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Original Research

Prevalence of hypermobility in primary school children: a Saudi experience

Abdullah M. Al-Shenqiti[1,2,*], Hatem A. Emara[1,3], Fahad S. Algarni[4], Osama A. Khaled[1,5], Asma A. Altaiyar[6], Tarek M El-gohary[1,7]

[1]Faculty of Medical Rehabilitation Sciences, Taibah University, 42353 Medina, Kingdom of Saudi Arabia
[2]Centre for Rehabilitation Science, University of Manchester, M13 9PL Manchester, UK
[3]Department of Growth and Developmental Disorders in Children and its surgery, Faculty of Physical therapy, Cairo University, 11432 Cairo, Egypt
[4]Rehabilitation Health Sciences Department, College of Applied Medical Sciences, King Saud University, 11543 Riyadh, Kingdom of Saudi Arabia
[5]Basic Science Department, Faculty of Physical therapy, Cairo University, 11432 Cairo, Egypt
[6]Medical Rehabilitation Hospital, Ministry of Health, 42316 Medina, Kingdom of Saudi Arabia
[7]Department of Biomechanics, Faculty of Physical therapy, Cairo University, 11432 Cairo, Egypt
*Correspondence: monuhama@yahoo.co.uk (Abdullah M. Al-Shenqiti)
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Abstract

Background: This study aims to determine the prevalence of generalized joint hypermobility (GJH) in primary school children in relation to age (6–12 years) and gender. It also aims to ascertain whether musculoskeletal pain (MSP) is associated with GJH among these children. **Methods**: This cross-sectional study was conducted in five primary schools in Al-Madinah al-Munawarah city. The demographic profile recorded includes age, gender, ethnic group, height, weight, and body mass index. The existence of GJH was assessed by the Beighton score ($\geq$4 was acknowledged as hypermobility). Prior to physical examinations, the Nordic Musculoskeletal Questionnaire was distributed to the students to assist in detecting symptoms in their neck, back, shoulders, and extremities. **Results**: The study included 563 students (392 boys, 171 girls; average 10.12 $\pm$ 1.588 years; range 6–12 years). GJH was found in 144 students (25.6%). Out of the total number of participants, the occurrence of GJH in male students was 30.87% versus 13.5% in female students, indicating a significant difference (p = 0.001) by gender. GJH was also more prevalent in the age group of 6–9 years (29.2%) compared to that of 10–12 years (23.7%), with a considerable significant difference of $p < 0.05$. With respect to MSP, 222 (39.4%) of the total number of students reported pain. Pain was found in 31 (21.5%) and 191 (45.6%) of hypermobile students and non-hypermobile students, respectively, with a significant difference of p = 0.029. MSP was also not associated with GJH among these children. **Conclusions**: The prevalence of GJH in the primary school children in this study was markedly higher than the range revealed in some countries in the region but somewhat within the range reported internationally.

Keywords: Hypermobility; School-age children; Beighton score; Musculoskeletal pain

1. Introduction

Joint hypermobility is defined as the excess movement of a joint's range of motion [1]. Hypermobility in multiple joints is commonly termed as generalized joint hypermobility (GJH) [2]. Ten to twenty percent of people display joint hyper mobility, which is more notable in children and adolescents [3].

GJH is a multi-factorial condition involving age and gender. Previous investigators also reported an influence of ethnic backgrounds on the possibility of the presence of GJH. Notably, GJH is highly prevalent among the populations of Asia and Africa compared to those in Western countries [4–6]. The reported prevalence of hypermobility in children varies in epidemiological studies; it has been recorded to be 8.8%–64.6% in different populations [7–12], a discrepancy largely due to the use of varying screening and diagnostic criteria.

The common consensus is that joint hypermobility is a possible cause of chronic pain and fatigue seen in at least 3% of the general population, probably owing to genetic or environmental factors [13]. However, the role of genetics in the development of chronic pain in joint hypermobility is controversial. The pain may become more evident later during adolescent years [14,15] and most commonly in the larger joints, for instance, in the back, hips, elbows, and knees. However, smaller joints such as wrists and fingers might also be affected [16].

