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# Assessment of lower limbs in people with Generalized Joint Hypermobility – preliminary report

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# **Abstract**

**Introduction:** Generalized Joint Hypermobility (GJH) is defined as an increased range of motion in joints. There is no causal treatment of GJH, therefore the therapy should be based on the individual needs of patients after the comprehensive diagnostic of body posture. The occurrence of deformities of lower limbs in people with GJH should be an indication for the therapy. The aim of this study was the assessment of the impact of Generalized Joint Hypermobility on the lower limbs position.

**Materials and methods:** The research was conducted on 30 children, aged 7–13 ( $10.1 \pm 1.7$ ), 51 adults aged 20–29 ( $23.2 \pm 1.6$ ). The study included the assessment of external and internal rotation of hips, tibial torsion, axis of lower limbs, longitudinal and transverse arch of the feet. Females with  $\geq 5$  and males with  $\geq 4$  scores in the Beighton test were included in the GJH groups.

**Results:** Both children and adults with GJH presents higher internal rotation of hips in comparison to the control group (p = 0.03 for right and p = 0.00 for left side, and p = 0.00 for right and p = 0.00 for left side, for children and adults, respectively). Children with GJH obtained higher values of the Clarke angle for the right foot compared to the control group (p = 0.00).

**Conclusions:** Regardless of age, subjects with Generalized Joint Hypermobility are characterized by higher internal rotation of the hip compared to healthy controls. Children with GJH present a higher longitudinal arch of the feet compared to peers, but the results fall within the normative ranges.

Keywords: hip joint, Generalized Joint Hypermobility, tibial torsion, arches of foot

### Introduction

Generalized Joint Hypermobility (GJH) is defined as an increased range of motion in large and small joints, taking into account age, gender, and ethnicity [1,2]. The prevalence of GJH in school-aged children from the east-central European region is reported to be from 5.7% to 19.2% [3] and at 10% of the young adults of Caucasian [4] and occurs three times more often in women than in men [5,6]. The varying estimates of prevalence are due to methodological differences [7].

The pathophysiology of the GJH lays in the disorders of a proportion of collagen, extracellular proteins, and hormones [8]. The lordotic posture, functional



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and idiopathic scoliosis, or flat feet co-exist with GJH [4,7,9–11]. The symptomatic (painful) form of GJH is called Benign Joint Hypermobility Syndrome (BJHS) [4]. When GJH is accompanied by other symptoms, it is defined as a health-related disorder, e.g. Joint Hypermobility Syndrome (JHS) or the Ehlers-Danlos Syndrome – Hypermobile Type (hEDS) [1].

The most commonly used diagnostic method of GJH is the Beighton 9-point test [1]. As a treatment of GJH is based on physiotherapy, its evaluation should be a part of a physiotherapeutic examination [7,12,13]. An important part of this evaluation should be the assessment of lower limbs and pelvis as a basis for the growth of the spine and the prevention of overload changes, and prevention of disorders of body posture. There is no causal treatment of GJH. Physiotherapy can be helpful in the treatment of the musculoskeletal consequences of GJH. Moreover, there are no evidence-based strategies in the treatment of GJH. Case studies confirm the effectiveness of physiotherapeutic interventions in the treatment of GJH but leave many ambiguities [14].

The aim of the study was to assess the relationship between GJH occurance and internal and external hip rotation, the axis of lower limbs, tibial torsion, and arches of the feet in two age groups. It was assumed the hypothesis that people with GJH are characterized by an increased range of hip motion, internal tibial torsion, valgus, and flat feet.

# Material and methods

A total of 130 subjects were included in the research. After taking into account the exclusion criteria the study group included 30 children aged 7–13 years (10.1  $\pm$  1.7), and 51 adults aged 20–29 years (23.2  $\pm$  1.6). Examination of two age groups may indicate that occurring disorders are independent of age and there are characteristic of GJH. The participants with GJH did

not differ significantly (p > .05) in terms of age, height, weight, and BMI in comparison to control groups. The exception is a significant difference (p < .05) in height of adults aged 20–29 years (Tab.1).

The exclusion criteria (interview information) were as follows: previous injuries of lower limbs, surgical procedures of lower limbs and spine and pain over 3 months of lower limbs and spine, the prevalence of Benign Joint Hypermobility Syndrome (the Brighton criteria), rheumatological, orthopedic, neurological, or genetic disorders.

