



The Effect of Hypermobility on Pain and Quality of Life in Young Adults

Genç Yetişkinlerde Hipermobilitenin Ağrı ve Yaşam Kalitesi Üzerine Etkisi

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ABSTRACT

Objective: Hypermobility is the excessive range of motion of joints, and related to various musculoskeletal and extra-articular problems that may significantly impair quality of life (QoL) by causing pain. The aim of the study is to examine the prevalence of hypermobility in young adults, and its relationship with pain in various regions of body and QoL.

Methods: Two hundred and twenty five volunteers, aged between 17 and 23, were classified as subjects-with-hypermobility or subjects-without-hypermobility according to the Beighton Criteria. Chronic pain was identified by using Nordic Pain Questionnaire, QoL was identified by Short Form-36 (SF-36) Questionnaire. Pain presence in 9 body regions and SF-36 scores were compared between groups using chi-square test and Independent Samples T-test, respectively.

Results: Of the participants, 164 (64%) were female, 91 (36%) were male, 119 (46.7%) had hypermobility. Upper back was the body region with the highest pain prevalence where 79% of hypermobile and 74% of non-hypermobile subjects reported pain at least once in past 12 months. Pain prevalence in body regions did not differ between groups ($p>0.05$). In terms of QoL, physical and mental component scores of SF-36, as well as all subgroup scores except social function were significantly lower in hypermobile subjects ($p<0.05$).

Conclusion: Pain prevalence in different body regions did not differ between subjects with and without hypermobility whereas the QoL

ÖZ

Amaç: Hipermobilitate eklem hareket açıklığının normal olan değerlerden fazla olması olarak tanımlanmaktadır. Çeşitli muskuloskeletal yaralanmalar ve ekstra-artiküler problemlerle ilişkilidir ve vücudun farklı bölgelerinde kronik ağrılara sebep olarak yaşam kalitesini (YK) belirgin şekilde etkileyebilir. Bu çalışmanın amacı, genç yetişkinlerde hipermobilitenin görülme sıklığını incelemek ve hipermobilitenin vücudun çeşitli bölgelerindeki ağrı prevalansı ve YK üzerindeki etkisini araştırmaktır.

Yöntemler: Araştırmaya yaşları 17-23 arası olan toplam 255 gönüllü katıldı. Katılımcılar Beighton Hipermobilitate Kriterleri'ne göre hipermobilitesi olan veya olmayan bireyler olarak sınıflandırıldı. Nordic Ağrı Anketi ile kronik ağrının varlığı ve lokalizasyonu; Kısa Form-36 (KF-36) anketi ile de YK değerlendirildi. Gruplar vücudun 9 bölgesi için bildirilen ağrı açısından ki-kare testiyle, KF-36 anket skorları açısından ise Bağımsız Örneklem t-testiyle karşılaştırılarak değerlendirildi.

Bulgular: Katılımcıların 164'ü kadın (%64) 91'i erkek (%36) idi. Katılımcıların 119'unda (%46,7) hipermobilitate saptandı. Sırt bölgesi katılımcıların en sık ağrı hissettiği bölgeydi ve son 12 ay içerisinde hipermobilitesi olan olguların %79'unun, hipermobilitesi olmayan olguların ise %74'ünün bu bölgede ağrı hissettiği saptandı. Gruplar arasında vücut bölgelerindeki ağrı prevalansları açısından anlamlı fark bulunmadı ($p>0,05$). YK açısından hipermobil bireylerde KF-36 anketinin fiziksel ve mental total skorları ile sosyal fonksiyon haricindeki alt grup skorları daha düşüktü ($p<0,05$).

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was significantly impaired in hypermobile subjects. Hypermobility is a substantial anatomical finding in young adults that should not be disregarded. Education, emotional support and encouraging about strengthening and proprioception exercises may contribute to their quality of life.