Negligence in diagnosis and treatment may result in loss of pain control and a significant negative impact on the quality of life in these cases [17,18]. However, the frequency of musculoskeletal pain (MSP) arising from GJH in childhood is quite variable, and conflicting evidence has clearly been noted in research studies to date [19–21]. GJH identification is crucial during childhood and early adolescent years of children's lives, and identifying whether it is the cause of pain may help guide clinical practitioners and healthcare policy makers in formulating more appropriate treatment strategies and rehabilitation programs.

The current study aims to ascertain the prevalence of GJH in primary school children in relation to age (6–12 years) and gender. It also aims to find whether MSP is associated with GJH among these children.

Table 1. Personal characteristics of participants.

Characteristics	Frequency (%)/Mean (SD)
Gender	
Male	392 (69.62%)
Female	171 (30.38%)
Age (Years)	10.12 ± 1.588
Height	133.466 ± 10.122
Weight	32.492 ± 11.285
BMI	
Under weight	93 (16.52%)
Normal weight	283 (50.26%)
Overweight	91 (16.16%)
Obese	96 (17.05%)
Hand Dominance	
Right	519 (92.18%)
Left	44 (7.81%)

2. Material and methods

The study population included 563 school-aged children (392 boys and 171 girls) from five different educational institutions (primary schools) chosen randomly. The population sample had a specific attribute: the sample consisted of a specific age group (6–12 years). The participants, all of Saudi nationality, were from Al-Madinah al-Munawwarah city, Saudi Arabia.

Any student with neurological deficits and/or acute trauma that may influence joint mobility was excluded from the study. Students who had been diagnosed previously with an inflammatory musculoskeletal disease or connective tissue disorders were also excluded.

The students underwent full medical history and physical examination. Demographic data including age, gender, race, height, weight, and body mass index was recorded. Data collection was conducted from March to May 2019. The degree of GJH was determined using the Beighton score, a valid measure scored on a scale between 0 and 9. Children with a score of 4 or higher were considered to have GJH [13,22].

2.1 Steps for performing Beighton score for screening GJH

The screening standards include five tests; participants received 0 or 1 depending on their capability to accomplish each test. Results have been summed with totals range from 0 to 9.

Joints were considered hypermobile when the examiner could extend the 5th metacarpophalangeal joint passively to greater than 90; elbow hyperextension $>$10; thumb-to-forearm opposition and knee hyperextension $>$10; and if the participant was able to perform trunk flexion with palms resting easily on the ground with fully extended knees. All examinations were executed bilaterally except for trunk flexion, which followed the guidelines of Norkin and White [23] and the steps recommended by Boyle *et al.* [24–26]. Physical examinations were carried out by two qualified physical therapists previously trained in a specialized medical rehabilitation center in pediatric assessments. A universal goniometer was utilized to measure the range of motion of the joints. The intra-rater reliability of the examiners was assessed prior to the data collection in the study.

2.2 Musculoskeletal pain assessment

The Nordic Musculoskeletal Questionnaire (NMQ) which is considered valid in exploring neck, back, shoulders, and the limbs' problems [27] was used in the study. Participants were asked about pain within last 12 months in the cervical region, shoulders, elbows, hands/wrists, upper back, lower back, and hip/thigh. The body chart of the NMQ was included in the questionnaire to increase explanation of the questionnaire [28].

2.3 Statistical analyses

The data was recorded in a Microsoft Excel sheet and analyzed using the Statistical Package for the Social Sciences (SPSS) for Windows, version 22 (SPSS, Inc., Chicago, IL, USA). Histograms and the Kolmogorov–Smirnov test were used to assess the normality and the symmetry of data. Descriptive statistics were used for personal characteristics; the prevalence of hypermobility was measured by the Beighton scale and MSP was measured by the NMQ. Categorical data from questions were used as percentage of the total participant pool, or a portion of this pool, while means with standard deviations were used for quantitative data.

