ORIGINAL ARTICLE

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The Effects of Joint Hypermobility on Quality of Life in Healthy School Children

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Abstract

Objective: This study aimed to determine the prevalence of joint hypermobility in healthy primary school children and to investigate the quality of life differences between children with and without joint hypermobility.

Methods: Joint hypermobility was investigated in healthy children aged 8-12 years. The "Beighton" score was evaluated for the diagnosis of joint hypermobility. According to these scores, children were divided into three groups as "joint hypermobility," "increased joint mobility" and "no joint hypermobility." All children were evaluated by the Pediatric Quality of Life Inventory (PedsQL) version 4 and quality of life scores were measured. The PedsQL results were compared between the groups in terms of statistical significance.

Results: The mean Beighton score of 378 students enrolled in the study was 2.4±2.2. The PedsQL mean total score was 77±13.3. Age, sex distribution, pediatric PedsQL total score, psychosocial score and physical health score did not differ significantly between groups.

Conclusion: Our results show that joint hypermobility does not affect the quality of life in healthy children but more extensive studies in this field are needed.

Keywords: Beighton score, joint hypermobility, quality of life, school

INTRODUCTION

Musculoskeletal pain is often seen in childhood. Sometimes it can be long-lasting and repetitive. One of the major causes of these pains is joint hypermobility. Joint hypermobility is defined as having a joint range of motion greater than normal limits (1,2). Joint hypermobility and joint laxity are used by some authors interchangeably (3). Although there are no definitive definitions, hypermobility is the ability of the joint to exceed its normal limits within its motion axis (especially in extension), and hyperlaxity is the ability of the joint to move in unusual motion axes (4). When more than one joint exceeds the joint motion limits within its own motion axes it is called generalized joint hypermobility. In hypermobility syndrome, the patient should have joint hypermobility as well as muscle and skeletal system symptoms and should not have hereditary connective tissue disease (5).

The incidence of joint hypermobility and benign joint hypermobility syndrome may differ greatly in different publications. This may be caused by the differences between the tests and the criteria used in the diagnosis of joint hypermobility. The lack of consensus on the cut-off value of the Beighton score, which is one of the most commonly used diagnostic

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tests, may also lead to different incidence rates. In his original article, Beighton accepted the score of 4 and for the diagnosis of hypermobility (6). Some authors accepted scores higher than 4 as the cut-off value (7-9). The frequency of joint hypermobility varies according to age, sex, and race (6,10,11). The frequency of asymptomatic hypermobility varies between 5 and 10% (11,12). In children, this rate was found as 10%-25% (13,14).

The measurement of quality of life is preferred today to evaluate the treatment results of rheumatic diseases in children (15). To assess the quality of life related to health, version 4 of the Pediatric Quality of Life Inventory (PedsQL) was created in 1999 by Varni et al. (16). There are different versions according to age groups. This scale, which is applied in the form of a questionnaire and can be completed by children aged 8-12 years, includes 23 questions about 4 different topics (16).

This study aimed to determine the prevalence of joint hypermobility in healthy primary school children and to investigate the quality of life differences between children with and without joint hypermobility.

METHODS

A total of 378 students, 211 (55.8%) females, 167 (44.2%) males, were included in the study. The mean age was 9.87 (range, 8-12) years. The criteria for inclusion (aged between 8-12 years, student of selected primary school, not having a physical, mental or developmental chronic disease and having informed consent) and exclusion (having a trauma in the last month which makes the patient unsuitable for evaluating joint movements or having sequela in extremities due to a recent trauma that limit joint movements) were applied to the students.

The students' Beighton scores were measured. All measurements were performed by two pediatric physicians using a goniometer. The Beighton score was established by Beighton in 1973 by modifying the Carter and Wilkinson criteria (6). Passive dorsiflexion of the little fingers beyond 90°, passive apposition of the thumbs to the flexor aspects of the forearms, hyperextension of the elbows beyond 10°, hyperextension of the knees beyond 10°, and forward flexion of the trunk with knees straight so that the palms of the hands rested easily on the floor, score points in the test. A total of nine points are evaluated. In his original article, Beighton accepted 4 as the cut-off value (6). Scores above 4 are required for the diagnosis of hypermobility.

The PedsQL version 4 questionnaire was then given to the students. In this survey, there were 23 questions under 4 main headings. In this questionnaire, the general health and activities of children (8 questions), emotional status (5 questions), social relations (5 questions), and school status (5 questions) were questioned. The answers and scores were as follows: No problems: 0 points; almost no problems: 1 point; problems sometimes: 2 points; problems often: 3 points; almost always problems: 4 points. The total score was inverted and was rated on a 0-100 scale. The mean score of the eight questions related to the "my health and activities" section was taken for the mean score of physical health, and the total mean score of the emotional status, social relations, and school status sections were taken for the psychosocial mean score.