Females with  $\geq 5$  and males with  $\geq 4$  scores in the 9-point Beighton score were included in the GJH groups [1,15]. The test consists of the abduction of the thumb to the forearm, knee hyperextension above 10 degrees, the extension of the metacarpophalangeal joint in the 5th finger above 90 degrees, elbow hyperextension above 10 degrees, and placing flat hands on the floor. Each positive test would score 1 point. The assessment was carried out for the left and right sides at the goniometer (MSD, Poland) [1,11]. The rest of the participants were qualified to the control groups.

The following measurements were performed to assess the lower limbs:

- a. the range of motion of hip internal and external rotation. The participant was in a prone position with the hip in a neutral position and the knee was in 90 degrees flexion. Researcher #1 controlled and stabilized the participant's pelvis. Researcher #2 measured the passive range of motion of internal and external rotation of the hip by placing the axis of rotation of the goniometer within the knee, and the fixed arm was parallel to the ground, and mobile along the calf [16–18].
- b. the tibial torsion. The measurement was carried out using the Thigh-Foot Angle (TFA) test. The position of the participant was the same as during the measurement of hip rotation. Researcher #2 measured the axis of rotation of the goniometer on the calcaneus,

<b>Tab. 1.</b> Characteristics of participants with GJH and from the CG (n =	81	.)
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	Children 7–13 years (n = $30$ )			Adults	20-29  years  (n = 51)	1)
	GJH	CG		GJH	CG	
	n = 13	n = 17	p	n = 19	n = 32	p
	$Mean \pm SD$	$Mean \pm SD$		$Mean \pm SD$	$Mean \pm SD$	
Age (years)	$9.3 \pm 1.4$	$10.4 \pm 1.5$	0.06	$23.4 \pm 1.0$	$23.1 \pm 2.0$	0.10
Weight (kg)	$30.4\pm10.2$	$33.3 \pm 7.4$	0.08	$64.6 \pm 10.8$	$72.4 \pm 15.2$	0.12
Height (m)	$1.38 \pm 0.1$	$1.42\pm0.1$	0.19	$1.71 \pm 0.1$	$1.76 \pm 0.1$	0.03*
BMI $(kg/m^2)$	$15.8 \pm 2.6$	$16.5 \pm 2.5$	0.46	$22.0 \pm 2.4$	$23.1 \pm 3.2$	0.46

the fixed arm along the long axis of the thigh, and the mobile one pointing between the 2nd and 3rd metatarsal bones [17,19,20].

- c. the axis of the lower limbs. The measurement was performed in a standing position with the feet hipwidth apart [21,22]. The measurement of the Q angle was conducted using a goniometer (MSD, Poland).
- d. the longitudinal and transverse arch of the feet. The Clarke angle [23–25] and the Wejsflog index [26,27] were measured on a computer podoscope (Koordynacja, Poland). The Wejsflog index was determined by calculating the ratio of the length to the width of the foot [26].

The local ethics committee (no. 6/2019) consent was obtained before the study. Written legal guardians' of the children and participants' consent were obtained. The recruitment process began with the announcement of a research project at the university and the pediatric center of rehabilitation. The next step was the informing meeting about the aim and protocol of the study. The study was conducted on healthy participants willing to participate in the research project. The participants were informed about the target of the research project, the possibility of insight into results, and the option to

discontinue their participation at any time. The singleblind test was applied.

## Statistical analysis

The statistical analysis was performed using Statistica 13.1 (StatSoft, Poland). The descriptive statistics were calculated separately for the group with GJH and the CG. Normal distribution was assessed with the use of the Shapiro-Wilk Test. T-test and Mann-Whitney U Test were applied to assess the differences between study and control groups.  $Chi^2$  test and the Spearman's correlation were carried out. The value p = 0.05 was adopted as the level of significance.

## **Results**

## Generalized Joint Hypermobility (GJH)

Children and adults obtained 5.6 points on average in the Beighton test (Tab. 2).

In children with GJH, the increased range of motion is concerned most often the knees (37.5% left side and 34.4% right side) and the elbow (34.4% left side).

In adults with GJH, the increased range of motion is concerned most often the 5th finger (29.4% left side)

Tab. 2. The results of the Beighton test

		GJH	CG		
	$Mean \pm SD$	Median (interquartile range)	$Mean \pm SD$	Median (interquartile range)	
Children	$5.6 \pm 0.8$	5.0 (5–6)	$1.7 \pm 1.6$	2.0 (0-4)	
Adults	$5.6 \pm 1.5$	5.0 (5–7)	$1.0 \pm 1.0$	1.0 (0-2)	

GJH – Generalized Joint Hypermobility, CG – Control group.