The prevalence of GJH was obtained by dividing the number of participants with GJH (Beighton cutoff score of $\geq$4) by the whole number of participants. Chi-square statistic and the Phi coefficient test were used to calculate the relation between binary variables and whether GJH exists or not. Moreover, the Chi square test of independence was computed to determine if there is a significant relationship between GJH and MSP. Significant difference was set at $p < 0.05$.

3. Results

Table 1 presents subjects' characteristics. The study comprised 563 primary school students (392 boys, 171 girls; average age 10.12 ± 1.588 years; range 6–12 years). GJH was found in 144 students (25.6%) (Table 2). Out of all participants, the frequency of GJH in male students was 30.87% while in female students it was 13.5%, indicating a significant difference (p = 0.001) in terms of gender. GJH was also detected more obviously in the age group of 6–9 years (29.2%) compared to that of 10–12 years (23.7%), with a considerable significant difference (p = 0.000) (Table 3).

Table 2. Generalized joint hypermobility and gender.

Characteristic		Gender		Total count (%)	p value
		Boys count (%)	Girls count (%)		
Hypermobility	Yes	121 (21.5%)	23 (4.1%)	144 (25.6%)	0.001
	No	271 (48.1%)	148 (26.3%)	419 (74.4%)	0.002
	Total	392 (69.6%)	171 (30.4%)	563 (100.0%)	

Table 3. Generalized joint hypermobility and age groups.

Characteristic				Hypermobility (Frequency/%)
Age				
Age group	Number	(Mean ± SD)	(Median)	
6–9	195	7.53 ± 0.68	7	57 (29.2%)
10–12	368	10.72 ± 0.71	11	87 (23.6%)
Total	563	9.46 ± 0.71	10	144
p value				0.000

Table 4. Prevalence of joint hypermobility at various sites used in the Beighton criteria (Cut-off ≥4).

Region	Total N = 563	Male N = 392	Female N = 171	Chi square (χ^2)	p value
	Frequency (%)	Frequency (%)	Frequency (%)		
Right thumb	42 (7.46%)	35 (8.92%)	7 (4.09%)	4.020	0.045
Left thumb	43 (7.63%)	36 (9.18%)	7 (4.09%)	4.367	0.036
Right little finger	25 (4.44%)	21 (5.36%)	4 (2.34%)	3.940	0.047
Left little finger	24 (4.26%)	20 (5.10%)	4 (2.34%)	2.219	0.136
Right elbow	51 (9.06%)	41 (10.46%)	10 (5.84%)	3.008	0.079
Left elbow	54 (9.59%)	44 (11.42%)	10 (5.84%)	4.215	0.401
Right knee	18 (3.19%)	17 (4.37%)	1 (0.58%)	5.480	0.0192
Left knee	12 (2.13%)	12 (3.06%)	0 (0.00%)	5.337	0.0209
Trunk	4 (0.71%)	3 (0.76%)	1 (0.58%)	0.055	0.815

Table 5. Prevalence of musculoskeletal symptoms in each body location.

Region	Pain last 12 months	
	Male total (392)	Female total (171)
Neck	17 (4.34%)	12 (7.02%)
Shoulders	14 (3.57%)	11 (6.43%)
Elbows	19 (4.85%)	14 (8.19%)
Hands/wrist	17 ((4.34%)	11 (6.43%)
Upper back	15 (3.83%)	13 (7.60%)
Lower back	19 (4.85%)	14 (8.19%)
Hip/thigh	21 (5.36%)	5 (2.92%)
Knees	16 (4.08%)	4 (2.34%)
Total	138	84

The subjects' joint hypermobility distribution is showed in Table 4. The elbows' hypermobility was (9.59%), thumbs (7.63%), little fingers (4.44%), knees (3.19%), and the trunk (0.71%). The ratio of boys with hypermobility of the right thumb, the left thumb, the right little finger, the right knee, and the left knee was statistically higher than girls ($p < 0.05$). The percentage of hypermobile participants did not differ by gender in the left little finger, the right elbow, and the left elbow. Unusually, none of the female participants had hypermobility in the left knee.

