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Hypermobility in Turkish Schoolchildren: Musculoskeletal Pain, Physical Activity, Balance, and Quality of Life

Türk Okul Çocuklarında Hipermobilite: Kasiskelet Ağrısı, Fiziksel Aktivite, Denge ve Yaşam Kalitesi

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Abstract

Aim: Joint hypermobility is a term used to describe an excessive range of joint motion. Joint hypermobility can be symptomatic or not. The present study aimed primarily to define the prevalence of joint hypermobility in healthy schoolchildren, and secondly, to determine the relationship between hypermobility and pain, physical activity, joint injury, quality of life, and balance.

Material and Method: In this cross-sectional study, the joints of 737 schoolchildren, aged 8 to 15 years, were examined according to the Beighton score (BS). Generalized joint hypermobility was defined by using a cut-off point of ≥6 joints. The participants with a BS between 1 and 5 were accepted as localized hypermobile. If the Beighton score was 0, the participants were accepted as non-hypermobile. Participants were evaluated using questionnaires or tests for pain, balance, physical activity, and quality of life.

Results: The 350 (47.5%) males and 387 (52.5%) females had a mean age of 11.47 ± 1.3 (8-15) years. The prevalence of generalized hypermobility was 13.4%, and we observed localized hypermobility in 65.9% of children and non-hypermobility in 20.6% of children. The most common pain localizations in children were neck (15.9%), lower back (13.7%), upper back (10.6%), shoulders (10.2%), and knees (7.9%). There was no association between pain and hypermobility in children aged 8 to 15 years.

Conclusion: The generalized joint hypermobility group was younger, shorter, and thinner than other groups. Additionally, we observed that hypermobility did not make a difference in terms of pain, quality of life, physical capacity, and balance in school-age Turkish children.

Keywords: Balance, injury, joint hypermobility, pain, quality of life, physical activity

Öz

Amaç: Eklem hipermobilitesi, aşırı eklem hareket aralığını tanımlamak için kullanılan bir terimdir ve semptomatik olabilir. Bu çalışma öncelikle sağlıklı okul çocuklarında eklem hipermobilitesinin prevalansını belirlemeyi ve ikinci olarak hipermobilite ile ağrı, fiziksel aktivite, eklem yaralanması, yaşam kalitesi ve denge arasındaki ilişkiyi belirlemeyi amaçlamıştır.

Gereç ve Yöntem: Bu kesitsel çalışmada, 8-15 yaşları arasındaki 737 okul çocuğunun eklemleri hipermibilite açısından Beighton skoruna göre incelendi. Beighton skoru 6 eklem ve üzeri ise jeneralize eklem hipermobilitesi, 1 ile 5 arasında ise lokalize eklem hipermobilitesi, puan 0 ise hipermobil olmayan olarak kabul edildi. Katılımcılar ağrı, denge, fiziksel aktivite ve yaşam kalitesi için anketler veya testler kullanılarak değerlendirildi.

Bulgular: Katılımcıların 350'si (%47,5) erkek, 387'si (%52,5) kadın ve ortalama yaşları 11,47 ± 1,3 (8-15) yıldı. Jeneralize eklem hipermobilite prevalansı %13,4 idi. Çocukların %65,9'unda lokalize hipermobilite ve %20,6'sında hipermobilite olmadığını gözlemledik. Çocuklarda en sık ağrı lokalizasyonları boyun (%15,9), bel (%13,7), üst sırt (%10,6), omuzlar (%10,2) ve dizler (%7,9) idi. 8-15 yaş arası çocuklarda ağrı ve hipermobilite arasında anlamlı bir ilişki yoktu.

Sonuç: Jeneralize eklem hipermobilitesi olan çocuklar diğer gruplara göre daha genç, daha kısa ve daha inceydi. Ayrıca okul çağındaki Türk çocuklarında hipermobilite varlığının ağrı, yaşam kalitesi, fiziksel kapasite ve denge açısından fark yaratmadığını gözlemledik.

