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Prevalence of generalised joint hypermobility in school-aged children from east-central European region

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Background: *There is no literature regarding joint mobility in children of the Central and Eastern Europe. Studies describing clinical characteristics and functional outcomes are still needed. The aim of this study was to assess the prevalence of generalised joint hypermobility (GJH) in the group of school-aged children from Vilnius, the capital city of Lithuania, in relation to different cut-off values of the Beighton score (BS), and to identify possible patients with joint hypermobility syndrome.*

Materials and methods: *The representative sample of this study was calculated to be 760 subjects. A total of 778 children from different schools were screened for the mobility of joints. The medical examination included an assessment of joints' hypermobility according to the BS. The presence of specific signs (marfanoid habitus, antimongoloid slant and drooping eyelids) was assessed additionally. Parents of all involved children were asked to answer the questions developed based on the Brighton criteria regarding the medical history of children.*

Results: *The prevalence of GJH in school-aged children from Vilnius, depending on the BS cut-off value, was 19.2% (BS ≥ 4), 9.5% (BS ≥ 5) or 5.7% (BS ≥ 6). The increased range of mobility was most frequently detected in thumbs of school-aged children. The frequency of hyperextension > 10º in knees was 7- to 8-fold lower than the frequency of hyperextension > 10º in a passive opposition of the thumb. The evaluation results were similar on the left and right sides in 87.4% cases of thumb opposition, 90.1% cases of hyperextension of 5th finger, 87.9% cases of elbow manoeuvres, and 94.8% attempts to hyperextend knee.*

Conclusions: *The prevalence of GJH in school-aged children from Vilnius depends on the BS cut-off value and ranges from 5.7% to 19.2%.* (Folia Morphol 2016; 75, 1: 48–52)

Key words: generalised joint hypermobility, prevalence, school-aged children, Beighton score, Brighton criteria

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INTRODUCTION

The decreased range of joint motion is often presented in different joint diseases and periarticular damages. Therefore, an increased joint mobility or hypermobility is still associated with some clinically irrelevant casuistry for the part of medical professionals. Despite the fact of the simple palpability and detectability, a generalised joint hypermobility (GJH) was recognised as a medical pathology only in 1967 [10]. Chronic pain related to joint hypermobility was was considered as one of the 5 most common conditions of chronic pain in paediatric rheumatology [4]. In these cases, when GJH is associated with musculoskeletal symptoms in the absence of demonstrable systemic rheumatologic disease, a definition of joint hypermobility syndrome (JHS) is used [6].

The studies of populations show that GJH is observed more often in children than in adults. The prevalence of GJH was studied in different societies and populations. It ranges from 2% to 71% [3, 7, 8, 11, 14, 16–19]. Age, sex, and ethnic subjection have influenced the prevalence of GJH in different societies [2, 3, 7, 13, 15].

The Beighton score (BS) is a widely accepted tool to identify GJH. The cut-off value of this score is ≥ 4, as recommended by the British Society of Rheumatology [17]. Murray noticed that such cut-off point of BS was extrapolated from the populations of adults into paediatric population and the high prevalence of GJH could be the consequence of such unreasoned extrapolation [14]. As it has been showed by Juul-Kristensen et al. [9], the significant reduction in prevalence of GJH has occurred when different cut-off values of BS have been used.

In 2000 the revised Brighton criteria of JHS diagnosis were introduced [6]. According to the Brighton criteria, the different combinations of major and minor signs can result in the diagnosis of JHS. Theoretically, BS, used as single criterion, could be sufficient for the diagnosis of JHS in the presence of other small symptoms. Taking into consideration the increasing numbers of people with hypermobility and chronic joint pain [4], the simultaneous use of the BS and Brighton criteria could be beneficial in screening for patients with joint hypermobility.

Studies describing clinical characteristics and functional outcomes are still needed [5, 14]. The prevalence of GJH in Lithuania could be interesting as a data from the relatively homogenous population of Caucasians. On the other hand, there is no literature regarding joint mobility in children in the Central and Eastern Europe.

The aim of this study was to assess the prevalence of GJH in the group of school-aged children from Vilnius, the capital city of Lithuania, in relation to different cut-off values of the BS, and to identify possible patients with JHS.

MATERIALS AND METHODS

Study design and population

The study took place in Vilnius, the capital city of Lithuania. There were 78,000 children in Vilnius schools grading from 5 to 12 classes at the moment of the study. The population of Vilnius is dominantly Caucasians with a rate of immigrants less than 1%.

The representative sample of this study was calculated to be 760 subjects. The schools were selected randomly using the list of institutions for secondary education. All headmasters of the selected institutions were visited to explain the goals and circumstances of the study, to get the agreement of participation and to clarify how long the data will be collected. Parents of potential participants were informed by providing an explanation letter in which they were asked if they agree that their children will be involved in the study and will answer several questions associated with Brighton criteria. The testing of Beighton manoeuvres was organised during the lessons of physical culture.

The study was approved by the Child Right Service Committee of Vilnius. The study was approved by the Ethical Committee of Vilnius University.

