignos scale classifies children with DMD into different levels, ranging from 1 to 10. Level 1 indicates that the patient can walk independently and climb stairs without assistance, while Level 10 indicates that the patient is confined to bed. The children with DMD who were included in this study were those who

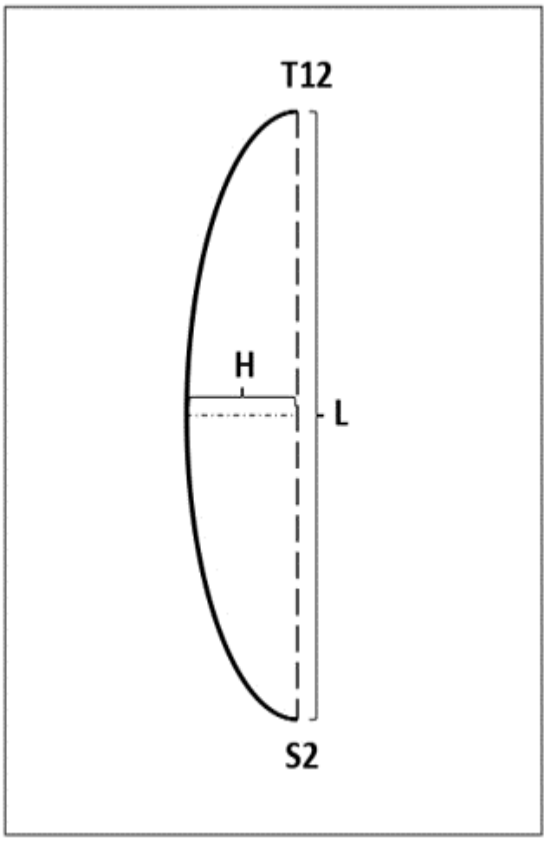

Figure 1. The measurement of lumbar lordosis with the flexible ruler

could climb stairs with or without support (Levels 1-3). This scale has been shown to have high ICC in the assessment of DMD children (16).

Lumbar Lordosis

A flexible ruler was used to assess lumbar lordosis in the children in this study (17, 18). The children were asked to stand barefoot between two parallel bands with the feet 15 cm apart, and their weight evenly distributed on both legs, their arms at the sides of the trunk and facing the opposite wall in a comfortable position for the assessment. In this position, a removable mark was placed on the children's spinous processes of T12 and S2. The flexible ruler was placed between these two marked points in the lumbar region and the obtained shape was transferred onto paper without distortion. The T12 and S2 points on the paper were connected vertically (L), and the distance between this vertical line and the maximum curvature was calculated (H) (Figure 1). The lumbar lordosis angle (θ) was calculated according to the following formula (19); θ=4X[Arctan2H/L]. Although this method has been shown to be highly reliable in children, as it was being used for the first time in the DMD population, reliability was tested in a preliminary study.

Postural Alignment

Postural characteristics were assessed with the New York Posture Rating Scale (NYPR) modified by Howly and Franks (20). Postural changes in 10 different parts of the body such as the head, trunk and hips were observed either lateral or posterior, and scored. Each body part of each child was evaluated by giving a score of good posture (10 points), fair (5 points), or poor posture (0 points). The NYPR was determined to have moderate ICC (0.70) value (21).

Functional Performance

Timed performance tests consisting of (a) supine to stand (Gowers' maneuver), (b) ascending and descending four steps of 25 cm width and 17 cm height with handrails, and (c) 10-m walk/run test were used to determine the functional performance of the children with DMD (22). Each timed performance test score was recorded in seconds.

Motor Function

The 32-item Motor Function Measure (MFM-32), which has been shown to be valid and reliable in neuromuscular diseases in the age range of 6-60 years, was used to assess motor function in the children with DMD. This outcome measure assesses function in 3 different dimensions [sitting and transfers (D1), proximal/axial (D2) and distal (D3)] over a total of 32 items. Items are generally scored on a four-point Likert scale; (0) cannot initiate any task and cannot maintain the starting position, (1) partially completes the task, (2) performs the task slowly, with some help and compensations, and (3) performs the task in a standardized pattern. Each sub-dimension and the total score are converted into a percentage score. Higher scores indicate better motor functioning (23, 24). This assessment was performed by a certified physiotherapist (NB).