Anahtar Kelimeler: Denge, yaralanma, eklem hipermobilitesi, ağrı, yaşam kalitesi, fiziksel aktivite

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INTRODUCTION

Joint hypermobility (JH) is used to describe an excessive range of joint motion. JH across multiple joints is termed generalized joint hypermobility (GJH). Although GJH is usually isolated and asymptomatic, it can be a part of syndromic diseases (heritable disorders of connective tissue such as Ehlers-Danlos syndromes (EDS), Marfan syndrome, and Osteogenesis imperfecta), and can be symptomatic with pain. Before 2017, the recommended terminologies for symptomatic and/or syndromic GJH were similar and there was a lack of globally accepted description or nomenclature for joint hypermobility.[1] In 2017, the International Consortium on EDS recommended using "hypermobility spectrum disorder" (HSD) for symptomatic, non-syndromic JH.[2,3] The symptomatic JH included single, localized or generalized subtypes. Joint hypermobility without musculoskeletal symptoms was termed asymptomatic GJH.

The Beighton score (BS) is a valid scale to identify JH in children and is calculated through 5 movements and 9 points.

In recent studies, the prevalence of GJH in children was reported between 7% and 36%. The discrepancy of outcomes might be a result of differences in study design; such as the threshold of BS, age of study groups, characteristics of study populations (healthy children, athletes, dancers, or symptomatic cases), and ethnicities. Therefore, the rates of GJH in children are seen as highly variable.[4]

In addition, patients with HSD may present with chronic musculoskeletal pain (MSP), soft tissue injuries, fatigue, decrease in physical capacity, balance, proprioception, and problems with motor development.[4] On the other hand, these hypermobile people may have an advantage in some activities like dancing, gymnastics.[5]

In the present study, we aim primarily to define the prevalence of JH in Turkish schoolchildren, and to classify the children as non-hypermobile (BS=0), localize hypermobile (BS=1-5), and generalized hypermobile (BS=6-9). A second aim is to evaluate these three groups in terms of hypermobility-related conditions such as MSP, physical activity, joint injury, quality of life, and balance in healthy schoolchildren.

MATERIAL AND METHOD

Study Group and Design

This cross-sectional study took place in Denizli, a city westward of Turkey. Initially, the document describing the study was shared with directors of schools and the Ministry of Education for permission. After approving the study, a written consent form and a letter that included information about JH, and the study design were sent to parents or legal guardians of 1100 students. Participation in this study was optional, and children with chronic diseases or without signed consent forms were excluded. The study included 737 school children, aged 8 to 15 years, from four different schools. The clinical research ethics committee of Pamukkale University, Faculty of Medicine (decision date, 03/05/2016) approved this study and all procedures were conducted according to the Declaration of Helsinki.

Clinic features and screening tests

1. Demographic and anthropometric parameters

   Age, gender, weight, and height of participants were collected and the body mass index (BMI) was calculated. Children were categorized according to BMI, as underweight, normal weight, overweight and obese.

2. The screening of generalized joint hypermobility

   Joint hypermobility was evaluated using the BS.[5] The BS includes 5 movements and ranges from 0 to 9 points. Four movements are calculated for the left and right sides separately (**Figure 1**). The BS has a high inter-researcher reproducibility in adults and children. It is a well-known, commonly used, valuable, and reliable scale of JH in children.[5-8] Ten pairs of qualified physiotherapists measured the joint angles with a goniometer. There is no fully accepted cut-off value of BS in children; we calculated the prevalence of GJH according to threshold 4, 5, and 6. However, GJH was defined using a cut-off point of ≥6 joints in this study. The participants with a BS between 1 and 5 were accepted as localized hypermobile (LJH). If the BS was 0, the participants were accepted as non-hypermobile (NH).

