The incidence of hypermobility syndrome in girls aged 16-18

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Summary

Introduction. Joint hypermobility is a rarely diagnosed abnormality, which may be due to the fact that young people excessive range of motion is considered to be normal, and among adults and olders causes of reported pain is seen in/believed to be other diseases. The aim of this study was to determine the prevalence of generalized joint hipermobility in girls aged 16-18 years.

Material and methods. The study included 96 girls aged 16-18 years. To assess hypermobility the Beighton scale and Bulbena scale were used. Measurements of height and weight were also made. The study was complemented by an original survey/ questionnaire. Queries related to the presence of pain and its location, the history of the injury and the subjective assessment of vulnerability to injury.

Results. Based on the obtained results, it was found that the incidence of hypermobility among examined girls was 28% in the assessment using Beighton scale and 45% based on the Bulbena scale. The correlation between the two scales was defined as high (r = 0.58). The relationship between BMI factor and the amount of points in Bulbena scale was poor (r = -0.21), and between the BMI and the amount of points in Beighton scale that was found no correlation (r = 0.01). When determining the susceptibility to injury, a recurrence of an injury was reported in 42% girls with hypermobility examined using Beighton scale and in 29% of girls with hypermobility examined using Bulbena scale.

Conclusions. The incidence of hypermobility in girls aged 16 - 18 years is high and varies depending on the adopted scale. There is no significant correlation between body composition determined on the basis of BMI factor and the number of points achieved in hypermobility evaluation scale. People with excessive mobility of the joints are more prone to injuries than those with a normal range of movement.

Key words: hypermobility, Beighton scale, Bulbena scale

Streszczenie

Wstęp. Hipermobilność stawów jest rzadko diagnozowaną przez lekarzy nieprawidłowością, co wynikać może z faktu, że wśród młodych osób nadmierny zakres ruchu uważany jest za normalny, a wśród dorosłych i starszych, przyczyn zgłaszanego bólu upatruje się w innych schorzeniach. Celem badań było określenie częstości występowania uogólnionej hipermobilności stawów u dziewcząt w wieku 16-18 lat.

Materiał i metoda badań. Badaniami objęto 96 dziewcząt w wieku 16-18 lat. Do oceny hipermobilności wykorzystano skalę Beighton oraz skalę Bulbeny. Dokonano również pomiarów wysokości i masy ciała. Badania uzupełniono autorską ankietą. Pytania dotyczyły m.in. obecności bólu i jego lokalizacji, historii doznanych urazów oraz subiektywnej oceny podatności na urazy.

Wyniki. Na podstawie uzyskanych wyników stwierdzono, że częstość występowania hipermobilności wśród badanych dziewcząt wyniosła 28% przy ocenie skalą Beighton i 45% w oparciu o skalę Bulbeny. Korelację między obiema skalami określono jako wysoką (r = 0,58). Zależność między współczynnikiem BMI, a ilością punktów hipermobilności w skali Bulbeny była słaba (r = -0,21), a pomiędzy współczynnikiem BMI i ilością punktów w skali Beighton nie stwierdzono korelacji (r = 0,01). Przy określaniu podatności na urazy, powtórne wystąpienie urazu odnotowano u 42% badanych hipermobilnych wg skali Beighton i u 29% przy ocenie skalą Bulbeny.

Wnioski. Częstość występowania uogólnionej hipermobilności u dziewcząt w wieku 16 – 18 lat jest wysoka i jednocześnie różna w zależności od przyjętej skali oceny. Nie występuje istotna korelacja między budową ciała określaną na podstawie współczynnika BMI a ilością punktów osiągniętych w skali hipermobilności. Osoby z nadmierną ruchomością stawów wykazują większą podatność na urazy, niż osoby z prawidłowym zakresem ruchów.

Słowa kluczowe: hipermobilność stawów, skala Beighton, skala Bulbeny

INTRODUCTION

Hypermobility is rarely diagnosed and described, therefore, its incidence is difficult to determine and its causes and effects are little explored. It is defined as excessive range of joint movement as compared to the range typical to age, gender and ethnicity.