Assessment Procedure

The assessments of functional level, motor function and functional performance were only applied to the children with DMD, while all other assessments were performed on all children. One physiotherapist carried out the postural assessments and the other performed the motor performance tests. Short resting periods were allowed after each assessment to avoid

Table 1. Demographic and postural characteristics of children with Duchenne muscular dystrophy at different functional levels and healthy peers.

Variables	Children with DMD[a]		*p (comparison between DMD groups)*	Healthy peers (n=10)	*p (comparison between groups)*
	Level 1 (n=10)	Level 2 (n=10)			
Age (years)	8.04 (7.19-9.04)	8.29 (7.52-10.12)	0.4	7.96 (7.23-8.92)	0.6
Height (m)	1.23 (1.17-1.33)	1.28 (1.20-1.35)	0.4	1.31 (1.27-1.40)	0.06
Body weight (kg)	23.50 (20.5-29.37)	30.75 (22.62-35.12)	0.2	28.80 (22.25-34.50)	0.5
Body Mass Index (percentiles)	45.50 (15.75-76.00)	84.50 (65.25-91.25)	0.06	62.50 (35.50-80.50)	0.5
Lumbar Lordosis (°)	43.15 (37.23-46.40)	53.89 (47.62-63.01)	0.005*	36.10 (33.36-38.91)	<0.001*
NYPR (points)	75.00 (70.00-90.00)	65.00 (60.00-75.00)	0.01*	95.00 (90.00-100.00)	0.002*

a According to Vignos Scale. Data expressed as median (25th and 75th percentiles).*Mann Whitney-U Test DMD: Duchenne Muscular Dystrophy, NYPR: New York Posture Rating

Table 2. The results of functional assessments of children with Duchenne Muscular Dystrophy.

Functional Assessments	Children with DMD[a]		*p*
	Level 1 (n=10)	Level 2 (n=10)	
Motor Function Measure			
Total Score (%)	85.42 (82.03-91.93)	80.21 (69.53-84.64)	0.02*
Dimension 1 (%)	73.08 (66.03-82.69)	57.69 (44.87-90.97)	0.004*
Dimension 2 (%)	97.22 (94.44-100.00)	94.44 (90.97-97.92)	0.1
Dimension 3 (%)	90.48 (89.29-96.43)	95.24 (90.48-95.24)	0.7
Supine to stand (sec)	4.80 (2.81-8.59)	7.12 (4.91-17.61)	0.09
Ascending four stairs (sec)	2.41 (2.18-2.91)	3.79 (2.99-9.49)	0.01*
Descending four stairs (sec)	2.22 (1.80-3.23)	3.50 (2.00-5.09)	0.2
10 meter walk/run test (sec)	4.70 (4.24-5.02)	5.91 (4.82-8.40)	0.03*

a According to Vignos Scale. Data expressed as median (25th and 75th percentiles).*Mann Whitney-U Test, DMD: Duchenne Muscular Dystrophy.

fatigue in the children. The assessments lasted about 50 minutes in total for the children with DMD and 10 minutes for the healthy children.

Statistical Analysis

Data obtained in the study were analyzed statistically using IBM SPSS vn 23.0 software (SPSS Inc., Chicago, IL, USA). Conformity of the data to normal distribution was evaluated with the Shapiro-Wilk test and it was determined that the data did not fit normal distribution. Scores were expressed as median and percentiles (25th and 75th percentiles). The Mann-Whitney U test was used to compare the lumbar lordosis and postural alignment scores of both children with DMD at different levels and children with DMD and their healthy peers. The relationship between postural alignment parameters and motor performance scores in children with DMD was analyzed using Spearman rank correlation coefficient (rs). The strength of correlations were determined as; rs = 0.7–0.99 strong; rs = 0.4–0.69 moderate; and rs =0.01-0.39 weak (25). A value of $p<0.05$ was accepted as statistical significance level.

In the post-hoc power analysis performed with G*Power 3.1 using the lumbar lordosis values of DMD and healthy peers, the effect size (Cohen's *d*) was 1.38 and the power of the study was 93.0%.