3. The screening of musculoskeletal pain and injury during daily life and physical activity

Little fingers[1]	Thumbs[2]	Elbows[3]	Knees[4]	Trunk[5]
2 points	2 points	2 points	2 points	1 points

1. Passive dorsiflexion of the little fingers over 90º (bilateral)
2. Passive apposition of the thumbs to the anterior of the forearm (bilateral)
3. Hyperextension of the elbows over 10º (bilateral)
4. Hyperextension of the knee over 10º (bilateral)
5. Anterior flexion of the trunk without bending knees and the palms flat on the floor

Figure 1. The 9-point maneuvers of Beighton score for diagnosis joint hypermobility

A questionnaire was designed to investigate MSP in schoolchildren. A visual analogue scale (VAS) and a diagram of body parts were used to define the intensity and location of the pain. Five physiotherapists asked the following questions to the participants one by one.

- Have you ever suffered from musculoskeletal pain during your daily life (MSP-DL) in the last month?
  - If yes, show the painful areas on the body diagram.
  - Give a number for your pain severity in daily life between 0-10. (VAS-DL)
- Have you ever suffered from musculoskeletal pain during or after physical activity (MSP-PA) in the last month without injury and trauma?

- If yes, show the painful areas on the body diagram.
- Give a number for your pain severity after physical activity between 0-10. (VAS-PA)

• Have you ever suffered from musculoskeletal injuries?

- If yes, show the injury areas on the body diagram.

4. The screening of hypermobility related conditions

The NH, LJH, and GJH groups were evaluated for physical activity, quality of life, and balance using the physical activity questionnaire for older children (PAQ-C), Paediatric Quality of Life Inventory 4.0 (PedsQL 4.0), Flamingo Balance Test (FBT). The experienced physiotherapists ensured the correct filling of the questionnaires and evaluated FBT carefully. The validity and reliability of PAQ-C and PedsQL 4.0 tests were shown in Turkish children.[9,10]

• PAQ-C is a set of questions that focus on physical activities (sport, dance, game) and their frequency in a week.[11] The replies to questions are evaluated according to a 5-point score (1 point = no activity, 5 points = 7 times or more) and higher points indicate a greater rate of physical activity.

• PedsQL 4.0 is a self-reported questionnaire that includes 4 parts, and 23 items. It determines the physical, emotional, social, and school life of children.[12] The range of scores is between 0 and 100 where higher scores indicate better quality of life.

• FBT is a static balance test used in children.[13] The children were asked to stand barefoot on a balance board with one leg for 60 seconds. The dimensions of the board were 50 cm in length, 4 cm in height, and 3 cm in width. The test score was the number of floor touches with a free foot for 60 seconds. A higher score indicated poor static balance.

Statistical Analysis

Data of the present study were assessed using SPSS (Version 22.0). In this study, 850 participants must provide 90% power and 95% confidence level even in a weak correlation between the parameters (r=0.1).

The quantitative variables were evaluated using the Kolmogorov-Smirnov test, detrended Normal Q-Q Plot, and histogram to define whether they were normally distributed. Normally distributed data were expressed as mean, standard deviation (SD). Non-normally distributed data were presented median, minimum, and maximum. The categorical data were expressed in count and percentage.

Parametric tests (Student -t, ANOVA tests) were used to compare normally distributed independent quantitative variables. If a parametric test was not provided for quantitative parameters, the Mann-Whitney U test or Kruskal Wallis Variance Analysis were used to compare the independent groups. Differences between categorical data were analysed using the Chi-square test. In addition, relationships between variables were evaluated by Spearman or Pearson correlation tests. P values <.05 with a 95% confidence interval were considered significant.

RESULTS

The Demographic Parameters, Hypermobility, and Hypermobility Related Conditions

1. Demographic Parameters

The 350 (47.5%) males and 387 (52.5%) females had a mean age of 11.47 ± 1.3 (8-15) years. The median values of all participants in weight, height, and BMI were 43 (23-109) kg, 151 (126-185) cm, (11.89-36) kg/m2, respectively. According to BMI, the participants were 46.7% underweight, 45.6% normal weight, 6.5% overweight, and 1.2% obese.

There was no significant difference between girls and boys in parameters of age, weight, height, and BMI (**Table 1**).