Keywords: Joint laxity, hypermobility, pain, quality of life, anatomy

Sonuç: Hipermobiliteye sahip genç yetişkinlerde hipermobilitesi olmayan bireylere kıyasla vücudun farklı bölgelerindeki ağrı prevalansları açısından fark saptanmamıştır; ancak YK’inde belirgin bir etkilenme mevcuttur. Hipermobilitte, bu yaş grubunda göz ardı edilmemesi gereken bir anatomik bulgudur. Bireylerin bu konu ile ilgili eğitimi, emosyonel yönden desteklenmesi ve güçlendirme ve propriocepsiyona yönelik egzersiz programı açısından cesaretlendirilmesi ile YK’sine katkı sağlanabilir.

Anahtar Sözcükler: Eklem laksitesi, hipermobilitte, ağrı, yaşam kalitesi, anatomi

Introduction

Joint hypermobility is defined as the range of motion of the joint that is greater than normal values according to age, gender and ethnicity. It is called Generalized Joint Hypermobility (GJH) when joint hypermobility is asymptomatic and Benign Joint Hypermobility Syndrome (BJHS) in cases accompanied by musculoskeletal symptoms (1). Therefore, joint hypermobility can be considered as a descriptive statement rather than a diagnosis (2). The primary cause of hypermobility is ligamentous laxity which is determined by genes encoding collagen, elastin, and fibrillin (3).

The prevalence of hypermobility varies according to age, gender and ethnicity. It is more common in females, Asians and Africans, and its prevalence is higher in childhood and tends to decrease towards adulthood (4,5). In addition to this genetic predisposition, it can be acquired as a result of intense training and stretching. Recently, it has been considered that it may overlap with or be a mild form of Heritable Connective Tissue Disorders (HCTDs) such as Ehlers-Danlos syndrome (EDS), Marfan Syndrome and Osteogenesis Imperfecta, in which connective tissue matrix proteins are affected. It is even thought that BJHS is the same as EDS type III (hypermobile type) (3). There are various scoring systems in diagnosis. Beighton scoring system and the revised Brighton criteria are commonly used for GJH (6) and BJHS (7), respectively. Lately, updates have been made emphasizing that the Beighton score should have different cut-off values according to the different ages (8). The results regarding the prevalence of GJH in the literature are highly variable due to the different evaluation methods, cut-off values, populations and age groups chosen in the studies.

Hypermobility may not cause any symptoms. Moreover, it may be advantageous for musicians and individuals who is engaged in gymnastics, martial arts, ballet (9-11). However, changes in the connective tissue matrix lead to joint and soft tissue injuries, and an inability of fast and adequate recovery through the instability of the joint capsule and decreased flexibility of ligaments and tendons (1,12). Besides, it has been observed to be associated with a wide variety of extra-articular clinical conditions such as muscle weakness, decreased motor abilities, skin problems, rectal-uterine prolapsus, pan intestinal dysmotility, fibromyalgia, low bone density, anxiety and panic disorders (13-19). Hypermobility is also associated with chronic pain which leads to avoidance of

exercise to prevent pain. This sedentary life causes functional disability, chronic fatigue, sleep disorders, deterioration in work and social lives, anxiety, depression, and a decrease in quality of life (QoL) all of which make the individual more inactive as a vicious circle (1,5). The aim of this study is to investigate the prevalence of hypermobility in young adults, and evaluate its impact on pain and QoL, which is often disregarded.

Methods

This observational and cross-sectional study was conducted with a total of 255 participants aged between 17-23 years. Participants were selected among the university students by using the university notice board, and volunteers were included in the study. Those with a diagnosis of orthopedic or neurological diseases, and a history of musculoskeletal surgery that might affect mobility were excluded. Participants were informed about the purpose of the study and the evaluations to be applied. An informed consent form was obtained from each participant. This study was approved by the Non-interventional Clinical Research Ethics Committee and conducted in accordance with the Declaration of Helsinki.