**Tab. 3.** The results of internal and external rotation of the hip joint

			GJH	CG	p
	Lower limb	Rotation (°)	$Mean \pm SD$	$Mean \pm SD$	
	Right	Internal	$60.2 \pm 10.4$	$50.2 \pm 13.0$	0.03*
Children	Left		$60.8 \pm 12.2$	$48.2 \pm 10.3$	0.00*
Children	Right	External	$38.1 \pm 8.8$	$35.7 \pm 6.3$	0.42
	Left		$39.5 \pm 8.8$	$40.0 \pm 9.7$	0.89
Adults	Right	Internal	$56.6 \pm 11.6$	$43.6 \pm 9.9$	0.00*
	Left		$56.9 \pm 11.2$	$44.6 \pm 11.6$	0.00*
	Right	External	$43.5 \pm 10.7$	$41.7 \pm 7.2$	0.47
	Left		$40.6 \pm 17.8$	$44.1 \pm 12.0$	0.40

and the thumbs (27.5% left and right side). 21.6% of adults presented knee hyperextension.

# Rotation of the hip joints

Children and adults with GJH received statistically significantly higher (p > .05) internal rotation of the hip (Tab. 3). They obtained the range of motion over  $10^{\circ}$  more compared to the CG.

There were no significant differences (p > .05) between participants with GJH and from the CG in terms of external rotation of the hip joint (Tab. 3).

#### **Tibial torsion**

Children and adults with GJH received similar (p > .05) results of tibial torsion in comparison to the control groups (Tab. 4). Each group is characterized by the high value of standard deviation (SD).

#### Axis of lower limbs

In children and adults, there was no significant correlation (p > .05) between knee valgus or varus and GJH (Tab. 5).

# Arches of the foot

Children with GJH received a significantly higher (p = .00) longitudinal arch of the right foot compared to the CG. There was no difference in the case of the left foot. All of the mean results were within the norm taking into account the age. Adults with GJH obtained similar results (p > .05) of the longitudinal arch of feet in comparison to the CG (Tab.6).

In the case of the transverse arch of feet, there were no significant differences (p > 0.05) between participants with GJH and the control groups (Tab. 6). Wejsflog index oscillated from 2.55 to 2.69.

**Tab. 4.** The results of tibial torsion

	Lower limb	GJH		CG		p
		$Mean \pm SD$	Min:Max	$Mean \pm SD$	Min:Max	
Children	Right (°)	$4.9 \pm 9.9$	-10:15	$7.4 \pm 7.1$	-10:20	0.46
	Left (°)	$7.9 \pm 12.4$	-11:22	$9.4 \pm 6.6$	-10:20	0.67
Adults	Right (°)	$2.3 \pm 6.7$	-18:10	$2.8 \pm 10.0$	-20:16	0.54
	Left (°)	$1.7 \pm 8.9$	-20:10	$4.2 \pm 8.0$	-10:20	0.63

GJH – Generalized Joint Hypermobility, CG – Control group; The "–" sign in the Min:Max columns indicates the inside foot position [18].

**Tab. 5.** The occurrence of knee valgus and varus

	Lower limb	Axis of limb -	GJH	CG	
	Lower mino	AXIS OI IIIIIO -	n (%)	n (%)	— р
		Norm	11 (91.7)	17 (94.4)	
	Right	Valgus	0 (0.0)	1 (5.6)	0.33
Children		Varus	1 (8.3)	0 (0.0)	
Children		Norm	11 (91.7)	17 (94.4)	
	Left	Valgus	0 (0.0)	1 (5.6)	0.33
		Varus	1 (8.3)	0 (0.0)	
		Norm	16 (84.2)	29 (90.6)	
	Right	Valgus	2 (10.5)	1 (3.1)	0.55
A -J14		Varus	1 (5.3)	2 (6.3)	
Adults		Norm	16 (84.2)	30 (93.8)	
	Left	Valgus	2 (10.5)	0 (0)	0.17
		Varus	1 (5.3)	2 (6.3)	

