



# Joint hypermobility in school-aged children and adolescents with idiopathic scoliosis – A chance for more accurate screening?

Barbara Katarzyna Dobies-Krześniak<sup>1,2,A-F</sup>, Agnieszka Werblińska<sup>1,B,E-F</sup>, Beata Tarnacka<sup>1,2,A,C,E-F</sup>

<sup>1</sup> Rehabilitation Clinic, Medical University of Warsaw, Warsaw, Poland

<sup>2</sup> Rehabilitation Clinic, National Institute of Geriatrics, Rheumatology and Rehabilitation, Warsaw, Poland

A – Research concept and design, B – Collection and/or assembly of data, C – Data analysis and interpretation, D – Writing the article, E – Critical revision of the article, F – Final approval of the article

Dobies-Krześniak BK, Werblińska A, Tarnacka B. Joint hypermobility in school-aged children and adolescents with idiopathic scoliosis: A chance for more accurate screening? *Ann Agric Environ Med*. doi: 10.26444/aaem/150010

## Abstract

**Introduction and Objective.** Joint hypermobility (JH) can be diagnosed in the case of increased range of joint mobility and the absence of systemic diseases. Lack of stability of the spinal joints is mentioned as one of the possible causes of scoliosis. Simplicity of hypermobility examination could help identify children at higher risk of developing scoliosis. The aim of this study was to assess JH prevalence in children with idiopathic scoliosis and to analyse coincidence of joint hypermobility, scoliosis features, age, gender and bone maturity.

**Materials and method.** The case-control study enrolled 125 children aged 7–18 years (mean 13.2±2; Cobb angle range 10°–53°; mean 24.3±11.7) diagnosed with idiopathic scoliosis. The control group included 83 volunteers. The Beighton scale was used to determine joint hypermobility. The relationship between joint hypermobility and scoliosis was tested. Secondly, hypermobility prevalence according to age, gender, curve severity, number of curvatures and Risser sign in scoliosis group was summarised. Data were compared by Student's t-test, U Mann-Whitney, chi-square test or Fisher's exact test where appropriate. P < 0.05 was considered statistically significant.

**Results.** JH was diagnosed in 64 (51.2%) scoliotic patients and in 34 (41%) control group children. The difference found was not statistically significant (p = 0.148). No significant difference was found comparing scoliosis subgroups with curve size cut-off point from 20 degrees, single/double curve scoliosis, male/female gender, age cut-off point 13,2 years of age, and Risser test score 0–2/3–5.

**Conclusions.** This study shows that children with JH features do not have a statistically significant increased risk of scoliosis co-diagnosis.

## Key words

joint instability, joint laxity, Beighton score, posture, spinal curvatures

## INTRODUCTION AND OBJECTIVE

Joint hypermobility (JH) should not be considered as a medical problem. It is a condition in which joint range of motion exceeds normal for age, gender, and ethnic background in the absence of any systemic disease [1]. To emphasize the difference, the condition in which the musculoskeletal and non-musculoskeletal complaints without systemic disease occur is known as joint hypermobility syndrome (JHS) [2]. The reported prevalence of JH varies between 2%–57% depending on age, gender, ethnicity, and race. It is more common in young women and of Asian descent. In Europe, the prevalence of JH in children fluctuates around 15–20% [3].

JH is commonly diagnosed with the Beighton score that evaluates the ability to perform a selection of joint manoeuvres [4], with a maximum total of 9 points. A value of 4 or more is typically considered positive for JH. Researchers studying JH confirm, that the Beighton score is a valid instrument to be used in children [5]. JH occurrence correlates with articular pain (mainly knee, foot and spine), postural disorders and

planovalgus feet. Previous studies found that JH was more frequent in children and adolescents with idiopathic scoliosis [6, 7].