Demographic data of participants including age, gender, height, weight, body mass index, smoking status, sports status, and the department they studied were recorded. Participants were classified as “subjects with hypermobility” and “subjects without hypermobility” according to the Beighton criteria. Presence and localization of chronic pain were evaluated with the Nordic Pain Questionnaire (NPQ). QoL was evaluated with the Short Form-36 questionnaire (SF-36). Participants with and without hypermobility were compared in terms of chronic pain and QoL.

The Beighton criteria examines the presence of hypermobility with 9 movements involving the extremities and trunk (Table 1). Items about extremities are evaluated symmetrically as right and left, while the trunk is evaluated with only one item. Each item is scored as “1” if the movement can be done, “0” if it cannot be done. In this evaluation, which is made out of 9 points in total, individuals with a score of 4 and above are considered to have joint hypermobility (6).

The NPQ is used to assess the localization and severity of chronic pain. “Did you feel pain in the last 12 months?”, “Did the pain in that area prevent you from doing your job in the last 12 months?”, “Did you feel pain in the last 7 days?” are asked to

Table 1. Beighton criteria

	Right	Left
Passive dorsiflexion of the little fingers beyond $\geq 90^\circ$		
Passive apposition of the thumbs to the flexor aspects of the forearm		
Hyperextension of the elbow beyond $\geq 10^\circ$		
Hyperextension of the knee beyond $\geq 10^\circ$		
Forward flexion of the trunk with knees fully extended so that the palms of the hands rest flat on the floor		
Total score (9 points)		

evaluate the pain in neck, shoulders, upper back, elbow, wrists-hands, lower back, hips-thighs, knees, ankles-feet (20). The validity and reliability in Turkish language were demonstrated (21).

The SF-36 is used to assess health-related QoL physically and mentally. It evaluates 8 contents of health with 36 items: physical function, limitation due to physical and emotional problems (physical and emotional role limitations), vitality (energy/fatigue), mental health, social function, bodily pain and general perception of health. Scores vary between 0-100, with 100 points indicating the best and 0 points the worst health situation. In addition to giving a score for each scale, two separate total scores can be calculated as physical and mental component scores (22). Turkish validity and reliability of SF-36 were demonstrated (23).

Statistical Analysis

The SPSS v.20 program (IBM Inc. USA) was used for data analysis. The distribution characteristics of the data were analyzed with the Kolmogorov-Smirnov test. Independent Samples t-test was used for numerical variables with normal distribution. Mann-Whitney U test was used for non-normally distributed or ordinal variables to compare pain and QoL scores of participants with and without hypermobility. Chi-square test was used to compare categorical (nominal) variables between groups. The level of significance was accepted as $p < 0.05$ for all analyses.

G-Power 3.1 (Universitat Dusseldorf, Germany) computer program was used to determine the sample size (24). It was shown that the QoL score measured by SF-36 was 55 ± 25 in individuals with hypermobility and 87 ± 18 in healthy controls (25). In order to detect a similar difference in this study with 95% confidence level and 80% power, it was calculated that at least 10 patients should be included in each of the “subjects with hypermobility” and “subjects without hypermobility” groups. Considering the prevalence of hypermobility was at least 5% in the healthy population (5), it was calculated that including at least 200 participants in the study would be appropriate to determine hypermobility in at least 10 participants among the total participants.

Results

Of the participants, 164 (64%) were female, 91 (36%) were male. The mean age of the participants was 19.81 ± 1.41 years. According to the Beighton Criteria, 119 (46.7%) of the

participants were hypermobile, while 136 (53.3%) were not hypermobile. Hypermobility was present in 50% of females and 40.7% of males. The demographic characteristics of the participants are shown in detail in Table 2.

Painful regions of the participants in the last 12 months were examined according to NPQ and it was observed that 79% of the hypermobile subjects felt pain in the upper back and 70.6% in the lower back region. Similarly, in the group without hypermobility, the most affected regions were upper back (74.3%) and lower back (69.9%). There were no statistically significant differences between participants with and without hypermobility in terms of pain prevalence in all regions ($p > 0.05$) (Table 3).