The term „hypermobility syndrome" (HS) was first used by Kirk et al in 1967, who interpreted it as the presence of symptoms associated with the musculoskeletal system in patients with general joint hypermobility (GJH), in the concurrent absence of systemic rheumatic diseases [1]. Later on, the terms Joint Hypermobility Syndrome (JHS) and Benign Joint Hypermobility Syndrome (BJHS) started to be used interchangeably. The adjective „benign" was introduced in 1990 to emphasize that the symptoms are not highly hazardous to health or life. However, many patients struggle with chronic pain or other debilitating symptoms, therefore, the term „benign" cannot apply to all cases [2]. Polish terminology applies the following terms: "nadmierna wiotkość stawów" (excessive joint flaccidity) or "zespół hipermobilności konstytucjonalnej" (constitutional hypermobility syndrome). Hypermobility syndrome should be differentiated from general joint hypermobility, which is a painless ailment. Currently, it is recognized that BJHS is a congenital, hereditary defect of connective tissue [3]. In each case different abnormalities may have varying severity [4]. So far, gene mutations responsible for collagen synthesis in patients with BJHS have not been detected, also, the genetic background of the syndrome is barely known, consequently, the diagnosis is based on clinical symptoms [5,6].

The spectrum of symptoms in BJHS is very broad. Simpson describes it as neuro-musculo-skeletal symptoms and divides them into two groups. The first one involves severe or traumatic symptoms, which include: sprains, acute or recurrent dislocations or subluxation. The second group of symptoms includes chronic or non-traumatic symptoms: soft tissue rheumatism, chondromalacia, back pain, scoliosis, Raynaud's phenomenon, flat feet, genu valgum, delayed motor development [7].

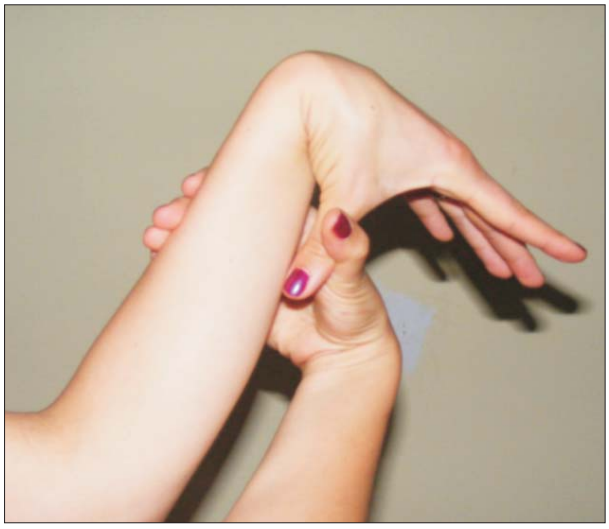
BJHS patients often describe a sense of bumping, shifting and instability of limb joints [7]. Changes are also observed in other systems, such as autonomic, gastrointestinal, cardiovascular or in connective tissue [8,9,10].

There are no standard, uniform criteria for diagnosing JHS. The most commonly used diagnostic criteria were introduced by Beighton et al, who modified Carter and Wilkinson scoring system. Both scales are based only on the joint-related JHS symptoms. Carter and Wilkinson included into the examination the following criteria:

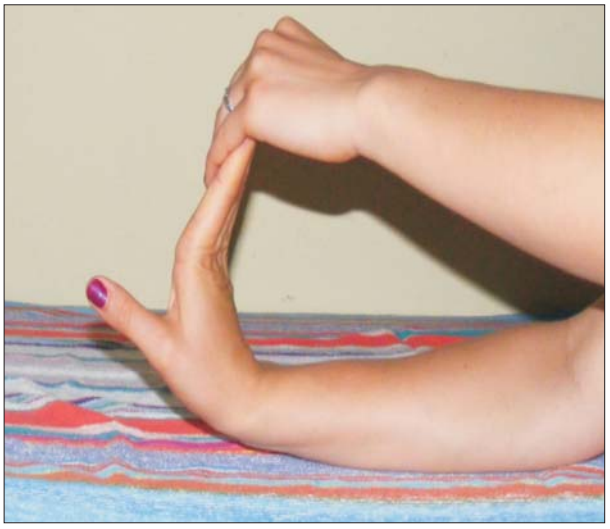
- Passive apposition of the thumb of the forearm [Fig 1],
- Passive hyperextension of the fingers so that they lie parallel with the forearm [Fig. 2]
- Hyperextension of the elbow [Fig. 3]
- Hyperextension of the knee,
- Increased dorsiflexion of the ankle and foot eversion [Fig. 4] [11].

Beighton et al. modified the scale by adding the fifth finger to the passive hyperextension in metacarpophalangeal joint [Fig. 5] as well as examining the ability of placing the palms flat on the floor while maintaining the knees in full extension [Fig. 6] [12].