The cut-off Beighton score for the diagnosis of joint hypermobility was accepted as 7. Children with ≥7 Beighton scores were diagnosed with joint hypermobility (group 3). Children with ≤4 Beighton scores were accepted as normal (group 1). Children with 5-6 Beighton scores were included in the "increased joint mobility" group (group 2). The difference between these groups in terms of the PedsQL results was investigated.

The study was approved by University of Health Sciences Turkey, Okmeydani Training and Research Hospital Ethics Committee (approval no: 25/10/2016-535) and a permission certificate was issued by the Istanbul Provincial Directorate of National Education (date: 13/12/2016, no: 14083890). Then, a "Child Consent Form for Research Purposes" and an "Informed Consent Form" for the approval of their families were distributed to the classes of students aged between 8 and 12 years in a public primary school.

Statistical Analysis

The mean, standard deviation, median, maximum, frequency, and ratio values were used in descriptive statistics (Table 1). The distribution of variables was measured using the Kolmogorov-Smirnov test. The Kruskal-Wallis test was used to analyze quantitative data. The chi-square test was used for the analysis of qualitative independent data. The SPSS 22.0 program was used for analysis.

RESULTS

The mean Beighton score was 2.39±2.2 (0-9) (Table 2). Positivity of apposition of the thumbs was found in 90 children, passive dorsiflexion of the little fingers in 118 children, hyperextension of the elbows in 85 children, hyperextension of the knees in 108 children, and resting of the palms of the hands on the floor with flexion of the trunk in 160 children (Table 3).

There was no difference between groups 1, 2, and 3 in terms of age and sex ($p>0.05$) (Figure 1). The PedsQL total score,

Table 1. Descriptive statistics				
		Min-max	Median	Mean ± SD
Age		8.0- 12.0	10.0	9.9±1.2
Sex	**Female**	-	-	211±55.8%
	Male	-	-	167±44.2%
Beighton scoring		0.0-9.0	2.0	2.4±2.2
Apposition of the right thumb		-	-	84±22.2%
Apposition of the left thumb		-	-	79±20.9%
Dorsiflexion of the right little finger		-	-	115±30.4%
Dorsiflexion of the left little finger		-	-	108±28.6%
Hyperextension of the right elbow		-	-	81±21.4%
Hyperextension of the left elbow		-	-	69±18.3%
Hyperextension of the right knee		-	-	108±28.6%
Hyperextension of the left knee		-	-	106±28.0%
Placing the palms of the hands on the floor		-	-	160±42.3%
Pediatric quality of life inventory total score		24-100	78.3	77.0±13.3
Physicosocial score		22-100	79.2	77.1±14.6
Physical total score		28-100	78.1	76.7±15.4
General health and activities question (GHAQ) 1		0-100	75.0	72.8±29.5
GHAQ 2		0-100	100.0	73.3±31.1
GHAQ 3		0-100	100.0	87.2±22.3
GHAQ 4		0-100	50.0	58.0±34.5
GHAQ 5		0-100	100.0	89.5±23.7
GHAQ 6		0-100	100.0	86.8±23.3
GHAQ 7		0-100	100.0	75.3±29.4
GHAQ 8		0-100	75.0	70.6±29.8
Emotional status question (ESQ) 1		0-100	100.0	75.5±29.6
ESQ 2		0-100	100.0	78.4±27.0
ESQ 3		0-100	100.0	75.5±29.1
ESQ 4		0-100	100.0	73.1±34.1
ESQ 5		0-100	100.0	74.9±30.7
Social relations question (SRQ) 1		0-100	100.0	83.9±25.8
SRQ 2		0-100	100.0	83.7±26.9
SRQ 3		0-100	100.0	85.7±25.2
SRQ 4		0-100	100.0	80.1±26.2
SRQ 5		0-100	100.0	77.6±31.4
School status question (SSQ) 1		0-100	100.0	80.3±28.5
SSQ 2		0-100	75.0	73.3±28.1
SSQ 3		0-100	100.0	82.6±25.6
SSQ 4		0-100	100.0	74.2±30.5
SSQ 5		0-100	50.0	58.1±33.6
SD: Standard deviation, min: Minimum, max: Maximum				