Scoliosis is defined as 3-dimensional deviation of the spine (lateral with Cobb angle of 10 degrees or higher) and axial rotation [8, 9]. Scoliosis can be defined as secondary (when associated with known medical problem) or idiopathic (without defined cause in other-wise healthy children) [10]. Prevalence of idiopathic scoliosis (IS) have a wide range of 0.93–12%, with value of 2–3% as most often repeated [10]. It is more common in girls (except for infantile idiopathic scoliosis) with the ratio increasing from 1.4:1 with Cobb angle between 10°–20° to 7.2:1 for above 30° [11, 12]. In retrospective studies, Fadzan et al. summarized that the prevalence of scoliosis is higher in adolescents participating in sports activities, such as dancing, ballet, swimming, tennis, table tennis, hurling, javelin, volleyball, and gymnastics [13].

Understanding of IS etiopathogenesis is still limited. There is a pattern of familial occurrence of IS suggesting genetic etiology. Researchers noted that IS may be connected to disorder of estrogen receptor structure and function, mucopolysaccharide and lipoprotein synthesis, melatonin synthesis and co-existent disorders of blood platelet and collagen function. Latest studies point for calmodulin as a protein connecting IS, melatonin level and platelet's function.

Address for correspondence: Barbara Katarzyna Dobies-Krześniak, Rehabilitation Clinic, Medical University, Spartańska 1, 02-637 Warsaw, Poland  
E-mail: bdobies@wum.edu.pl

Received: 23.02.2022; accepted: 11.05.2022; first published: 11.07.2022

Other researchers suggest that MMP-3 and IL-6 promoter polymorphisms or increased BNC2 expression could possibly be genetic markers for IS [10].

There are many risk factors that may predispose to spinal instability. Joint hypermobility, delayed puberty, asymmetric spinal load, placing the thoracic spine in a lordotic position, growth-related factors (greater body height and growth velocity of more than 2 cm per year), low body mass index and abnormal levels of leptin are also suspected to play a role in scoliosis development [13]. Finding the association of JH with the development of IS is an important part of medical care for children and adolescents especially from rural regions where diagnostic imaging and therapeutic intervention may be delayed due to reduced accessibility [14, 15]. The diagnosis of joint hypermobility during a simple physical examination could identify children at higher risk of developing scoliosis.

## MATERIALS AND METHOD

Patients were recruited in the Orthopaedic and Rehabilitation Centre for Children and Youth in Konstancin-Jeziorna, Mazowiecki Province, Poland, from 01.2018–07.2019. Admitted patients were pre-screened to generate lists of potential study subjects. The patients and their legal guardians were then approached and both provided informed written consent. Inclusion criteria: age 7–18; IS diagnosis based on anteroposterior radiogram with Cobb angle  $\geq 10^\circ$  and vertebral rotation, exclusion criteria: secondary scoliosis, connective tissue disease in medical interview. Control group included children and adolescence volunteers examined from April–June 2019. To minimize confounding of gender and age, which are known to affect the Beighton score, the group was matched by average age and gender ratio. Inclusion criteria: age range 7–18, exclusion criteria: previously diagnosed incorrect posture (irregularities in the shape of the spine with suggestion of a dedicated therapeutic intervention) and connective tissue disorder in medical interview. The following information was collected from the medical records: medical history, age and gender. Maximum lateral spinal curve size was measured by BDK (intra-rater reliability 0,941 with  $2^\circ$  error range) using the Cobb method [16]. Number of curves and the US Risser sign [17] were also determined on anteroposterior radiograph of whole spine. Classification of IS depending on angle of lateral deviation used in this study was excerpted from SOSORT guidelines and adopted (Tab. 1) [10].

Joint mobility was examined by BDK using the Beighton score (between 0–9) via the Beighton Scale (Tab. 2) [4]. Transparent plastic goniometer (Saehan Plastic Goniometer 08–030111, Masan, Korea) was used to assess knees and elbow hyperextension  $>10^\circ$ . The side of the body on which the bilateral measurements were made first was selected at random. Patient with result of 4 or more was identified as hypermobile.