Another scoring system used to diagnose hypermobility syndrome is Antonio Bulbena's scale, also known as Hospital del Mar criteria. Hospital del Mar criteria incorporate three criteria of the Beighton scale, four from the Carter and Wilkinson scale, as well as the evaluation



Pic. 1. Abduction of the thumb to the forearm



Pic. 2. Passive hyperextension of all MCPs of the II-V fingers to a position parallel to the extensor aspect of the forearm

of movability of five additional joints. It was the first scale to take into account extra-articular symptom, specifically appearance of ecchymoses and to introduce a different boundary value for diagnosing hypermobility syndrome in men and women [13].

In 1998 the Brighton criteria were developed with the application of the Beighton hypermobility score and a number of additional symptoms, including characteristic extra-articular ones [Tab 1].

Hypermobility can be diagnosed in the presence of either both 'major', one 'major' and one 'minor' or four „minor" criteria of the above mentioned scales [4].

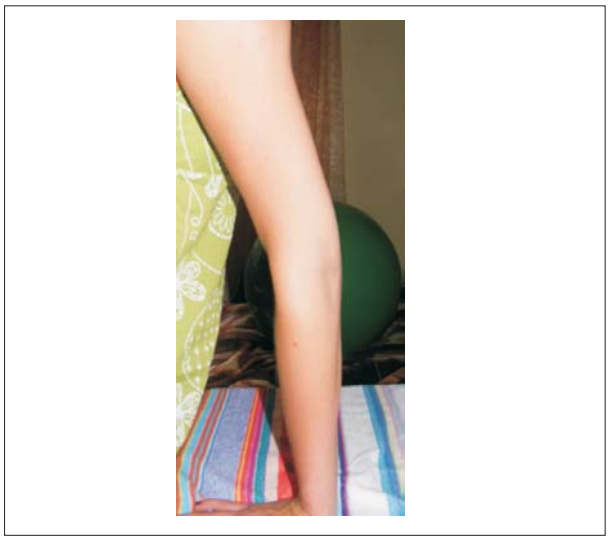
When diagnosing hypermobility syndrome it is important to exclude conditions accompanied by increase hypermobility syndrome, such as Ehlers-Danlos syndrome and Marfan syndrome [7,14].

There is no agreement among clinicians and scientists which scale should be generally applied, nonetheless, the most commonly used scale in clinical conditions is the Beighton hypermobility score [15].

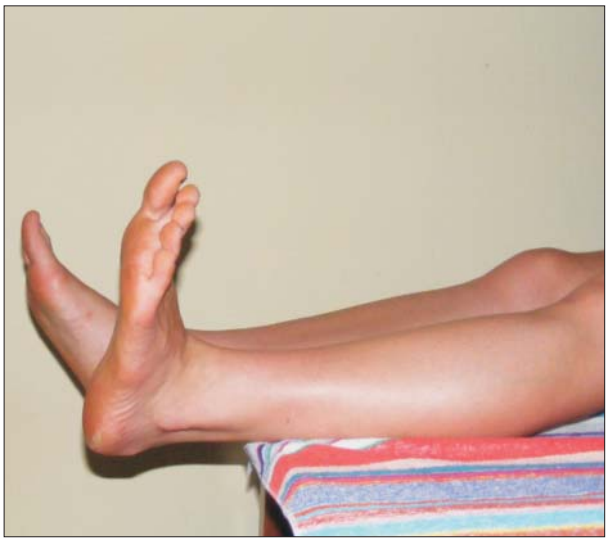
Occurrence of joint hypermobility differs greatly depending on gender, age and ethnic group [16]. In the European population it may reach 10%, while studies of African and Asian populations report occurrence of 25-30% [17,18]. According to Kopff women suffer from hypermobility 3-5 times more often than men [4]. More than 5% of healthy women report ailments associated with hypermobility compared to 0.6% in men [19].

Frequency of joint hypermobility declines with age due to changes resulting from maturing and aging [4]. The research carried out by Stodolny and Tybinkowska estimate the occurrence of hypermobility at 12% of the studied population, with occurrence among women (16%) being twice as high as in men (8%) [20].