Table 5 shows the prevalence of MSP in each body location. For males, pain was primarily reported on the hip and thigh while the lowest prevalence was reported for the shoulders. For females, pain was principally reported on the elbows and lower back while the lowest prevalence was reported for the knees.

3.1 Correlation between GJH and musculoskeletal pain

Regarding MSP, 222 (39.4%) of the total number of primary school students reported pain. Pain was also evident in 31 (21.5%) and 191 (45.6%) of hypermobile children and non-hypermobile children, respectively, with a significant difference ($p = 0.029$). Children without hypermobility were more likely to have chronic MSP.

MSP was demonstrated with a higher proportion of 29.9% in the hypermobile students in the 10–12 years age group compared to 8.8% in that of 6–9 years with a significant difference ($p = 0.003$), whereas MSP was present in the non-hypermobile students with a proportion of 47.8% in the age group of 6–9 years compared to 44.5% in that of 10–12 years, with no significant difference ($p = 0.533$) (Table 6). Based on the results of the Phi coefficient test, there was no association between hypermobility and chronic MSP (Ø = 0.024, $p = 0.571$).

Table 6. Musculoskeletal pain and hypermobility including age groups.

		6–9	10–12	Total	p value
Hypermobility	Pain	5 (8.8%)	26 (29.9%)	31 (21.5%)	
	No Pain	52 (91.2%)	61 (70.1%)	113 (78.5%)	0.003
	Total	57	87	144	
No hypermobility	Pain	66 (47.8%)	125 (44.5%)	191 (45.6%)	
	No Pain	72 (52.2%)	156 (55.5%)	228 (54.4%)	0.533
	Total	138	281	419	
Sum of All		195	368	563	

4. Discussion

This study exhibited a relatively high prevalence of GJH (25.6%) in primary school students aged 6–12 years, utilizing a Beighton score (cut-off ≥4/9). The study also showed that MSP was substantially higher in children who were not associated with GJH with a proportion of 45%, compared to those with GJH with a proportion of 21.5%, with a difference of $p = 0.029$.

The prevalence of GJH as shown in this study was higher than that reported in studies with relatively similar designs from different countries, including Egypt (16.1%), Turkey (11.7%), the Netherlands (15.5%), the United Kingdom (19.2%), and Lithuania (19.2%) [29–33]. However, the United Kingdom (4.2%) and Lithuania (5.7%) showed an even lower prevalence when they used a higher Beighton score cut-off (≥6/9) in their assessments [32,33]. Conversely, a considerably high prevalence was clearly shown in Brazil (64%) and India (58.7%) [34,35]. However, these studies included either a lower (4–7 years) or a wider range (3–19 years) of age groups in their adopted criteria compared to the current study.

The Beighton score has been utilized globally to determine GJH in different age groups, and studies have frequently used different cutoffs, ranging from ≥4/9 to ≥6/9 [25]. This may also explain the substantially wide variable findings of the prevalence found in this study compared to that from different parts of the globe. Of the total participants, the current study showed a higher frequency of GJH in boys (30.87%) compared to girls (13.5%). This was also reflected in the results of the statistical analysis ($p = 0.002$).

A study with a comparative design to the present study by Qureshi *et al*. [36] revealed similar observations in Pakistan where a higher frequency of GJH was observed in boys (39.5%) compared to girls (34.2%), whereas some studies in different countries show a higher occurrence of GJH in girls, including in the Middle East, Europe, and the USA [2,31,37]. However, no significant differences between boys and girls were reported in these studies.