When the QoL of the participants was evaluated with SF-36 (Table 4); subgroup scores of physical function, role limitation (physical-emotional), energy, mental health, bodily pain, general health and total scores of physical and mental component were statistically lower in hypermobile individuals compared to individuals without hypermobility ($p < 0.05$). There was no

Table 2. Demographic characteristics of the participants

Age (years) (mean \pm SD)	19.81 \pm 1.41
Body mass index (kg/m ²) (mean \pm SD)	21.93 \pm 2.88
Gender	n (%)
Female	164 (64.3)
Male	91 (35.7)
Smoking	n (%)
Smokers	35 (13.7)
Non-smokers	220 (86.3)
Regular physical exercise	n (%)
Doing regular physical exercise	68 (26.7)
Not doing regular physical exercise	187 (73.3)
Body mass index (kg/m ²)	n (%)
Underweight (<18.5)	21 (8.2)
Normal weight (18.5-24.9)	203 (79.6)
Overweight (25-29.9)	26 (10.2)
Obesity class I (30-34.9)	5 (2)
Hypermobility status	n (%)
Subjects with hypermobility	119 (46.7)
Subjects without hypermobility	136 (53.3)
SD: Standard deviation	

Table 3. Pain presence reported in last 12 months

	Normal subjects (n=136)	Hypermobile subjects (n=119)	p value
Neck	94 (69.1%)	83 (69.7%)	1.000
Shoulder	75 (55.1%)	75 (63%)	0.251
Upper back	101 (74.3%)	94 (79%)	0.376
Elbow	20 (14.7%)	16 (13.4%)	0.858
Wrist	56 (41.2%)	54 (45.4%)	0.528
Lower back	95 (69.9%)	84 (70.6%)	1.000
Hip	36 (26.5%)	39 (32.8%)	0.275
Knee	37 (27.2%)	40 (33.6%)	0.277
Ankle	41 (30.1%)	33 (27.7%)	0.681

Table 4. Quality of life outcomes based on SF-36

	Normal subjects (n=136)	Hypermobile subjects (n=119)	p value
Physical function	93.78±8.15	90.84±13.78	0.036
Role limitation-physical	81.06±30.19	72.47±35.71	0.039
Role limitation-emotional	50.73±42.16	38.65±44.04	0.026
Energy	54.08±19.70	47.94±20,26	0,015
Mental health	67.58±14.59	63.36±18.65	0.044
Social function	74.54±21.37	70.75±23.94	0.184
Bodily pain	80.11±12.54	74.40±17.67	0.003
General health	66.83±16.17	61.47±19.86	0.018
Physical component score	80.45±11.75	74.79±16.32	0.002
Mental component score	61.73±19.21	55.11±20.31	0.008

Short form-36: SF-36

statistically significant difference between the groups in terms of social function ($p=0.184$).

Discussion

In present study, hypermobility was detected in 46.7% of the participants. Pain prevalence in different body regions in the past 12 months did not differ between participants with and without hypermobility. In terms of QoL; physical function, role limitation due to physical and emotional problems, energy, mental health, bodily pain, general health, physical and mental component scores of hypermobile participants were lower than the participants without hypermobility.

The prevalence of GJH was reported between 10-30% in some studies examining the adult population (26,27). This rate was 43% in Nigeria (28), and 38.5% in females and 25.4% in males in Iraq (29). In UK, it was reported as 34% between the ages of 20-30 and 18.4% in those aged 60 and above in Caucasian female twins (30). In American university students; Reuter