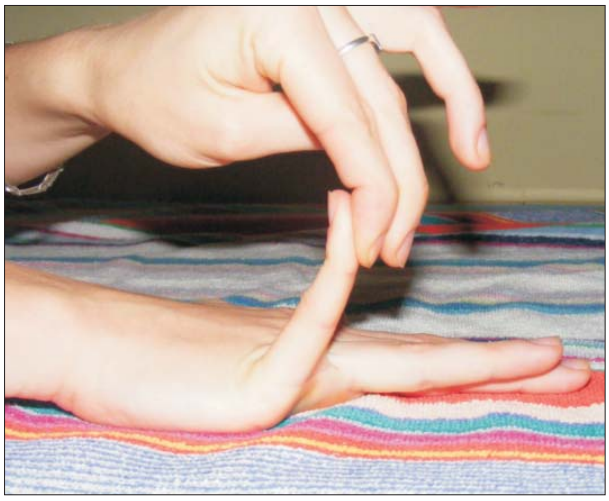
The aim of the study was to estimate the incidence of GJH among girls aged 16–18 years.



Pic. 3. Knee hyperextension



Pic. 4. Ankle dorsiflexion



Pic. 5. Extension of the MCP joint of the fifth finger

Pic. 6. Touching the floor with the palms of the hands

MATERIAL AND METHOD

96 adolescent girls from Rzeszow's 1st High School aged 16-18 years (average 16,67) participated in the study. GJH was diagnosed using the Brighton criteria and the Bulbena scale. When using the Beighton hypermobility score both right and left side of the body was examined [Table 2]. One point was allocated for performing one task. When applying the Bulbena scale only the dominant side of the body was examined [Table 3]. One point was given for each criterion met.

In addition, the examination included measurement of movement range by goniometer and a survey. For the former a standardized two-armed metal goniometer SH5105 manufactured by MSD Europe BVBA was used. GJH was diagnosed when the patient obtained 4/9 points in the Beighton hypermobility score and 5/10 in the Bulbena scale.

BMI (body mass index) was calculated for each participant based on the weight and height measurement. The survey involved questions related to previous injuries and their location, subjective assessment of the susceptibility to injuries, pain and its location, and other symptoms that are associated with hypermobility syndrome. The obtained data were evaluated using the Statistica 10.0 with the significance level of $p<0.05$. Pearson's correlation coefficient was calculated for each scale in order to find out if there is a relationship between BMI and the results obtained in hypermobility score.

RESULTS

Almost 1/3 of the respondents were diagnosed with hypermobility using the Beighton hypermobility score while the Bulbena score recognised GJH in 43 girls, which accounted for almost 45%. Taking into account both scales, Ł of the respondents met the criteria [Table 4].

Graphs 1 and 3 present the point binomial in each scale. In the Beigton scale most girls were appointed 1-3 points, which is a standard value. Hypermobility, which equals 7-9 points, was diagnosed in 3 patients; in 8 subjects no signs of hypermobility were found. A score of 4-6 points was observed in 24 patients. Graph 2 indicates even distribution of points in the range of 2-7. There were no cases of extreme hypermobility (9-10 points); also there was not an incident of a negative score in either test.

Statistical calculations based on the data presented in Figures 1 and 2 reveal a significant relationship between

Tab. 1. Brighton criteria

Major criteria	Minor criteria
1. Beighton score > 4/9 points (either now or in the past) 2. Joint pain for longer than three months in four or more joints	1. Beighton score = 1, 2, or 3/9 points (0, 1, 2, 3 if > 50-years old) 2. Joint pain > 3 months in 1 - 3 joints, or back pain for > 3 months, spondylosis, spondylolysis/spondylolisthesis 3. Dislocation/ subluxation, or partially dislocating, more than one joint or the same joint more than once 4. Soft tissue rheumatism > 3 lesions (e.g., epicondylitis, tenosynovitis, bursitis) 5. Marfanoid habitus, arachnodaktylia (positive Steinberg/wrist signs) 6. Abnormal skin – striae, hyperextensibility, thin skin, papyraceous scarring 7. Eye-related symptoms: drooping eyelids, myopia or antimongoloid slant 8. Varicose veins, or a hernia, a rectal or uterine prolapse

Tab. 2. Beighton scale

Test	Right side	Left side
Ability to extend the MCP joint of the fifth finger back beyond 90°		
Ability to bend the knee backwards/hyperextend beyond 10°		
Ability to bend right elbow backwards /hyperextend beyond 10°		
Ability to bend the thumb back on the front of your forearm		
Ability to put hands flat on the floor with knees straight		
Total		

the Beighton hypermobility score and the Bulbena scale. Pearson's correlation coefficient for these variables amounts to r = 0.58.