Tah 6	The result	s of arches	of the foot

		Foot -	GJH	CG	p
		root	$Mean \pm SD$	$Mean \pm SD$	
	Children	Right	$50.5 \pm 3.5$	$43.8 \pm 7.2$	0.00*
Clarka angla (°)	Cilidien	Left	$49.5 \pm 4.9$	$41.3 \pm 13.1$	0.11
Clarke angle (°)	Adults	Right	$45.2 \pm 7.1$	$44.0 \pm 8.8$	0.89
		Left	$46.2 \pm 7.8$	$44.4 \pm 6.7$	0.49
Wejsflog Index	Children	Right	$2.55 \pm 0.13$	$2.55 \pm 0.19$	0.98
	Children	Left	$2.64 \pm 0.24$	$2.56 \pm 0.20$	0.37
	A dulta	Right	$2.58 \pm 0.13$	$2.63 \pm 0.14$	0.28
	Adults	Left	$2.56 \pm 0.13$	$2.64 \pm 0.16$	0.08

GJH - Generalized Joint Hypermobility, CG - Control group, \*statistically significant difference.

#### Discussion

The impact of Generalized Joint Hypermobility on the musculoskeletal system of children and adults is an important issue in the research and clinical practice of physiotherapists. The best therapeutic methods for patients with increased range of motion in joints are still sought [28]. The physiotherapy procedures should be based on assessment of body posture, including diagnostic tests. Its inseparable element should be a comprehensive assessment of the lower limbs and the pelvis as the basis for the growth of the spine.

For the assessment of the lower limbs of people with Generalized Joint Hypermobility, there were used reliable diagnostic tools which are available to clinicians [29–32]. Therefore, in our study, we did not assess the reliability and repeatability of the tests performed. Future studies may update the data.

In the literature, no studies were found about the assessment of the rotation in the hip joints, the axis of the lower limbs, tibial torsion, and arches of feet in subjects with GJH at different ages. This issue seems to be important for posturology and the causes of gait disorders [19]. Sass et al. [33] claimed that rotational and angular disorders of the lower limbs are the most common disorders in children whose therapy is based on conservative treatment. Moreover, the authors suggest that diagnostics of the musculoskeletal system should be performed, taking into account the interview with the patient and physical examination, including a comprehensive assessment of the spine, hips, knees, and ankles, arches of the feet, and the presence of joint hypermobility [33].

In this study, the 9-point Beighton test was used. It characterizes by reliability and repeatability [1]. This test does not include the lower limb joints assessment, such as the hip, ankle, and foot joints. Another test for

the assessment of the GJH is the Lower Limb Assessment Score (LLAS), which is characterized by good sensitivity and moderate specificity [34]. However, this test, like the Beighton test, does not take into account the rotational movements in the hip joint, but only the flexion and abduction. Due to, the assessment of the range of hip rotation should be included in the physiotherapeutic examination.

## **Rotation of hip joints**

In this study, there were no differences in hip external rotation in people with GJH compared to the control groups. Significantly higher values of internal rotation were received in people with GJH compared to peers. In this study, the hip rotation was measured with the neutral position of the hip joint [18]. For children, the normative values of internal rotation were 40°-41° and external rotation were 44°-48° [35] and for adults 40°-45° internal rotation and 45°-50° external rotation [36]. Subjects from the GJH groups obtained an average of 16°-21° over the norm of internal rotation. Children from the CG achieved the range of internal rotation 8°-10° more than the norm, and adults from the CG presented the normative value. For the all of examined groups, the external rotation of the hip was about 5° lower than the assumed norms. Standard goniometric tests include measurements of hip rotation with the lower limb positioned with 0 ° or 90 ° flexion in the hip joint. According to Cannon [36], the values obtained in these angles may not be the maximum achievable by the patient. Therefore there is a probability that in people with GJH, the range of internal rotation might be even bigger during the assessment of gait. Increased range of internal rotation may be the cause of the in-toeing gait. It was found that despite numerous publications, the reason of in-toeing gait is still unknown [19]. The

most common cause is forefoot adduction, an increase of internal tibial torsion, and femoral anteversion [33]. Nikolajsen et al. [37] claimed that the kinematics of gait is the same in people with and without GJH. Different results are presented by Engelbert [38], who stated the occurrence of different gait patterns in people with the Joint Hypermobility Syndrome and Ehlers-Danlos Syndrome.

In the study of Quanbeck et al. [17], it was found that the increased number of scores in the Beighton test did not correlate with obtaining a greater range of motion of hip and knee (p = .63 and p = 0.65). Our methodology takes into account the cut-off criteria (4 and 5 scores) and division into GJH groups and control groups. Such an approach makes it impossible to perform a correlation analysis suggested by Quanbeck et al. [17].