RESULTS

The demographic and postural characteristics of the children with DMD at different functional levels and their healthy peers are shown in Table 1. The demographic characteristics were similar in both the children with DMD (median age: 8.29 years) and their

Table 3. The correlations between postural alignment parameters and functional assessments (n=20).

	Lumbar Lordosis		New York Posture Rating	
	r_s	*p*	r_s	*p*
Motor Function Measure				
Total Score	-0.5*	0.01	0.6*	0.002
Dimension 1	-0.6*	0.002	0.8*	<0.001
Dimension 2	-0.3	0.1	0.3	0.1
Dimension 3	-0.08	0.7	0.01	>0.9
Supine to stand	0.6*	0.002	-0.6*	0.006
Ascending four stairs	0.7*	<0.001	-0.7*	0.001
Descending four stairs	0.3	0.1	-0.4	0.09
10 meter walk/run test	0.8*	<0.001	-0.5*	0.01

*Spearmann Correlation Coefficient.

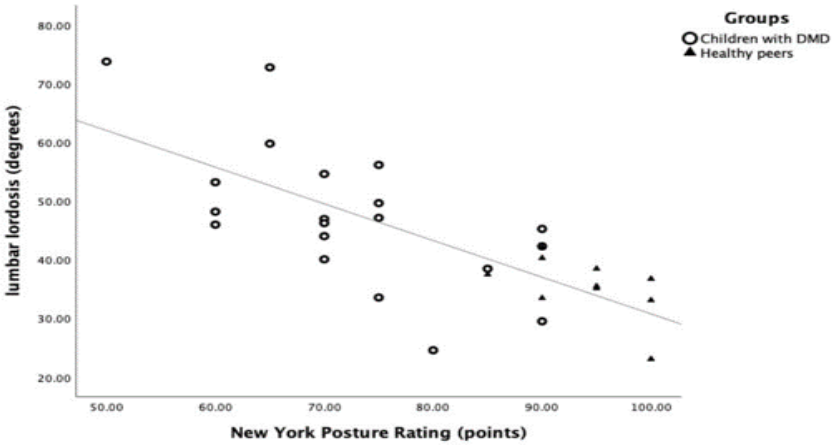

Figure 2. The results of postural alignment of children with Duchenne muscular dystrophy and healthy peers (DMD: Duchenne muscular dystrophy)

healthy peers (median age: 7.96 years) (p>0.05). All the children included in the study were right-handed.

The degree of lumbar lordosis was determined to be statistically significantly higher in the children with DMD compared to the healthy peers (median angle: 46.58° vs. 36.10°) (p<0.001). The flexible ruler was found to have high inter-rater reliability (ICC: 0.925; p<0.001) in a preliminary study involving 14 children with DMD. The lumbar lordosis values measured with the flexible ruler and the NYPR scores of each child are shown in Figure 2. A strong positive correlation was determined between the NYPR scores and lumbar lordosis in the children with DMD (rs= 0.8, p<0.001).

The functional assessment results of the children with DMD are presented in Table 2. The MFM total score, MFM D1, ascending four stairs and 10 m walk/run test results were significantly better in the children with Level 1 DMD (p<0.05).

Moderate to strong relationships were determined between postural alignment and motor performance test parameters, as shown in Table 3 (p<0.05).

DISCUSSION

The results of this study, which assessed lumbar lordosis in children with DMD using a flexible ruler, showed that children with DMD had increased lumbar lordosis in the ambulatory period compared to their

healthy peers. It was determined that with the worsening of functional ability, the lumbar lordosis increased, and postural alignment disturbances might be associated with the worsening functional performance. Considering the studies in the literature in which different methods have been used in the evaluation of lumbar lordosis (3, 12), it can be stated that the evaluation of lumbar lordosis with the flexible ruler reveals similar results in differentiating healthy children and those with DMD and provides information about physical performance.