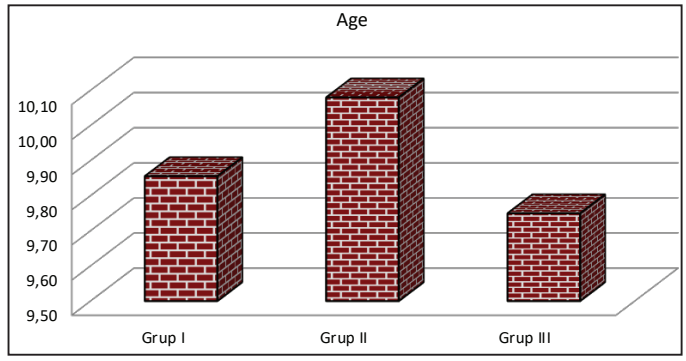

Figure 1. Distribution of age in the groups

Table 2. The distribution of students according to the Beighton score		
Beighton score	**Number of students**	**Percentage in all students**
9	11	2.91
8	4	1.06
7	29	7.67
6	18	4.76
5	32	8.47
4	16	4.23
3	51	13.49
2	29	7.67
1	30	7.94
0	158	41.8

psychosocial score, and physical health score did not differ significantly between groups 1, 2, and 3 ($p>0.05$). There was no difference between groups 1, 2, and 3 in terms of sections in the PedsQL ($p>0.05$) (Table 4).

DISCUSSION

Beighton et al. (6) determined the cut-off value for the diagnosis of joint hypermobility as 4 and it was reported that joint range of motion reduced with age from childhood. According to this, the Beighton cut-off value for the diagnosis of joint hypermobility in children should be a higher value than the cut-off value for adults. Jansson et al. (10) scanned hypermobility in 1.845 children and stated that the Beighton cut-off should be 8 for children aged 9 years; 7 for girls and 6 for boys aged 12 years; and 8 for girls and 6 for boys aged 15 years to be able to define 95% of the children as normal. Mikkelsson et al. (7) stated that the cut-off was 6 in hypermobility scans in 1.637 students. Remvig et al. (17) stated in a literature review of generalized joint hypermobility and benign joint hypermobility that the cut-off value of the Beighton score ranged from 2 to 7 in several publications. To investigate the validity and reproducibility of

Table 3. The rate of the Beighton score positivity among all students				
Beighton score	**Positivity in the right**	**Positivity in the left**	**Bilateral positivity**	**Total**
Apposition of the thumbs	11 (2.91%)	6 (1.59%)	73 (19.31%)	90 (23.8%)
Passive dorsiflexion of the little fingers	10 (2.64%)	3 (0.79%)	105 (27.78%)	118 (31.21%)
Hyperextension of the elbows	16 (4.23%)	4 (1.05%)	65 (17.19%)	85 (22.48%)
Hyperextension of the knees	2 (0.52%)	0 (0%)	106 (28.04%)	108 (28.57%)
Placing the palms of the hands on the floor with lomber flexion	-	-	-	160 (42.32%)

Table 4. The distribution of the pediatric quality of life scores in the groups								
	Group 1		**Group 2**		**Group 3**		**p**	
	Mean ± SD	**Median**	**Mean ± SD**	**Median**	**Mean ± SD**	**Median**		
The pediatric quality of life inventory								
Total score	76.7±13.1	78.3	77.6±13.5	77.7	77.8±14.9	79.3	0.719	[K]
Psycosocial score	76.7±14.3	78.3	78.8±14.8	80.0	78.0±16.4	80.0	0.454	[K]
Physical health score	76.8±15.1	78.1	75.3±16.3	78.1	77.3±6.5	79.7	0.776	[K]
[K]Kruskal-Wallis test, SD: Standard deviation								

the Beighton score, Smits-Engelsman et al. (18) conducted this test on 551 children aged between 6 and 12 years and reported a cut-off value of 7. Hypermobility frequency is reported to vary according to age, sex, and race in various publications (10,19,20).

In a study conducted in our country, Koldaş Doğan et al. (21) accepted the cut-off value of the Beighton score as 4 in children aged 7-12 years with attention-deficit/hyperactivity disorder. Yazgan et al. (22) accepted the cut-off value of the Beighton score as 4 in a study of 922 children aged between 5 and 10 years and 363 (39.3%) children were accepted as having joint hypermobility. Yıldırım (23) accepted the Beighton cut-off value as 6, and 118 (13.8%) of 857 children were diagnosed with joint hypermobility.

In our study, to distinguish children with joint hypermobility from children without joint hypermobility, children were divided into three groups according to their Beighton scores as Smits-Engelsman et al. (18) did and a group of children with increased joint mobility was added to the groups comprising children with and without joint hypermobility. The proportion of children diagnosed with joint hypermobility was found to be consistent with the literature.