BMI was calculated on the basis of anthropometric measurements and the following was found: 3/4 of respondents were within the norm, a large number of respondents (17.7%) was in the underweight range, i.e. below 18.5 [kg/m2], and a small percentage of the respondents represented BMI above normal.

The correlation coefficient between BMI and the Beighton hypermobility score was r = 0.01, whereas between BMI and the scores in Bulbena scale it amounted to r = - 0.21, which leads to a conclusion that there is no correlation in the case of the Beighton hypermobility score while the correlation between BMI and the Bulbena scale is weak. It is worth noting that the correlation with the Bulbena scale is negative, which indicates that when the BMI increases, the average value of points on a hypermobility scale decreases.

Responders were divided into two groups: patients with hypermobility and a normal movement range, and survey questions on the prior injuries, their type and possible recurrence were analyzed [Table 5].

It was found that the incidence of injuries in each group is similar and equals over 70%. When estimated with the Beighton hypermobility score, more injuries were reported in the the group with normal joint mobility, whereas almost 1/3 of responders with hypermobility had not suffered any injury. A very significant difference can be captured in repeatability of injuries. Almost half of the respondents with hypermobility had suffered a second injury, while in the second group the number

Tab. 3. The Bulbena scale

Criterion	Points
External rotation of the shoulder up to more than $>85^0$	
Passive hyperextension of the elbow is 10° or more	
Passive apposition of the thumb to the flexor aspect of the forearm at a distance of less than 21 mm	
Passive dorsiflexion of the MCP joint of the fifth finger is 90° or more	
Passive hip abduction to an angle of 85° or more	
Knee flexion allows the heel to make contact with the buttock	
Passive shift of the patella to the lateral side of the tibia	
Foot dorsiflexion $>20^0$	
Passive dorsal flexion of the toe of 90° or more	
Ecchymoses after minimal trauma	
Total	

Tab. 4. The prevalence of generalized joint hypermobility

	Beighton scale score		Bulbena scale score		Score in both scales	
	n	%	n	%	n	%
hypermobility	27	28.13	43	44.79	22	22.92
norm	69	71.87	53	55.21	74	77.08

Tab. 5. The presence and repeatability of injuries

	Beighton scale score				Bulbena scale score			
	hypermobility		*norm*		*hypermobility*		*norm*	
	n	%	n	%	n	%	n	%
Presence of injury	19	70.37	50	72.46	31	72.09	38	71.70
Lack of injury	8	29.63	19	27.54	12	27.91	15	28.30
Repeated injury	8	42.11	12	24	9	29.03	11	20.75

Tab. 6. Subjective assessment of the susceptibility to injury (the Beighton scale)

Frequency of undergoing an injury	n respondents	n hypermobile
very often due to clumsiness	8	1
very often in sport	0	0
average	43	13
very seldom	36	11
never	9	2

amounted to almost 1/4. Unlike in the case of the Beighton hypermobility score, more responders with a previous injury were in the hypermobility group when evaluated by the Bulbena scale. When analyzing the recurrence of injury in the Bulbena scale evaluation it was noted that it is greater in the hypermobility group. Almost 1/3 of respondents in this group had suffered a recurrent injury, and among patients with a normal range of movement the number was a little more than 20%. These differences were not statistically significant.

Next, we analyzed of the number of particular types of injuries in groups of patients, defined by the Beighton hypermobility score and the Bulbena scale. This showed that the most common injuries were a dislocated finger or muscle strain, respectively 30% and 38% in each group. The least frequently occurring injury is a ligament rupture, its occurrence amounted to 4% in both groups. There is a noticeable disparity in the assessment with the Bulbena scale. In subjects with hypermobility the frequency of ligament rupture is almost 7%, in patients with normal movement range it is less than 2%.

The incidence of dislocations in subjects with hypermobility constituted almost 1/4 of all injuries both when assessed by the Beighton hypermobility score and the Bulbena scale. In subjects with normal range of joint movement a dislocation is the fifth most common type of injury. Only one in ten respondents with normal joint mobility suffered from a dislocation.

In the Bulbena scale assessment, in both groups, sprains appeared in more than L' of the respondents. Sprains occurred in 37% of hypermobility patients and only in 23% in the group with normal range of movement when assessed by the Beighton hypermobility score.

According to the assessment with the Bulbena scale fractures are much more common among subjects with normal joint mobility (26%) than in the second group (14% of all injuries). When assessed by the Beighton hypermobility score 22% of patients with a normal range of movement declared a previous fracture while in the second group, it was reported by more than 18% of the respondents.