and Fichthorn (12) reported two different rates as 12.5% and 18.2% with two separate cut-off values, 5 and 4, respectively, and Russek and Errico (31) reported as 26.2% when the cut-off value was approved as 5. In Turkey, the prevalence was shown between 12.4% and 22% in females; between 6.1% and 7.7% in males in pediatric and adult populations (32-35). In the only study conducted in Turkey with different cut-off values, the prevalence in university students was shown as 25.9% and 34.9% when cut-off values were approved as 5 and 4, respectively (36). Conspicuously, it was observed that different results were obtained with different cut-off values in the same populations. Considering the decrease in range of motion of the joints with aging, it has been thought that using different cut-off values for adults and children when diagnosing GJH can prevent false positive and false negative results. Due to the diagnostic complexity and different demographic characteristics, the results regarding the prevalence of GJH are quite different in the literature. In present study, the cut-off value of Beighton criteria was determined as 4. Accordingly, the prevalence of GJH was 46.7%, and its prevalence was higher in females (50%) than males (40.7%), similar to the literature. The prevalence of GJH detected was higher than the studies conducted with similar age groups in the literature. This may be due to the fact that the majority of the population in this study is women. Also, the determined cut-off value may also have increased the calculated prevalence levels.

Hypermobile individuals are thought to be more prone to musculoskeletal diseases, such as subluxations, dislocations, meniscal and muscle tears, degenerative joint diseases, synovitis, arthralgia, myalgia, spondylolysis/spondylolisthesis, due to the increased joint laxity with the changes in neuromuscular reflexes and the decrease in proprioception (5,37,38). The most dominant complaint of these individuals is pain. Pain may be acute, localized and recurrent as a result of the musculoskeletal problems mentioned above, or it may be presented as chronic widespread pain (39). Its relationship with chronic pain is thought to be related to the possible pathogenic mechanisms, such as repetitive microtrauma, sensitization of pain receptors, amplification of pain signals, and central hyperexcitability (5). Musculoskeletal pains are observed more frequently in weight-bearing joints, such as knees and ankles due to biomechanical loading, impaired proprioception, decreased muscle strength and endurance (40,41). In adolescents, hypermobility was reported to be associated with shoulder, knee, foot-ankle pain, while not associated with neck, upper back, upper arm, elbow, wrist-hand and hip pain (42). Seçkin et al. (34), reported that 30.7% of hypermobile high school students had low back pain, 16.8% had arthralgia, and 13.9% had sprain complaints. In patients with BJHS, Albayrak et al. (43), observed low back (32.2%) and knee pain (27.8%) most frequently. In another study on BJHS; upper back pain, sprain/strain, and muscle pain ratios were reported 60-74%, 66-74% and 54-72%, respectively (44). When the patients with BJHS and hypermobile type EDS were evaluated, the most common sites of pain were reported as neck (90.4%), shoulder (80.8%), knee and ventral side of the leg (76.9%) (25). Russek and Errico (31) reported that upper back pain, sprain,

and stress fractures were more common in those with BJHS than in those without BJHS, but there was no difference in the injury prevalence between those with and without GJH, and so hypermobility alone was not associated with symptoms. In present study, upper and lower back were observed as the regions that pain was mostly localized rather than weight-bearing lower extremity joints in hypermobile individuals. However, compared with healthy individuals, no statistically significant relationship was found between joint hypermobility and painful areas in whole body. The frequencies of upper back pain (74.3%) and lower back pain (69.9%) were also found to be high in healthy participants. This may be related to the situation that lower and upper back are frequently observed pain regions in normal populations regardless of hypermobility as reported in epidemiological studies (45). Besides, the studies in the literature were mostly conducted with BJHS and EDS types, while the present study was conducted with the participants from the healthy population rather than the patients with HCTDs, and this might also affect the results obtained. In addition, recall bias should be considered, due to the retrospective questioning of pain.