2. The Beighton Score and Generalized Joint Hypermobility

Out of 737 children, 20.6% had a BS of 0 (no hypermobile joint) and 0.9% of children had a BS of 9. The mean BS was 2.74±2.19 (0-9). The frequency and distribution of BS in girls and boys are shown in **Figure 2**. There was a significant difference between girls and boys (p=0.0001) (**Table 1**).

The prevalence of GJH in different thresholds was defined as 34.1% according to a cut-off of ≥4, 19.7% according to a cut-off of ≥5, and 13.4% according to a cut-off of ≥6. There was no significant sex difference in the three groups (**Table 1**).

By using the new terminology of the 2017 International Consortium on EDS, we observed GJH in 13.4% of children (BS: 6-9), LJH in 65.9% of children (BS: 1-5), and NH in 20.6% of children. In the NH group, the prevalence of boys was significantly higher than girls (p=0.004) (**Table 1**). However, there was no significant difference in the gender parameter between LJH and GJH (**Table 1**).

3. Musculoskeletal Pain and İnjury

Out of 737 participants, 33.4% had MSP-DL and 39.9% had MSP-PA. The mean severity score of MSP was 1.11±1.89 during DL and 1.29±2.02 after PA.

The most common localizations of MSP were neck (15.9%), lower back (13.7%), upper back (10.6%), shoulders (10.2%), and knees (7.9%). In the present study, the prevalence of MSP in the upper back and shoulders were significantly higher in girls (p=0.008, p= 0.019) (**Table 1**).

Over half of children (N=395, 53.6%) reported joint injury. The distribution of joint injury was ankle (N= 346, 46.9%), finger (N= 24, 3.3%), wrist (N= 23, 3.1%), and knee (N=2, 0.3%). However, only 14.8% of children needed medical care due to injury.

There was no significant difference between girls and boys in parameters of MSP-DL, MSP-PA, medical care, and joint injury (**Table 1**). We did not observe a significant difference between boys and girls in the parameter of severity scores of MSP (VAS-DL and VAS-PA) (**Table 1**).

Table 1. Comparison of the demographic characteristics, pain, joint injury, medical care necessity, pain intensity, physical activity, life quality and balance between girls and boys

	Boys	Girls	p value
Mean age (year) ±SD	11.37±1.3	11.56±1.3	**p=0.037***
Mean height (cm) ±SD	151±10.48	151.2±10.11	p=0.344*
Mean weight (kg) ±SD	45.61±12.9	44.45±11.53	p=0.486*
Mean BMI (kg/m²)±SD	19.72±3.8	19.21±3.57	p=0.069*
Mean BS ± SD	2.47±2.19	2.98±2.16	**p=0.0001***
GJH N (%)			
Cut off≥ 4	108(14.7%)	143(19.4%)	p=0.081**
Cut off≥ 5	60(8.2%)	85(11.5%)	p=0.1**
Cut off≥ 6	39(5.3%)	60(8.1%)	p=0.083**
HSD N (%)			
NH (BS=0)	88(25.1%)	64(16.5%)	**p=0.004****
LJH (BS=1-5)	223(63.7%)	263(68%)	p=0.225**
GJH (BS=6-9)	39(11.1%)	60(15.5%)	p=0.083**
MSP-DL N (%)	107(30.6%)	139(35.9%)	p=0.124**
MSP-PA N (%)	131(37.4%)	163(42.1%)	p=0.194**
Mean VAS of MSP ±SD during daily life	1±1.86	1.21±1.92	p=0.089*
Mean VAS of MSP ±SD after physical activity	1.23±2.1	1.34±2	p=0.209*
Pain location			
Neck N (%)	48(13.7%)	69(17.8%)	p=0.127**
Upper back N (%)	26(7.4%)	52(13.4%)	**p=0.008****
Lower back N (%)	47(13.4%)	54(14%)	p=0.621**
Shoulders N (%)	26(7.4%)	49(12.7%)	**p=0.019****
Knees N (%)	23(6.6%)	35(9.0%)	p=0.213**
Joint injury N (%)	192(54.9%)	203(52.5%)	p=0.514**
Medical care N (%)	56(16%)	53(13.7%)	p=0.379**
Mean PAQ-C score ±SD	27.72±7.1	25.14±7.3	**p=0.0001***
PedsQL4.0			
Mean physical ±SD	84.96±13.45	81.2±14.96	**p=0.0001***
Mean emotional ±SD	77.13±18.65	74.83±19.72	p=0.142*
Mean social ±SD	88.97±15.08	90.42±13.94	p=0.162*
Mean school ±SD	80.29±16.04	81.58±15.31	p=0.332*
Mean FBT ±SD	30.19±8.99	29.12±9.85	p=0.118*