**Statistical analysis.** Continuous data were presented as a mean value and standard deviation (SD). Categorical data were presented as a percentage. Normal distribution was verified by Kolmogorov-Smirnov test. Continuous data were compared by Student's t-test or U Mann-Whitney test where appropriate. Categorical data were compared by chi-

**Table 1.** Classification of scoliosis depending on angular deviation (Cobb)

Original classification		Adaptation	
Low	$\leq 20^\circ$	Low	$\leq 20^\circ$
Moderate	$21^\circ$ – $35^\circ$	Moderate	$21^\circ$ – $35^\circ$
Moderate to severe	$36^\circ$ – $40^\circ$	Severe	$36^\circ$ – $50^\circ$
Severe	$41^\circ$ – $50^\circ$		
Severe to very severe	$51^\circ$ – $55^\circ$	Very severe	$\geq 51^\circ$
Very severe	$\geq 56^\circ$		

**Table 2.** The Beighton score

The Beighton Score	R	L
	Passive dorsiflexion of the little finger beyond $90^\circ$	1
Passive apposition of the thumb to the flexor aspect of the forearm	1	1
Hyperextension of the elbow beyond $10^\circ$	1	1
Hyperextension of the knee beyond $10^\circ$	1	1
Forward flexion of the trunk, with knees straight, so that the palms of the hands rested easily on the floor	1	
<b>Total</b>	0–9	

square test and Fisher's exact test. P value less than 0.05 was considered statistically significant. Data were analyzed using IBM SPSS Statistics for Windows, v21 (IBM Corp., Armonk, NY, USA)

## RESULTS

**Basic statistics of the studied groups.** The study group consisted of 125 patients with idiopathic scoliosis and the control group of 83 children and adolescents [16]. Girls statistically more often showed hypermobility in all examined cases ( $p=0.01$ ) and the control group alone ( $p=0.01$ ), but not

**Table 3.** Basic parameters of the study and control groups

	Study group (n=125)	Control group (n=83)	p value
Age (years)	$13.2 \pm 2.0$	$13.2 \pm 3.0$	0.750
Male gender (%)	33 (26.4)	22 (26.5)	0.986

in the study group ( $p=0.24$ ). Basic parameters comparing both groups are shown in Table 3.

Cobb angle range was  $10^\circ$ – $61^\circ$ , mean  $21.85^\circ \pm 11.99$ . The quantitative division due to the angular size of the IS curvature is typical and presented in Figure 1.

**Comparative analysis of hypermobility in the study group and control group.** The differences found between patients and controls for JH were not statistically significant considering all used cut-off points.

Individual comparison of almost all tested manoeuvres in the groups did not show statistical significance. The only exception to this observation was the measurement of left knee hyperextension (Tab. 4).

**Scoliosis intra-group analysis.** The division of the AIS group into subgroups considering the features of scoliosis,

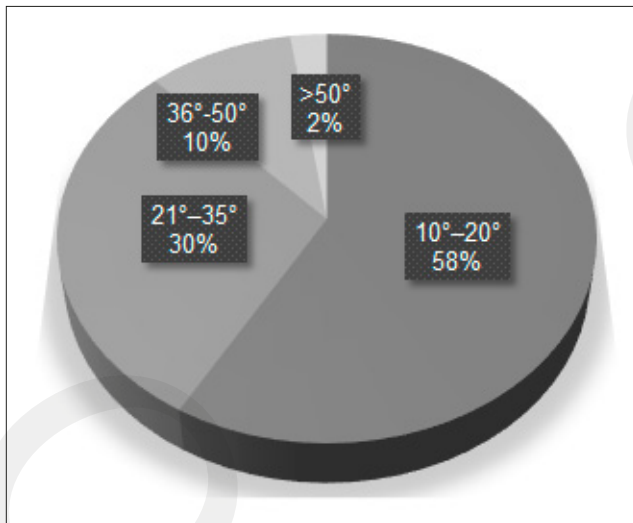


Figure 1. Angular size of scoliosis in study group (Cobb)

Table 4. Hypermobility prevalence considering different cut-off points for Beighton score and separate manoeuvres in inter-groups comparison