Measures

We have screened the joint mobility of 778 children aged from 10 to 18 years using the widely accepted Beighton method which helps to examine range of motion of the knees, trunk, fingers, thumbs, and elbows bilaterally and employs a 0 to 9 scoring scheme.

Additionally, the presence of specific signs (marfanoid habitus, antimongoloid slant and drooping eyelids) was assessed at the same time during the study.

Parents of all involved children were asked to answer the questions developed based on of the Brighton criteria regarding the medical history of children.

Statistical analysis

All analyses were conducted using SPSS for Windows, version 19.0. The results are shown as num-

Table 1. Demographic and anthropometric profile of participants

	N	Minimum	Maximum	Mean	Standard deviation
Age [years]	778	10	18	14.02	2.11
Height [cm]	778	130	197	167.55	12.23
Weight [kg]	778	24	120	56.42	12.77
Body mass index	778	13.17	35.44	19.8801	2.861

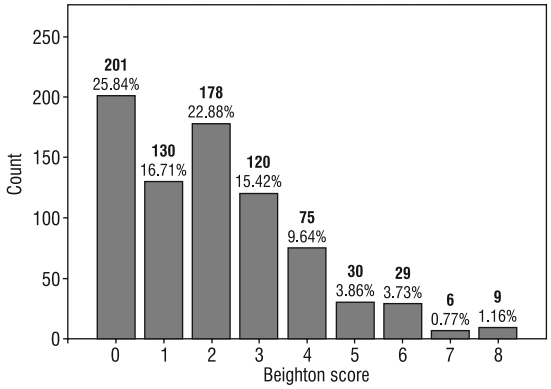

Figure 1. Absolute and percentile distribution of the total Beighton score of school-aged children.

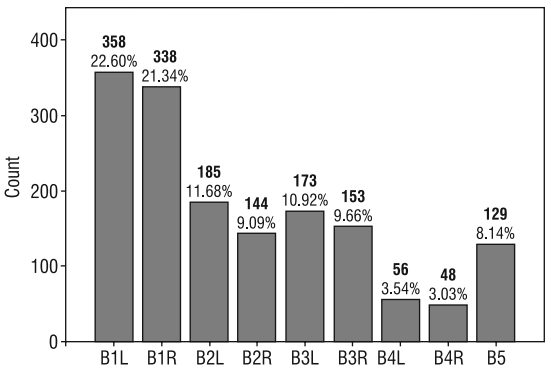

Figure 2. Prevalence of the Beighton manoeuvres in the study sample; B1 — passive abduction of thumb; B2 — passive hyperextension of fifth finger; B3 — hyperextension of elbow; B4 — hyperextension of knee; B5 — lumbar flexion; L — left; R — right.

bers and percentages for categorical variables. In the statistical analysis the Mann-Whitney test and χ^2 test were applied. Statistical significance was set at 0.05 for all tests.

RESULTS

The anthropometrical characteristics of the study sample are presented in Table 1. The detailed distribution of the BS is presented in Figure 1.

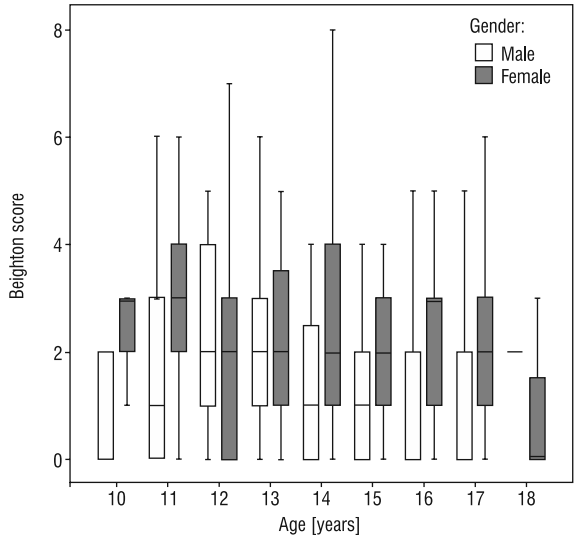

Figure 3. Impact of age and gender on the Beighton score of school-aged children (horizontal lines within bars depict median and vertical lines outside the box display the non-outlier range).

As showed in Figure 1, the prevalence of joint hypermobility in school-aged children from Vilnius, depending on the BS cut-off value, was 19.2% (BS ≥ 4), 9.5% (BS ≥ 5) or 5.7% (BS ≥ 6). The significance of found proportions depending on cut-off value was confirmed by using χ^2 test ($p < 0.001$).

The frequencies of positive responses based on separate tested manoeuvre are shown in Figure 2. As showed in Figure 2, the increased range of mobility was most frequently detected in the thumbs of studied school-aged children. The frequency of hyperextension > 10° in the knees was 7- to 8-fold lower than the frequency of hyperextension > 10° in the passive opposition of the thumb.

The median of BS, depending on the age and gender, is showed in Figure 3.