BMI- Body mass index, GJH-Generalized joint hypermobility, SD- Standard Deviation, HSD-Hypermobility spectrum disorders, NH-Not hypermobile, LJH-Localized joint hypermobility, VAS-Visual analogue scale, MSP-Musculoskeletal pain, DL- Daily life, PA- Physical activity, PAQC-Physical activity questionnaire for older children, PedsQL- Pediatric Quality of Life Inventory 4.0, FBT-Flamingo Balance Test, SD-Standard Deviation, *Mann Whitney U Test ** Chi-square test

Comparing Non-hypermobile, Localized Hypermobile, And Generalized Hypermobile Groups

1. Demographic parameters of groups

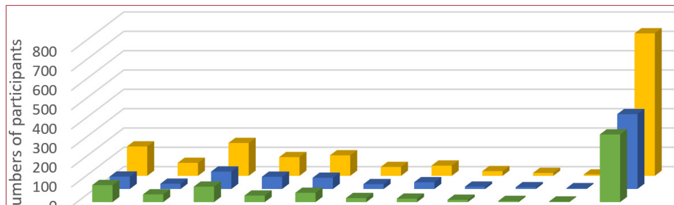
We observed significant differences in the demographic features of groups in the present study.

- There were significant differences in parameters of age, height, and weight between the groups (p=0.001, p=0.004, p=0.001, respectively) (**Table 2**).
- There was a significant difference in BMI between the three groups (p=0.006) (**Table 2**).
- The male/female proportions of groups were significantly different (p=0.008) (**Table 3**).

2. Musculoskeletal pain and injury

There were significant differences in MSP-DL and VAS-DL parameters between the groups.

- The rate of MSP-DL in the GJH group was significantly lower than the NH and LJH groups (p=0.039) (**Table 3**).
- The VAS-DL in the GJH group was significantly lower than that of the LJH group (p=0.03) (**Table 2**).
- There was no difference between the groups in the presence of MSP-PA, joint injury, and necessity of medical care (**Table 3**).



	0	1	2	3	4	5	6	7	8	9	Total
Boys	88	40	80	34	48	21	18	12	6	3	350
Girls	64	27	90	63	58	25	35	12	9	4	387
Total	152	67	170	97	106	46	53	24	15	7	737

Figure 2. The distribution of Beighton score in boys, girls and all participants

Table 2: Comparison of the demographic characteristics, pain intensity, physical activity, life quality and balance between non-hypermobile, localized hypermobile and generalized hypermobile groups

Characteristics	NH (N=152, 20.6%)	LJH (N=486, 65.9%)	GJH (N=99, 13.4%)	p Value	p Value NH-LJH	p Value NH-GJH	p Value LJH-GJH
Mean age (year) ±SD	11.57±1.21	11.54±1.3	10.98±1.35	**0.001***	0.568	**0.001**	**0.001**
Mean height (cm) ±SD	152.74±11.01	151.24±9.76	147.88±11.04	**0.004***	0.887	**0.004**	**0.011**
Mean weight (kg) ±SD	47.24±11.69	44.89±12.11	42.09±12.86	**0.001***	0.053	**0.000**	**0.041**
Mean BMI (kg/m²)±SD	20.02±3.36	19.38±3.75	18.89±3.77	**0.006***	**0.026**	**0.008**	0.568
Mean VAS-DL±SD	1.02±1.66	1.22±2.01	0.72±1.6	**0.037***	0.582	0.216	**0.030**
Mean VAS-PA±SD	1.41±2.06	1.28±2.03	1.16±1.93	0.661*	NP	NP	NP
Mean PAQ-C score ±SD	26.15±6.5	26.26±7.6	27.25±7.02	0.360*	NP	NP	NP
PedsQL 4.0							
Mean physical ±SD	84.11±12.77	82.28±15.07	84.69±13	0.276*	NP	NP	NP
Mean emotional ±SD	75.89±18.08	75.76±19.85	76.77±18.02	0.914*	NP	NP	NP
Mean social ±SD	88.0.5±15.94	90.04±14.02	90.82±14.47	0.236*	NP	NP	NP
Mean school ±SD	80.3±14.74	80.78±16.15	82.88±14.55	0.366*	NP	NP	NP
Mean FBT±SD	31.84±10.28	29.2±9.03	28.37±9.76	**0.020***	**0.033**	0.052	0.523