		Scoliosis Group n=125 (100%)	Control Group n=83 (100%)	p value
Passive dorsiflexion of the little finger	R	52 (41.6)	33 (39.8)	0.791
	L	56 (44.8)	37 (44.6)	0.975
Passive apposition of thumb to forearm	R	64 (51.2)	36 (43.4)	0.269
	L	65 (52.0)	38 (45.8)	0.380
Elbow hyperextension	R	36 (28.8)	26 (31.3)	0.697
	L	30 (24.0)	25 (30.1)	0.327
Knee hyperextension	R	37 (29.6)	16 (19.3)	0.094
	L	38 (30.4)	15 (18.1)	0.046
Forward flexion		28 (22.4)	24 (28.9)	0.288
<b>Total</b>		<b>3.2 ± 2.6</b>	<b>3.0 ± 2.3</b>	<b>0.495</b>
Hypermobility ≥4 Beighton score		64 (51.2)	34 (41.0)	0.148
Hypermobility ≥5 Beighton score		38 (30.4)	25 (30.1)	0.323
Hypermobility ≥6 Beighton score		20 (16.0)	11 (13.3)	0.209
Hypermobility ≥7 Beighton score		16 (12.8)	7 (8.4)	0.325

bone maturity or demographic characteristics, allowed for quantitative and statistical analysis.

Figure 2 shows a scatter plot of the relationship between the scoliosis angle and the Beighton score. The trend line shows a weak relationship consistent with the hypothesis made. With the division considering the angular size of scoliosis, patients with a curvature of more than 20 degrees more often presented hypermobility, but the difference was not statistically significant. Also, while comparing the subgroups with single and double curve scoliosis, no statistically significant difference was observed in the frequency of JH. The study did not show a statistically significant difference in the occurrence of hypermobility in the study group divided into subgroups by gender and by age (cut-off point at the median – 13.2 years), and no correlation with level of bone maturity represented by the Risser Sign. Table 5 summarizes the results and level of statistical significance.

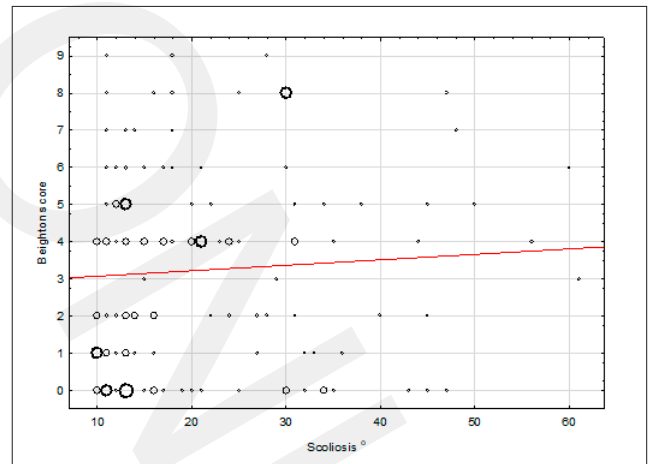


Figure 2. Beighton score – scoliosis angle values scatter plot

Table 5. Hypermobility prevalence in subjects with scoliosis divided into specified subgroups

		JH present – n (%)	JH absent – n (%)	p value
Cobb 10°-20°	n=73	36 (49%)	37 (51%)	p=0,62
	Cobb >20°	n=52	28 (54%)	
Single curve	n=56	30 (54%)	26 (46%)	p=0,64
	Double curve	n=69	34 (49%)	
Risser 0-2	n=63	34 (54%)	29 (46%)	p=0,54
	Risser 3-5	n=62	30 (48%)	
Age < 13.2 years	n=66	38 (58%)	28 (42%)	p=0,13
	Age ≥13.2 years	n=59	26 (44%)	
Females	n=92	50(54%)	42(46%)	p=0,24
	Males	n=33	14(42%)	