The results of the survey on pain and its location is presented in Figure 3. The vast majority of respondents did not report any pain. 10% have symptoms of knee pain and a very small percentage of patients declared suffering from a different kind of pain.

Finally, subject were asked to assessment their own susceptibility to injury. The vast majority of respondents declared that the frequency of their injuries is average or very rare. Eight of the responders, including one with diagnosed hypermobility, perceives themselves as often undergoing injuries, believing the reason for this is clumsiness. The complete absence of injuries was declared by nine respondents, including two with excessive range of movement [Table 6].

DISCUSSION

It is hard to clearly define the prevalence of GJH. It greatly depends on the adopted classification system. The obtained results estimated it at a level of 27-44% depending on the applied scale. Various results can be found in the literature. Seckin et al using the Beighton hypermobility score found hypermobility in 11.7% of over 800 Turkish students aged 13-19 years. The prevalence of the factor in women was 16.2% [21]. Similarly, researchers who applied the same scale diagnosed hypermobility in 13.6% of girls in a study conducted in Iran between 1994 - 2004 [22].

The percentage of subjects with hypermobility when assessed by the Bulbena scale and the Brighton criteria varies considerably, all the same, a strong correlation between the scales was found. Among the respondents there were patients who were diagnosed with hypermobility with the Beighton hypermobility score, yet in the Bulbena scale they received less than 5 points. The opposite was more frequent. In both scales GJH was

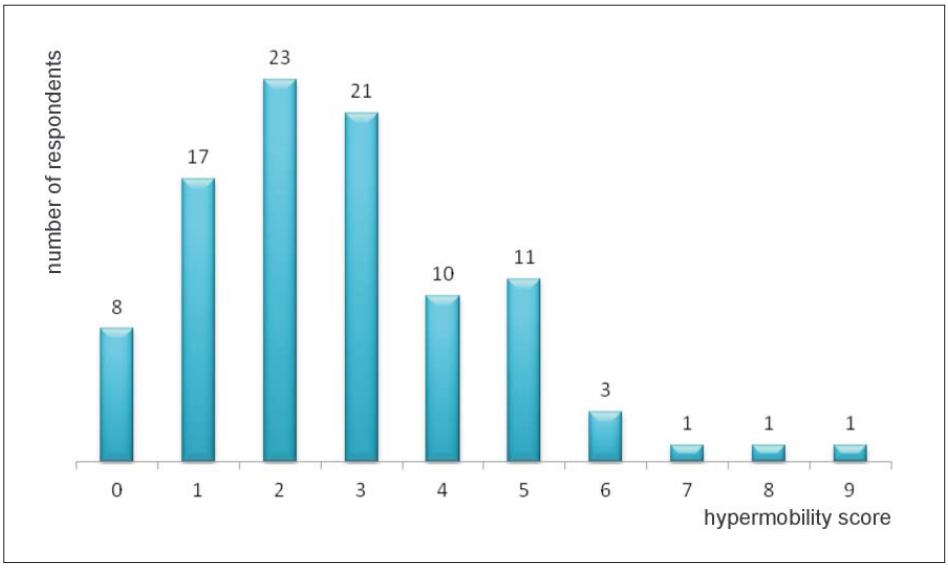


Fig. 1. Distribution of the Beighton scale scores

found in 22.92% of patients, which is a result similar to the observations of other authors. Ferrell et al compared the results received with the Beighton hypermobility scale and the Contompasis scale and observed a very high correlation (r = 0.87) [23]. Both scales are equally suitable for hypermobility diagnosis. Nevertheless, the Beighton hypermobility score is less complicated, quicker and easier, which is especially beneficial for physiotherapists and general practitioners.

The above described differences in the assessment of hypermobility prevalence, even within the same ethnicity, sex and age group, point to the need of establishing unified criteria of evaluation and diagnosis. Some reports indicate that some tests used to diagnose hypermobility are more specific than others, which should be applicable in primary diagnosis in children [24]. Smits-Engelsman et al postulate that for the Beighton hypermobility score 7/9 points should become the boundary value for children, not 5/9 which is used for adults. In this research a study was conducted in a group of children aged 6-12 years and it was noted that over 35% of respondents received 5 or more points. Thus the suggestion to increase the threshold in order to prevent overdiagnosis of this syndrome in young patients [25].