With respect to age, despite the narrow range between the age groups of the students in this study (group of 6–9 years and group of 10–12 years), there was a considerable, significant decline in GJH with increase in age ($p = 0.000$), while the frequency of GJH also diminished from 29.2% to 23.7%. Similar observations were also noted in a comparative Dutch study that showed a trend of GJH decline associated with the increase in age but with smaller proportions (15.5% to 13%) [31], and more prominently in a Greek study [7] with proportions of 11% in the younger age group compared to 7.6% in the older age group supplemented by a significant difference of $p = 0.0018$. However, these two studies included students with a wider range of age groups (4–13 years versus 12–17 years and 5–14 years).

With regard to MSP, there is an increasing amount of controversial evidence of the presence of MSP among individuals with GJH. The current study did not show an association between GJH and MSP where a Beighton score with a cut-off point of ≥4 was adopted. Similarly, no association between GJH and MSP was found when a slightly higher Beighton score cut-off (>5) was utilized in a cross-sectional study conducted by Leone *et al*. [38] for school children aged 7–15 years. Conversely, a study by Hasija *et al.* [35]. that adopted the Beighton score with a cut-off point of ≥4 in assessing hypermobility but including wider age groups (3–19 years old) showed MSP was more prevalent in children associated with GJH with a proportion of 26%, compared to those who did not have GJH with a proportion of 17.2%, with a significant difference ($p < 0.05$) [35].

In a recent study by Al-Jarallah *et al*. [39], the occurrence of MSP was found to be more frequent in hypermobile students with Beighton scores of 4–5 than those with hypermobility assessed with scores of 6–9 [39]. This may give us a justification for the variable findings in the current study in comparison to other studies. MSP was shown to be associated with GJH in childhood and, more frequently, during adolescence. For example, findings of some research studies showed causal associations between chronic and recurrent back pain and GJH [19,40]. The complaints may become more prominent from 10 years of age and older [14,15]. This might be due to the natural demand for increasing physical and sport activities associated with increasing age during the adolescent period, and it also explains the findings of the current study, which shows an increase in MSP in the older age group of the children associated with GJH. Therefore, a better understanding of these complaints is substantially valuable not only for providing effective preventative strategies to adults, but also for a better understanding of the origin of MSP.

Identifying the prevalence of GJH and the MSP associated with it is highly crucial and necessary for countries to plan favorable management strategies and rehabilitation programs. However, it remains unclear why some children associated with GJH become symptomatic while others remain symptom-free. The limitations of this study comprise its cross-sectional nature, where the analyses were restricted to the frequency of GJH associated with MSP in primary school children (aged 6–12 years), without considering the underlying cause for regional or widespread MSP in this particular age group.

5. Conclusions

The GJH prevalence in primary school students in this study is within the range reported from different parts of the world. The current study showed a remarkably decline of GJH with increase of age and a higher frequency of GJH in boys more than girls.

MSP was not more evident in children with GJH, but an increase in MSP was clearly shown in the older age group of children associated with GJH rather than the younger age group. Therefore, rigorous, controlled studies considering the underlying cause of MSP in GJH children are essential.

Author contributions

Study conceptualization, design and analysis of data were done by AMA and HAE. HAE, AAA and OAK collected data Interpretation of data was carried out by HAE, FSA and AMA and OAK. AMA, HAE and OAK conducted organization procedures, supervision and prepared the initial version of the manuscript. Final version of the manuscript was reviewed by TME and AMA, FSA and OAK. Similarity check was done by all authors using the software of authenticate.

Ethics approval and consent to participate

The study was approved by the ethics committee at the college of Medical Rehabilitation Sciences, Taibah University, in accordance with the declaration of Helsinki (CMR-PT-2019-010). Informed consent was obtained from and signed by legal guardians.

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Conflict of interest

The authors declare no conflict of interest.

Data availability statement

The datasets generated during and/or analysed during the current study are not publicly available but are available from the corresponding author on reasonable request.

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