Muscle weakness and imbalance, which are the main symptoms in children with DMD, cause joint contractures, postural deviations and deterioration in functional ability (3). Ankle contractures have been reported to occur in about half of children with DMD, even in the ambulatory phase, followed by hip and knee contractures (26). In addition to contractures, postural alignment disturbances such as an increased anterior position of the centre of mass and significant pelvic anteversion have been observed (3). Scoliosis and lordosis are also part of the postural alignment disturbances in DMD (11). Filiz et al. (12) reported that lumbar lordosis became more pronounced compared to healthy children, even at a stage that can be considered the early ambulatory phase according to the age of the children (mean age: 8.0 years). Similarly, it was seen in the current study that children with DMD had higher lumbar lordosis values and worse posture. This shows that the postural change is not only in the proximal region such as lumbar lordosis but also in the whole body. However, the lumbar lordosis values of both the healthy and DMD children in the current study were numerically different from those reported in other studies (12, 18). This may be due to differences in methodology and the age range of participants.

Due to the progressive nature of DMD, there are many reports of worsening symptoms such as contractures and gait. (27, 28). In other words, as performance decreases so the functional level worsens. Alkan et al. (29) stated that balance parameters deteriorated as the functional level worsened in children with DMD. The current study results showed that the lumbar lordosis values of children with Level 1 DMD, which is functionally better, were 10 degrees lower and their postural scores were 10 points higher than those of children with Level 2 DMD. From these results, it can be concluded that when functional loss occurs, it leads to holistic changes in the whole body.

In children who maintain their functional level until the age of 6-8 years, there are losses in both muscle strength and functional performance after this age, and children may be later confined to a wheelchair (30). In an 18-month follow-up study of steroid-using DMD children with a mean age of 7.6 years, it was reported that the MFM D1 sub-dimension and MFM total scores tended to decrease (31). Another DMD cohort study reported deterioration after 7 years of age in timed performance tests such as supine to stand, ascending four steps and 10 m walk/run test (22). The fact that the median age of the children with DMD in the current study was above 6-8 years, which is the critical threshold, may explain the difference in assessment parameters between the DMD and healthy peer groups. The age of children with DMD provides important information, but not all of it. Although the age of the children at two different functional levels was similar, the difference in proximal and axial motor function and in timed performance tests such as ascending four stairs and the 10-meter walk test can be explained by the holistic change mentioned above rather than by age.

Physical symptoms are expected to interact because many of the problems in DMD are related to muscle weakness. In this regard, there are studies showing a relationship between muscle strength and motor functions in children with DMD (32, 33). In addition, both proximal and distal postural alignment have been shown to be associated with balance and gait in children with DMD (3, 34). A study that focussed on lumbar lordosis reported a correlation between the lumbar lordosis values and gait quality and balance (12). The results of the current study showed an association of both lumbar lordosis values and postural alignment scores with proximal and functional performance parameters in the children with DMD. Thus, in addition to the information in the literature on the relationship between postural alignment and balance and gait, this study provides further evidence that postural alignment may be related to functional performance. This supports the view that problems which occur secondary to muscle weakness may influence each other. The main limitation of this study was the relatively low number of participants.

CONCLUSION

In conclusion, the assessment of lumbar lordosis using a flexible ruler can be used to differentiate between both children with DMD and healthy peers,

and children with DMD at different functional levels. In the light of these results, flexible ruler measurements may be an alternative option to the methods used for the assessment of lumbar lordosis in children with DMD. In addition to the existing knowledge about the relationships between postural alignment and gait and balance, this study may provide additional evidence in respect of the possible relationships between postural alignment and motor function parameters. To be able to confirm these results, there is a need for a cohort study including a wider spectrum of children. The use of flexible ruler measurements in future studies may be of guidance to researchers and clinicians for the determination of the cut-off value for lumbar lordosis.

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Author contribution: Numan Bulut: Conceptualization, Design, Data Collection, Literature Review Writing-Original Draft and Critical Review. İpek Gürbüz: Conceptualization, Design, Materials, Analysis, Writing Original Draft and Review. Öznur Yılmaz: Conceptualization, Design, Supervision, Interpretation, Writing Original Draft and Review. All authors approve the submitted and final version.

Conflict of interests: Approval for the study was obtained from the Hacettepe University Non-Interventional Clinical Studies Ethics Board (Decision Date: 18.10.2022, Number: 2022/16-27). Written informed consent was obtained from children and their families, and the study was conducted in accordance with the Declaration of Helsinki.

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