#### **Tibial torsion**

In this study, there were no significant differences in tibial torsion in people with GJH compared to the control groups. Similar results were obtained by Jeon et al. [19]. The high values of standard deviation (SD) indicate the high individual variability of the tibial torsion in participants. The values of 10°-15° were assumed as normative values [39,40]. None of the examined groups obtained mean values within the norms. The analysis of the individual results of participants showed that 20-30% of children and adults with GJH received the norms. Jeon et al. [19] used the Thigh-Foot Supporter as a measurement standardization. The participants obtained higher results on average, but the standard deviation stayed high [19]. The high individual variability of the tibial torsion suggests to increase the group size or to search for new, more precise than TFA Test measurement tools. X-ray, USG, CT, or MRI are more often used to assess the tibial torsion [19]. However, due to the high cost of diagnostics, in the clinical practice of physiotherapists, measurements of tibial torsion are performed in TFA test. However, a low correlation was found between the results obtained in goniometric measurements and the CT measurements [41]. Similarly, in studies with MRI, the obtained results are higher than those performed in the TFA test [42].

## Axis of lower limbs

In this study, there was no significant correlation between the occurrence of GJH and the knee valgus or varus. The typical value of the femo-tibial angle, assessed on X-ray, is obtained around the age of 7 and amounts to 6–7° [21]. In the study of Pietrzak et al. [21], hypermobility was present in 10% of all patients treated for disorders of the lower limbs axis. In this study, the incidence of knee valgus or varus was lower, however, it was assessed using the goniometer (Q angle). The

normative values were 10-13° for men and 13-18° for women [43]. Pietrzak's norm is 5° in the valgus setting [21]. An important part of a clinical assessment is the reference position. Significant proximity to the medial ankles can reduce knee valgus, and the wide spacing of the feet can intensify knee valgus. This is especially important in people with joint hypermobility who are susceptible to the temporary correction of body posture. A similar phenomenon can be observed when measuring the Q angle in the supine position. It is suggested to perform the assessment in a standing position as a real determinant of the axis of the lower limbs [21]. In the study of Keays et al. [44] a positive correlation between GJH and knee valgus was found. Moreover, it was found that GJH and axis disorders of lower limbs were significantly more common in families with injuries within the knee joints than in non-injured people. Screening for both features may help identify and prevent injury [44].

## Arches of foot

Flat feet are one of the most common orthopedic disorders in children [45]. According to Evans et al. [45], flat feet are associated with a reduced range of motion in the ankle joint, inversely related to age, and correlates with obesity and overweight, and joint hypermobility. The following thresholds were adopted for the value of the Clarke angle: ≤30° – flat foot, 31–41° - lower medial longitudinal arch of the foot,  $42-54^{\circ}$  correct longitudinal arch of the foot, ≥55° – hollow foot [26]. In this study, no reduction in the medial longitudinal arches of the feet in adults with GJH was found. Moreover, statistical analysis showed higher values of the Clarke angle in children with GJH only for the right foot, but the angular range was within the norms. Cimolin et al. [46] claimed that 45% of adults with genetic syndromes characterized by hypermobility had a high arch of the longitudinal arch of the feet, 27.5% of people were within the normal range, and 27.5% presented a lower arch, which corresponded to a flat foot.

In the case of the transverse arch of feet, there were no differences between people with GJH and the control groups. The values  $\geq 2.56$  were taken as the correct arches of the foot, and values  $\leq 2.55$  as the lowering of the transverse arch [26]. The Wejsflog index oscillated from 2.55 to 2.69. The lowest values of the transverse arch obtained children from the both groups and the adults with GJH. In the literature, no studies were found about the assessment of the arches of feet in people with GJH.

The foot examination might be supplemented with an assessment that takes into account the position of the calcaneal tumor and the condition of the deltoid ligament, which dysfunctions may not be visible during podoscopic examination.

## **Conclusions**

The assessment of lower limbs which included the external rotation in hip joints, tibial torsion, and arches of feet did not show differences between children and adults with and without Generalized Joint Hypermobility. Regardless of age, people with Generalized Joint Hypermobility are characterized by higher internal rotation of the hip. Decreasing this range of motion and stabilization of the hip should be a goal of the physiotherapeutic program dedicated to people with GJH.

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## **Conflict of interests**

The authors have no conflict of interest to declare.

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