NH-Not hypermobile, LJH-Localized joint hypermobility, GJH-Generalize joint hypermobility, VAS-Visual analogue scale, DL- Daily life, PA- Physical activity, PAQC-Physical activity questionnaire for older children, PedsQL- Pediatric Quality of Life Inventory 4.0, FBT-Flamingo Balance Test, NP-Not performed, SD-Standard Deviation, *Kruskal Wallis Test, p< 0.05 statistically significant

Table 3: Comparison of the gender, MSP, joint injury and medical care necessity between non-hypermobile, localized hypermobile and generalized hypermobile groups

Characteristics	NH (N=152, 20.6%)	LJH (N=486, 65.9%)	GJH (N=99, 13.4%)	p Value
Sex				
Boys N (%)	**88a (57.9%)**	223b (45.9%)	39b (39.4%)	**p=0.008***
Girls N (%)	**64a (42.1%)**	263b (54.1%)	60b (60.6%)	**p=0.008***
MSP-DL N (%)	52a (34.2%)	172a (35.4%)	**22b (22.2%)**	**p=0.039***
MSP-PA N (%)	63a (41.4%)	194a (39.9%)	37a (37.4%)	p=0.812*
Pain location				
Neck N (%)	30 (19.7%)	81 (16.7%)	**6 (6.1%)**	**p=0.011***
Upper back N (%)	19 (12.5%)	52 (10.7%)	7 (7.1%)	p=0.389*
Lower back N (%)	27 (17.8%)	65 (13.4%)	9 (9.1%)	p=0.348*
Shoulders N (%)	13 (8.6%)	56 (11.5%)	6 (6.1%)	p=0.198*
Knees N (%)	11 (7.2%)	39 (8%)	8 (8.1%)	p=0.948*
Joint injury N (%)	88a (57.9%)	254a (52.3%)	53a (53.5%)	p=0.478*
Medical care N (%)	21a (13.8%)	70a (14.4%)	18a (18.2%)	p=0.584*

NH-Not hypermobile, LJH-Localized joint hypermobility, GJH-Generalize joint hypermobility, MSP-Musculoskeletal pain, DL- Daily life, PA- Physical activity, *Pearson Chi-Square, p< 0.05 statistically significant

- There was no significant difference in the location rate of MSP between the NH, LJH, and GJH groups (**Table 3**).

3. Physical activity, quality of life, and balance

   The NH, LJH, and GJH groups were compared to each other for physical activity, quality of life, and balance.

   - In the present study, no significant difference was observed in the capacity of physical activity, quality of physical, emotional, social, and school life between the groups (**Table 2**).
   - There was a significant difference in the balance parameter between the groups. The NH group had a significantly higher score in FBT (p=0.033) (**Table 2**).

Comparing Symptomatic and Non-symptomatic Generalized Hypermobile Groups

The generalized hypermobile group was subdivided, according to whether the children had MSP during DL and after PA in last month, into symptomatic and non-symptomatic. We found out no significant differences in demographic features such as age, weight, height, BMI, and gender between the subgroups in the present study.