Median value of BS was never higher for boys, if the groups of marginal ages with the scarce number of individuals were not included in comparison. The total BS was higher in girls ($Z = -6.05$, $p < 0.001$). Table 2 contains the data regarding the differences in

Table 2. Differences in separate manoeuvres of Beighton score due to gender

	B1L	B1R	B2L	B2R	B3L	B3R	B4L	B4R	B5
Mann-Whitney U[a]	67417	63288	74796	74468	66276	67593	72405	75188	62327
Z	–3.007	–4.560	–0.326	–0.513	–4.110	–3.690	–2.242	–0.276	–6.553
p	0.002	< 0.001	0.745	0.608	< 0.001	< 0.001	0.02	0.782	< 0.001

[a]Grouping Variable: gender. B1 — passive abduction of thumb; B2 — passive hyperextension of fifth finger; B3 — hyperextension of elbow; B4 — hyperextension of knee; B5 — lumbar flexion; L — left; R — right

each tested manoeuvres between genders. An evident unidirectional tendency is presented in Table 2.

The scores were higher in girls than in boys for all manoeuvres (the differences between genders were statistically significant). Lumbar hypermobility was 3 times more often in girls than in boys.

Both sides, left and right, were evaluated as symmetrical in 87.4% cases of thumb opposition, 90.1% cases of hyperextension of 5th finger, 87.9% cases of elbow manoeuvres, and 94.8% attempts to hyperextend the knee.

The groups of subjects with and without hypermobility were compared according to their age, body mass, and height as well as body mass index (BMI). Independent samples Mann-Whitney U test revealed significant differences in age ($p = 0.016$), body mass ($p < 0.001$), height ($p = 0.001$) and BMI ($p < 0.001$) between the groups with and without hypermobility.

There were no subjects who answered positively to at least two questions which were based on Brighton criteria. This means that none of the examined children had JHS.

DISCUSSION

This was the first study researching the prevalence of GJH in school-aged children from Lithuania. No studies have been performed to assess the prevalence of GJH in the population of the Central and Eastern Europe. This study has confirmed the opinion that the prevalence of GJH depends on the cut-off value of the BS [17].

The wide range of prevalence estimates may be attributed to methodological differences across studies as well as actual differences in the prevalence of GJH between countries. In general, the result of this study is in accordance with the previous studies on Western populations [3, 8, 11, 16]. The fact that pupils without joint hypermobility were heavier, taller and older could be interpreted as the confirmation of hypothesis that the phenomenon of hypermobility disappears with age. Despite numerous investigations in this area of rheumatology, a direct comparison between studies is complicated due to different anthropometrical properties of involved subjects. The significant influence of age on the prevalence of GJH is remarked in majority of studies [7, 8, 11]. The range of age in current study was 10–18 years. Rikken-Bultman et al. [18] found 13.4% subject with hypermobility in Dutch school children aged 12–17 years. Seçkin et al. [19] noted the prevalence of GJH 11.7% in Turkish population of 13–19 years age. The range of age in both of these studies is very similar to the range of age in the current study. So, the prevalence of GJH in Vilnius is higher than in both above mentioned studies.

Analysis of the prevalence of separate Beighton manoeuvres in the current study revealed some tendencies. As showed in other studies too [3, 7, 16], the highest prevalence is peculiarity of palm signs. The ability to hyperextend knee can be described as the rarest kind of hypermobility in school-aged children from Vilnius. Majority of children who were able to reach the floor without bending the knees were involved in regular activities such as dancing or sports training. Trainability of this manoeuvre was suggested and remarked in other studies as well [8, 15, 16]. Dancing or sport training may maintain the presence of hypermobility or promote hypermobility through forced hyperextension [3]. So, our study supports the results from previous studies revealing that children who have a higher range of joint movement may be involved in sport and music activities.

Consistent with the findings of previous studies [3, 12], our study revealed the little evidence for laterality of hypermobility in school-aged children.

In current study, the largest difference in median BS between genders was found in children aged 11 years. The most variable data were collected from 14-year-old girls. Non-outlier range in this subgroup covers values of BS from 0 to 8, as is showed in Figure 3.

It could be associated with various phases of pubertal process in this age group. Temporary elevation of BS value at age of 15 years in girls was explained similarly in the study of Jansson et al. [8].

No cases of JHS were revealed using questionnaire developed based on Brighton criteria and addressed to the parents of investigated children. Besides low response rate, a couple other reasons could explain it. Testing of the BS was performed in school's environment. As it has been noted in the study of Adib et al. [1], half of JHS patients have been missing school. It could hide some part of those patients. The conflicting results regarding the prevalence of musculoskeletal pain were noted in subjects with and without hypermobility by several groups of researchers [6, 10, 20]. Recent study of Leone et al. [11] investigated the occurrence and frequency but not the duration of musculoskeletal complains.

CONCLUSIONS

Due to low response rate, the question regarding musculoskeletal pain revealed only a few of these persons in the current study. Parents were asked only about pain longer than 3 months. It could be an explanation of so low number of positive cases.

The prevalence of GJH in school-aged children from Vilnius depends on the BS score cut-off value and ranges from 5.7% to 19.2%.

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