The presence of hypermobility in females is higher than in males (10,20). In our study, although the ratio of girls in the group with joint hypermobility was higher, there was no statistically significant difference test between the groups. We think that this is because of differences in the children's age. Jansson et al. (10) determined different Beighton score cut-off values in different age groups according to sex in their publication. In our study, we believe that the use of the same cut-off value for all children aged between 8 and 12 years without regard to sex and age increased the rate of the girls in the group with joint hypermobility.

A decrease in the frequency of joint hypermobility with age has been reported in several publications (6,20). In spite of this, Mikkelsson et al. (7) and Ruperto et al. (24) were unable to confirm this in their studies. In our study, the age range in the inclusion criteria was narrow. We believe that the mean age between the groups was not different due to our inclusion criteria.

The frequency of joints with hypermobility in the Beighton scoring system varies in studies. El-Garf et al. (25) reported that the most frequently observed hypermobile joint was the finger joint, and the least frequently observed hypermobile joint was knee joint in 997 children in Egypt. Lamari et al. (26) and Silman et al. (27) indicated that the most frequently observed hypermobile joint was finger joint in children and adolescents. Adib et al. (28) indicated that the most frequently observed hypermobile joint the knee joint in children. In our study, 160 of 378 (42.32%) children rested easily on the floor with the palms of the hands with forward flexion of the trunk and with knees straight, and they scored points from this criterion in the Beighton score. The least frequently observed hypermobile joint was the elbow in our study. The apposition of the thumb was found in 90 (23.8%) children, passive hyperextension of the little finger in 118 (31.2%) children, hyperextension of the elbow in 85 (22.48%) children, and hyperextension of knee in 108 (28.5%) children; all these score points in the test.

The validity and reliability of the Turkish PedsQL 4 was tested in 2008 by Çakın Memik et al. (29). The total score, psychosocial score and physical health score did not differ significantly between the 3 groups in our study. When we examined each question, we found that the 4th question (Is it hard for me to lift something heavy?) of the "My Health and My Activities" section, which examines physical health, was the question with lowest average score in all three groups. Çakın Memik et al. (29) also encountered this finding and stated that this could be because the child perceived to be carrying more weight than the weight they could lift. The average score of the last question about school functionality (Are there times you can't go to school because you go to doctor or hospital?) was lower than other questions in all 3 groups. It was stated that this could be because the child perceived as they could not go to school when they became sick.

In the literature, the quality of life scores were found to be statistically lower in patients with joint hypermobility syndrome compared with the control group (15,30,31). Pacey et al. (30) correlated this difference in quality of life with pain, fatigue, and incontinence. Mastoroudes et al. (31) showed a significantly higher prevalence of urinary incontinence in females with joint hypermobility than controls and correlated the difference in quality of life with incontinence. Fatoye et al. (15) compared the quality of life of 29 children with hypermobility syndrome between the ages of 8 and 15 years and 37 healthy children and found that the quality of life score was lower in children with hypermobility syndrome. Although Beighton et al. (6) mentioned a positive relationship between mobility scores and musculoskeletal symptoms, we found no difference between the groups with and without joint hypermobility in terms of quality of life total scores, physical scores, emotional scores, and school scores. This may be due to "pain in four or more joints for 3 months or longer," a major Beighton criterion for the diagnosis of joint hypermobility syndrome. Russek and Errico (32) showed a statistically significantly higher frequency of sprain and back pain in patients with joint hypermobility syndrome than the control group, but they found no difference between patients with generalized joint hypermobility and the control group in terms of sprain and back pain. McCluskey et al. (33) observed no relation between musculoskeletal pain and joint hypermobility in European children, but they did find a relation in Afro-Asians in their meta-analysis on the relation between musculoskeletal pain and joint hypermobility in children. In a study of 1230 children aged between 7 and 15 years, Leone et al. (34) found no positive correlation between joint hypermobility and pain.

CONCLUSION

In conclusion, our results show that joint hypermobility does not affect the quality of life in healthy children. More extensive prospective studies on relation between joint hypermobility and musculo skeletal pain are needed. The appropriateness of beighton cut off values for age, sex and race can reduce the high prevalence rates of joint hypermobility.

Ethics

Ethics Committee Approval: The study was approved by University of Health Sciences Turkey, Okmeydani Training and Research Hospital Ethics Committee (approval no: 25/10/2016-535) and a permission certificate was issued by the Istanbul Provincial Directorate of National Education (date: 13/12/2016, no: 14083890).

Informed Consent: Consent was received.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Concept: O.Ö., A.S., Design: O.Ö., A.S., Data Collection or Processing: M.S.S., A.S., B.Y.D., Analysis or Interpretation: O.Ö., E.T., A.S., B.Y.D., M.S.S., Literature Search: B.Y.D., A.S., M.S.S., Writing: A.S.

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