DISCUSSION

The present study evaluated hypermobility in Turkish children and defined the frequency of joint hypermobility according to the 2017 new nomenclature of the International Consortium on EDS. It supplemented the information about the relationship between GJH and MSP, physical capacity, balance, and quality of life in healthy schoolchildren.

In this study, the GJH group was significantly younger, shorter, and thinner than the NH and LJH groups. The mean BS was significantly higher in girls than boys. However, in the GJH group, there was no significant difference between the proportion of girls and boys. The rates of MSP-DL and VAS-DL in the GJH group were significantly lower than in the NH and LJH groups. There was no significant difference between boys and girls in the VAS-DL and VAS-PA parameters but the rate of MSP in the upper back and shoulders was significantly higher in girls.

Mainly, we determined no higher MSP rate, poorer quality of life, limited physical capacity, and decreased balance ability in the GJH group than the NH and LJH groups in healthy schoolchildren between 8-15 years.

The prevalence of GJH in the present study was 13.4% in healthy schoolchildren (Cut-off≥6, Age between 8-15 years). The prevalence of GJH in other Turkish studies was between 11.7% and 18.4%. These studies evaluated hypermobility with different cut-offs in different age groups.[14-16] In Asian countries[17-20], the prevalence range was 10-65%, in Europe[21-25] the range was 7-30%, and in other countries (Australia, Egypt, Brazil) the prevalence range was 14.4-64.6%.[6,26,27] The prevalence of GJH has a broad range in children and adolescents in these studies. It seems that race, geographical location, age, study group (dancer, athletes, swimmer, rheumatology outpatient, and healthy schoolchildren or adolescents), and study design (cut-off point for BS) influence the prevalence of GJH.

The BS is a well-known, practical procedure for determining joint hypermobility. In the present study, the mean BS was 2.74±2.19. The mean BS was 2.47±2.19 in boys and 2.98±2.16 in girls, and the difference in the BS between girls and boys was significant. In an Italian study, the difference in the BS was significant between girls (median 3) and boys (median 2).[21] The mean BS in another Turkish study was 2.5.[15]

The female dominance was commonly observed in the JH studies.[6,17,20-22,25,27,28] Others reported no gender difference.[18,19,24,26,29] In our study, the difference in the BS between girls and boys was significant and the prevalence of GJH in girls was higher than boys, but this difference was not significant. Two Turkish studies showed significant female dominance and another one reported no gender difference.[14-16]

The other controversial issue in JH is the threshold of the BS for determining GJH. The cut-off point of the BS was highly variable for the diagnosis of GJH in children and adolescents. It ranged between 4 and 7 in other studies.[4] In this study, 6 was selected as the main cut-off point to define GJH according to recommendations of the International Consortium on EDS.[2]

In recent studies, GJH was defined in the same population according to different thresholds of the BS and these subgroups were compared to each other. In the present study, the prevalence of GJH was determined by 34.1% (cut-off≥4), 19.7% (cut-off≥5), and 13.4% (cut-off≥6). The results of an Italian study were 35.4% (cut-off≥4), 22.2% (cut-off≥5), and 15.1% (cut-off≥6).[21] A Danish study had higher hypermobility rates and the results were 43.2% (cut-off≥4), 27.9% (cut-off≥5), and 21.3% (cut-off≥6).[28] A study from Saudi Arabia reported the prevalence of GJH 15.2% (cut-off≥4) and 7.6% (cut-off≥6).[29] An Indian study reported 58.8% (cut-off≥4) and 44.4% (cut-off≥6)[30] An Australian study reported 48% (cut-off≥4) and 18.6 % (cut-off≥6).[6] The results of GJH in the United Kingdom were 19.2% (cut-off≥4) and 4.2% (cut-off≥6).[22] There was a great similarity between the Italian study and the present study. Both studies evaluated GJH in the same age group, and the geographical area of both studies was close.