JH – joint hypermobility

## DISCUSSION

It is well known that joint hypermobility and scoliosis co-occur in diseases involving connective tissue, especially the Ehlers-Danlos and Marfan syndromes [19, 20]. This study attempted to investigate whether the coexistence of these two conditions is coincidental, or maybe they can also be seen in a population without connective tissue disorder, and whether JH might be a factor in the idiopathic scoliosis origin. Perhaps the very existence of articular hypermobility, regardless of its cause, promotes the occurrence of scoliosis. To the best of the authors' knowledge, this is the second-largest group of IS patients examined in terms of JH, and the only study comparing patients with scoliosis in terms of bone age denoted by the Risser Sign. This topic has already been researched by scientists. The earliest identified article addressing this issue dates back to 1983. Mattson et al. examined 51 girls with untreated scoliosis and 65 controls. The study showed that patients with scoliosis had similar flexibility or were even less flexible than healthy controls [21]. In the same year, Veliskakis described a high incidence of JH in the families

of children with AIS (adolescent idiopathic scoliosis) [22]. In 1988, Binns observed that girls with scoliosis had greater JH defined by a shorter thumb-to-forearm distance in a wrist bending test [23]. The study by Veldhuizen et al. confirmed the observations of Mattson on 10 patients and 10 controls concerning bending stiffness of the trunk – analysis did not show a clear difference between patients and controls [24]. Compared to the presented study, none of those mentioned above used the Beighton score to determine mobility, but were based on different sets of flexibility measurements.

Studies with a similar methodology to the presented study were published relatively recently. Czaprowski et al. renewed the topic by publishing two papers in 2011 and 2014. The first, including 70 patients and 58 controls, showed a statistically significant ( $p = 0.00015$ ) difference in the incidence of JH in children with scoliosis (51%) compared to healthy control group (19%). Even considering the lack of groups matching in terms of gender additional calculations on the same-gender subgroups, still showed a statistically significant higher incidence of JH in the IS group [6]. The second study repeated the results in a narrow population of Caucasian girls with scoliosis, where JH (defined as Beighton score  $\geq 5$ ), was found in 23.2% of 155 patients and 13.4% of 201 controls – a statistically significant result ( $p = 0.02$ ) [7]. In the latest study by Bozkurt et al. on a population of 822 Turkish children, 43 of whom were diagnosed with AIS – no significant correlation was found between scoliosis and hypermobility occurrence [3].

In the current study, the incidence of JH was greater in the group of IS patients, but the difference was not statistically significant. In previous studies on healthy populations of similar race, culture, and geographic location, the typical JH (Beighton score  $\geq 4$ ) prevalence was 15–20%, the same as in 2011 article by Czaprowski [3, 6, 7]. In the current study, the incidence of JH in the control group was unusually high. This may have been due to the inclusion of children with previously undiagnosed posture defects in the control group. Unfortunately, there are no studies of JH prevalence in Polish children, adolescents and the general population.

In the presented study, the only measurements that were statistically significantly different between the research and control groups, were left knee hyperextension where the results of both knee hyperextension measurements were at the border of statistical significance. This may be a possible direction for further research.

In terms of JH prevalence within the IS subgroups for scoliosis features, the findings of the current study are consistent with those of Czaprowski and Bozkut who found no association between hypermobility and scoliosis curve size or curve pattern. Demographic features compared (age, gender) showed typical trends, with JH more common in younger females, but without statistical significance [3, 6, 7]. Bone age seems to be even less related to the occurrence of JH than the record age. Perhaps if the correlation between the occurrence of scoliosis and hypermobility is poorly marked, a subsequent study would require more numerous groups. At the same time, various research methods and cut-off points make it difficult to conduct a proper meta-analysis of existing studies.

**Limitations.** The study was carried out on both first-time and previously treated patients, which could have a positive (stretching exercises) and negative (corset treatment) impact

on joint mobility. However, some of the earliest articles examined the scoliosis treatment influence on joint mobility, showing that only elbow hyperextension decreased [21].

## CONCLUSIONS

As there is no statistically significant difference in the incidence of JH in the scoliosis group compared to the control group, special vigilance for IS detection in JH children seems unnecessary.

Population studies on the incidence of articular hypermobility in Polish children and adolescents are needed.

Despite the lack of statistical significance of the results of this study, the authors believe that the assessment of joint mobility should be a permanent element of the child's examination as it affects the stability of the joints and scoliosis treatment options. Hypermobility impact on the scoliosis occurrence remains ambiguous.

## Availability of data and materials.

The datasets [16] used and/or analyzed during the current study are available from the corresponding author on reasonable request.

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