According to Stodolny, hypermobility in mature age is often accompanied by obesity, which does not reduce the range of movement [26]. In our research the vast majority of subjects fit within the range of a normal weight, and only a few have a tendency to be overweight or obese. A weak negative correlation was observed between the number of points in the Bulbena scale and BMI, which indicates that an increase in body weight in relation to the increasing score caused a decrease in range of joint mobility. No relationship was observed between BMI values and the points obtained in the Beighton hypermobility score, therefore, there is no basis to conclude that at a young age hypermobility of joints in any way affects the body structure.

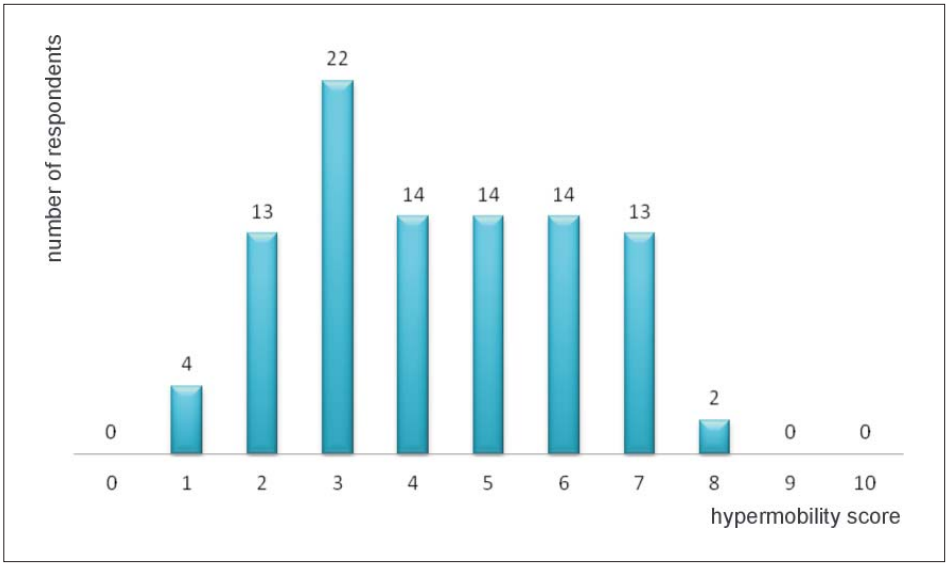


Fig. 2. Distribution of the Bulbena scale scores

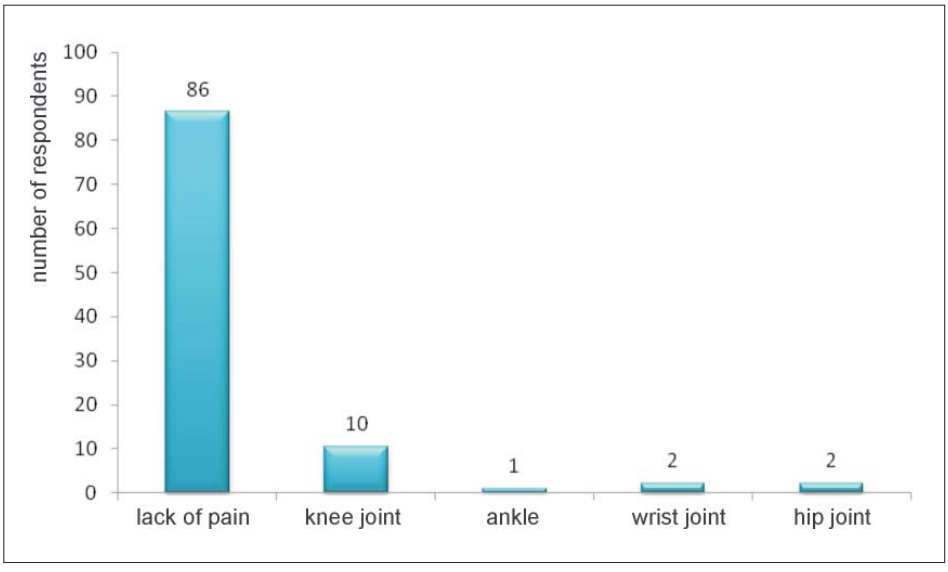


Fig. 3. Pain and its location

Generalized joint hypermobility is presented in the literature as a mild set of symptoms. The term „mild" is intended to emphasize that this disorder is not accompanied by other significant symptoms caused by defective collagen structure and that it is a nonprogressive and noninflammatory disorder of connective tissue. In our research 15% of subjects reported articular pain. Many publications show that 9.4-32% of children and adolescents aged 0-18 years complains about chronic musculoarticular pain, the most affected group being the 12-15-year-olds. 64% of children with diagnosed hypermobility syndrome complained of articular pain [27,28].