Hypermobility is associated with acute and chronic pain as well as various extra-articular problems, such as autonomic and psychiatric problems that affect physical functions, and cause chronic fatigue, sleep problems, and various psychosocial problems. This situation affects the QoL negatively (1). Voermans et al. (46), emphasized that pain was more common in the patients with hypermobile type EDS, and associated with deterioration in sleep quality and functional loss in daily living activities. Studies evaluating QoL with SF-36 generally yielded lower scores than the normal population. In a study on hypermobile type EDS, all scores of SF-36 were found to be lower than the normal population (47). Albayrak et al. (43), found the scores of physical function, physical and emotional role, and mental component lower in patients with BJHS. In the same study, it was observed that lower physical and mental component scores were associated with higher fatigue level and decreased sleep quality (43). Castori et al. (48), detected a lower bodily pain score in those with classical and hypermobile type EDS compared to the normal population which might be associated with chronic pain. In the same study physical function, physical role, general health, vitality and social function scores also created significant differences compared to the normal population (48). In a study conducted with a group, consisting of patients with BJHS and hypermobile type EDS, all physical scores except mental health and mental component scores were found to be lower (25). It is observed that the studies evaluating the QoL in the literature mostly focus on patients with a diagnosis of EDS and BJHS. In present study, the QoL of individuals with GJH was evaluated. Consistent with the literature, lower physical and mental scores were remarkable in the results. The major differences were found in bodily pain and physical component scores. Hypermobile participants did not differ from the participants without hypermobility only in terms of social function. Although there were no differences between the groups in terms of painful areas, the fact that bodily pain and physical component scores of SF-

36 were affected more prominently suggested that pain and accompanying physical limitations had a significant effect on hypermobile individuals' QoL. In addition, low mental scores may support that hypermobility is not only a physical problem. In the literature, it was presented that various psychiatric problems could accompany with hypermobility. Therefore, the effects of chronic fatigue, sleep problems, and psychiatric problems on QoL should be evaluated in hypermobile individuals. The fact that social functions were not affected by hypermobility might be due to the selection of the participants from healthy university students rather than a group of patients with EDS subtypes.

Study Limitations

The most important limitations of this study were that; it was limited to a specific population, and the majority of the population was already female participants where hypermobility is common. The prevalence of hypermobility in young adult population may be overestimated due to the significantly higher number of female participants. The results obtained mostly reflected young adults and could not be generalized to the whole population. Therefore, longitudinal studies with wider age ranges are needed. In addition to these, especially considering the updates in 2017, age-based assessments in the diagnosis of hypermobility can reflect the society better. Besides, evaluating the sleep quality, fatigue, and physical activity level in hypermobile individuals may be essential in investigating the underlying causes of deterioration in QoL.

Conclusion

Although hypermobility is a common condition in the population, it can be neglected due to the wide symptom scale, the nonspecific nature of some symptoms, and the belief that it is a benign condition that does not cause any problems. Therefore, it is important to recognize hypermobile individuals and be aware of the clinical importance of the problems that may be encountered in the future. Hereby, there can be an opportunity to support them with education and exercise programs, including strengthening and proprioception. Early recognition of hypermobility can prevent the inveteracy of pain. Since hypermobility is a common feature of HCTDs, it is important to evaluate the family history and other accompanying symptoms for the definition and prognosis of BJHS and other HCTDs. These individuals, for whom we often seek solutions to their pain problems in daily practice, should also be evaluated for sleep quality, physical activity status, and participation in daily life activities. Thus, it can be aimed not only to solve the pain and other symptoms, but also to increase their QoL.

Ethics

Ethics Committee Approval: Bezmalem Vakif University Non-Interventional Research Ethics Committee (date: 12/07/2018/ no: 11200).

Informed Consent: An informed consent form was obtained from each participant.

Peer-review: Externally peer reviewed.

Authorship Contributions

Concept: E.Z., Y.A., Design: E.Z., Y.A., Data Collection or Processing: E.Z., F.M., M.P., Analysis or Interpretation: Ç.A.K., M.Z., Literature Search: Ç.A.K., E.Z., F.M., M.P., Ö.K., H.B., Y.A., Writing: Ç.A.K., E.Z., M.Z.

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