Chronic MSP is another prominent issue that is commonly investigated in hypermobile children. The relationship was highly variable.[6,21,28,30,31] The studies that reported the association between MSP and GJH were usually cross-sectional hence the reason for MSP in hypermobility was not clear. Therefore, we still do not know why some hypermobile children were in pain or symptomatic while others were not. In addition, the studies used different methods to show a relationship between JH and MSP, such as odds ratio or comparing group tests. In the present study, hypermobile and non-hypermobile groups were compared to show an association between MSP and GJH. The participants defined the severity and locations of pain during daily life and physical activity using VAS and a body diagram. We observed no significant difference in the rate and severity of MSP between both groups. A previous study from Turkey showed a relation between GJH and MSP and another did not.[14,15] The Italian study that was similar to our study showed no association.[21-23] Some recent studies reported GJH as a risk factor for MSP.[6,28,30,31] Sohrbeck-Nohr et al.[28] stated that GJH contributed to MSP after 14 years of age. Similarly, according to prospective follow-up results of the ALSPAC cohort, the GJH group who had no significant MSP at 13.4 years showed significant association with MSP at 17.8 years.[22,31] The ages of children in our study were between 8-15 years and at these ages, the GJH group was as symptomatic as the LJH and NH groups. However, if JH continued in older ages, it would be frequently symptomatic and cause MSP.

Tobias et al.[31] reported the pain sites: the spine (lower back 16.1%, upper back 8.9%, and neck 8.6%), shoulder (9.5%), knee (8.8%), and ankle/foot (6.8%) in 17.8 years. In the present study, the locations of pain were neck (15.9%), lower back (13.7%), upper back (10.6%), shoulders (10.2%), knees (7.9%), and ankle (6.5%) in 11.5 years. The distribution of pain location in both studies was parallel, but the mean age of the study population and the relation between GJH and MSP was not identical. At an older age, we also observe the painful effect of GJH on our participants.

In this study, the overall self-reported joint injury rate was 53.6% and the distribution of joint injury was ankle (46.9%), finger (3.3%), wrist (3.1%), and knee (0.3%). There was no significant difference in joint injury between boys and girls, and HSD subgroups. Seckin et al.[14] reported the joint sprain (7.4%) as the most common injury in a Turkish study group, and there was a significant difference between boys and girls. Two studies in university students (aged between 17-26 years) from the USA reported no difference in the parameter of joint injury between hypermobile and non-hypermobile groups.[32,33]

In the present study, we observed no difference in physical activity, life quality, and static balance between the NH, LJH, and GJH groups. There was no negative impact of GJH on balance, physical capacity and, quality of physical, emotional, social, and school life. The other studies reported that there was no significant difference in physical function and capacity in the NH, LJH, and GJH groups.[21,28,34] Two studies reported poorer life quality in the hypermobile group due to stress incontinence and gastrointestinal dysfunction.[35,36]

In our study, the NH group had a higher score in FBT and poorer static balance. The LJH and GJH groups had no difference in balance. The other studies reported that the GJH group had no significant difference in dynamic balance, but had significantly better static balance.[23,37]

The main limitation of the present study was being cross-sectional in healthy schoolchildren, and it would have been informative about the follow-up of this GJH group over a longer duration, in terms of developing joint pain and other hypermobility-associated problems.

CONCLUSION

In the present study, we observed no association between MSP and GJH, and no negative influence of GJH on physical activity, life quality, and balance. However, the study population was young (mean age 11.47) and the study was cross-sectional. Prospective long-term studies are necessary to understand better the association of GJH with physical activity, life quality, joint injury, and balance. The follow-up of children with GJH in further studies may help to define the age of becoming symptomatic.

Key point

Joint hypermobility tends to be non-symptomatic at early ages because it is physiological. The well-known association between joint hypermobility and musculoskeletal pain appears in older ages, and long-standing prospective studies are necessary to define the occurring time of symptomatic hypermobility..

ETHICAL DECLARATIONS

Ethics Committee Approval: The clinical research ethics committee of Pamukkale University, Faculty of Medicine (decision date, 03/05/2016) approved this study.

Informed Consent: All patients signed the free and informed consent form.

Referee Evaluation Process: Externally peer-reviewed.

Conflict of Interest Statement: The authors have no conflicts of interest to declare.

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