In literature there are reports that describe pain symptoms of hypermobility syndrome usually relate to knee joint [29]. The same result was obtained in our study - knee pain was reported in 10% of patients. In addition, pain of hip, wrist and ankle was found in respectively, 2%, 2% and 1% of subjects.

Physical activity is essential for the proper development of children and adolescents, both healthy, with a reduced range of movement, as well as those with hypermobility. There are beneficial activities and sport disciplines for each of these groups. Those with excessive range of movement should focus on stabilizing exercises, exercises improving bathyesthesia and equilibrium, as well as exercises strengthening postural muscles. Ferrell et al presented a 8-week closed chain lower body exercise program. The following results were obtained in 16 of 18 patients: improvement in proprioception, equilibrium and strength of knee flexors and extensors [23]. Disorders of these mechanisms as well as a reduction in muscle strength can lead to a greater susceptibility to injury. Our findings suggest that excessive joint mobility predisposes to re-emergence of a prior injury, therefore, a total amount of injuries experienced by hypermobility patients is higher than in subjects with normal range of movement. Responders with increased mobility do not consider themselves to be more likely to experience an injury than their peers; according to their estimates they undergo injuries with a usual frequency or rarely.

Studies on the incidence of injuries among ballet dancers showed that tendon injuries are much more common among dancers with hypermobility, both women and men. They also need a lot more time to fully recover from an injury [30]. In our study the most common injuries among subjects with excessive mobility are dislocations and sprains. Susceptibility to this type injury suggests compromised mechanisms of balance and proprioception. It may, however, be associated with other factors, such as: the type of physical activity practiced, existing uncorrected vision impairment or other comorbidities.

Engelbert et al investigated reduced exercise tolerance in children and adolescents with excessive range of movement and musculoskeletal complaints. The authors pointed out that the reduced endurance resulted from limited physical activity, which in turn is caused by pain [27]. Reduced endurance and limited physical activity can also be connected to chronic fatigue syndrome, which is present in 82% of hypermobility children [31].

Lack of physical activity, decreased muscle strength and chronic fatigue can have other consequences, strongly influencing young organisms. One of them may be frequently occurring postural defects. In our research 36% of respondents had a diagnosed postural defect, with the most frequent one being scoliosis, followed by flat feet; genu valgum was rarely identified. Our results confirm data from the literature, i.e. 19% of hypermobility subjects suffers from scoliosis [31]. The research by Czaprowski et al showed that hypermobility is more common in patients with idiopathic scoliosis than in those without this impairment. It was also noted that hypermobility was more frequently observed in subjects with one arch scoliosis. No relationship was noted between the scoliosis angle, uppermost involved vertebra, the number of vertebrae in the primary curvature, applied therapeutic techniques and the incidence of hypermobility [32].

Considering all the above-mentioned ailments and disorders afflicting people with excessive mobility of the joints, the question arises whether and to what degree JHS affects the quality of life. It is known that a significant number of the afflicted subjects function well unaware of their ailment or treat it as their advantage, many lead active lifestyles, some are professional athletes who owe their good results to the disorder itself. However, a number of patients with hypermobility syndrome suffer from a chronic pain. Russek described a case in which pain introduced severe limitations to patient's life. The described patient was able to perform all activities of daily living despite the accompanying pain that required compensatory movements in certain situations. She stated that pain limited her hitherto active lifestyle [15].

The previously mentioned research by Ferrell et al investigated the effect of exercise on quality of life (SF-36 rating), and pain perception (VAS scale was used) in patients with JHS. After 8 weeks of recommended activity decline in VAS scale and significant improvement in both physical and mental health as well as in life quality [23].

Therefore, there is no doubt that the excessive range of movement with accompanying ailments affect quality of life, yet, appropriately selected training program can improve functioning and prevent some complications.

CONCLUSIONS

1. The incidence of general hypermobility syndrome in girls aged 16-18 years is different depending on the adopted scale of evaluation. When based on the Beighton hypermobility score it amounts to 28%, while applying the Bulbena scale hypermobility was present in 45% of patients.
2. There is no significant correlation between BMI and the points achieved on the hypermobility scale.
3. Patients with hypermobility are more vulnerable to injuries than those with a normal range of mobility as well as extremely vulnerable to recurrent injuries.

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