

Benign Joint Hypermobility Syndrome in Patients with Attention Deficit/Hyperactivity Disorders

Dikkat Eksikliği/Hiperaktivite Bozukluğu olan Hastalarda Benign Eklem Hipermobilitate Sendromu

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Objectives: In this study, we determined whether attention deficit/hyperactivity disorder (ADHD) is accompanied by benign joint hypermobility syndrome (BJHS) and evaluated the incidence of BJHS in patients with ADHD.

Patients and methods: This study was performed in the Ufuk University Faculty of Medicine, Departments of Physical Medicine and Rehabilitation and Psychiatry between November 2007 and February 2008. A total of 54 patients (45 males, 9 females; mean age 9.7±2.2 years; range 7 to 12 years) with ADHD and 36 healthy controls (19 males, 17 females; mean age 9.8±2.3 years; range 7 to 12 years) were included in the study. The sociodemographic and clinical characteristics of the individuals and the joint pain severity in the last month were recorded. Pain severity was assessed with the Faces Pain Scale-Revised (FPS-R) and the Likert Pain Scale. The joint hypermobility was evaluated by using Beighton scores.

Results: Benign joint hypermobility syndrome was found in 31.5% of the patients with ADHD and 13.9% of the individuals in the control group, and the difference was statistically significant ($p=0.05$). There were no statistically significant differences between the groups in FPS-R or Likert Pain Scale scores ($p>0.05$). A statistically significant increase was observed in the Beighton total score in ADHD patients compared with the control group ($p=0.004$).

Conclusion: The results of this study support that joint hypermobility may be associated with ADHD, and this condition should be taken into consideration in assessing the complaints of patients with ADHD-related musculoskeletal symptoms.

Key words: Hyperactivity; hypermobility; joint; pain.

Amaç: Bu çalışmada dikkat eksikliği/hiperaktivite bozukluğuna (DEHB) benign eklem hipermobilitate sendromunun (BEHS) eşlik edip etmediği belirlendi ve DEHB'li hastalarda BEHS'nin insidansı değerlendirildi.

Hastalar ve yöntemler: Bu çalışma Kasım 2007 - Şubat 2008 tarihleri arasında Ufuk Üniversitesi Tıp Fakültesi Fiziksel Tıp ve Rehabilitasyon ve Psikiyatri Anabilim Dallarınca yapılmıştır. Elli dört DEHB'li hasta (45 erkek 9 kız; ort. yaş 9.7±2.2 yıl; dağılım 7-12 yıl) ve 36 sağlıklı kontrol (19 erkek, 17 kız; ort. yaş ve 9.8±2.3 yıl; dağılım 7-12 yıl) çalışmaya dahil edildi. Bireylerin sosyodemografik ve klinik özellikleri ile son bir aydaki eklem ağrısı şiddeti kaydedildi. Ağrı şiddeti Revize-Yüz Ağrı Skalası ve Likert Ağrı Skalası ile değerlendirildi. Eklem hipermobilitesi Beighton skorları ile değerlendirildi.

Bulgular: Benign eklem hipermobilitate sendromu DEHB'li hastalarda %31.5, kontrol grubundaki bireylerde ise %13.9 oranında saptandı ve bu farklılık istatistiksel olarak anlamlı idi ($p=0.05$). Revize-Yüz Ağrı Skalası ve Likert Ağrı Skalası skorlarında gruplar arasında istatistiksel olarak anlamlı farklılıklar yoktu ($p>0.05$). Dikkat eksikliği/hiperaktivite bozukluğu olan hastalarda kontrol grubuna kıyasla Beighton total skorunda istatistiksel olarak anlamlı bir artış gözlemlendi ($p=0.004$).

Sonuç: Bu çalışmanın bulguları eklem hipermobilitatesinin DEHB ile ilişkili olabileceğini desteklemektedir ve DEHB'li hastaların kas iskelet sistemi ile ilişkili yakınmaları değerlendirilirken bu durumun göz önünde bulundurulması gerekmektedir.

Anahtar sözcükler: Hiperaktivite; hipermobilitate; eklem; ağrı.

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Attention-deficit/hyperactivity disorder (ADHD) is the most common psychiatric disorder in children with symptoms of inattention, hyperactivity, distractibility, and impulsivity. Patients with ADHD have cognitive complaints and difficulty concentrating, retaining short-term memory, and executing simple daily routines.^[1-6] Low performance levels in school, behavioral disruptions, and emotional liability are other signs of this condition.^[1,3,6,7] These children have poor self-esteem and a high risk of depression and anxiety because of adverse feedback from their teachers, friends, and family.^[1,3,6] The prevalence of ADHD is 5-10% in children and 4% in adults.^[3] The etiology of ADHD is still unknown. Pregnancy complications and genetic transmission are accused of being etiologic factors for this disorder.^[4,6,7]

Benign joint hypermobility syndrome (BJHS) is a musculoskeletal disorder characterized by excessive movement of the joints without a systemic rheumatological disease.^[8] This syndrome is seen in healthy patients suffering from arthralgia without any explanation, soft tissue rheumatism, joint laxity, and in healthy patients without Marfan and Ehlers-Danlos syndromes.^[9,10] The prevalence of BJHS varies from 5% to 38% depending on age, sex, and race.^[11,12] This syndrome is thought to be a hereditary connective tissue disorder resulting from a defect in collagen.^[13] Hypermobility and chronic pain in multiple joints are primary clinical signs of BJHS.^[14] An increased prevalence of psychological disturbances, such as anxiety and depression, has been found in patients with BJHS.^[15-17] Speech impairment, learning disabilities, poor coordination, delayed walking, and problems with handwriting tasks have also been reported in children with BJHS.^[14]

Many musculoskeletal findings have been reported in children with ADHD, including postural anomalies, chronic fatigue syndrome, widespread musculoskeletal pain, and fibromyalgia.^[5,7] Also, signs of ADHD have been reported in disorders associated with joint laxity.^[18-21] The aim of this study was to determine whether ADHD is accompanied by BJHS and to evaluate the incidence of BJHS in patients with ADHD.

PATIENTS AND METHODS

This study was performed in Ufuk University Faculty of Medicine, Departments of Physical Medicine and Rehabilitation and Psychiatry between November 2007

and February 2008. A total of 54 patients (45 boys, 9 girls; mean age 9.7 ± 2.2 years; range 7 to 12 years) with ADHD and 36 healthy controls (19 boys, 17 girls; mean age 9.8 ± 2.3 years; range 7 to 12 years) were included in the study.

The Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition (DSM-IV) and the Schedule for Affective Disorder and Schizophrenia for School-Age Children-Present and Lifetime version (K-SADS-PL) were used for diagnosing ADHD.^[22,23]

The DSM-IV has specific criteria that is required for diagnosis. These criteria include symptoms of inattention and hyperactivity/impulsivity. Six of nine characteristics of each symptom must be present and also be expressed for six months or more.^[3,7]

The K-SADS-PL is a semi-structured instrument developed by Kaufman et al.^[23] to screen the psychopathology in children and adolescents between the ages of six and 18. It screens the psychopathology by gathering information from both parents and their children. Mood disorder, psychotic disorder, anxiety disorder, disruptive behavioral disorder, elimination disorder, eating disorder, and tic disorder along with alcohol and other substance use disorders are the psychiatric conditions included in this instrument. The reliability and validity of the Turkish version of K-SADS-PL has previously been shown.^[24] After the psychiatric evaluation, patients with mental retardation, mood disorder, anxiety disorder, disruptive behavior disorder, cerebral palsy, epilepsy, brain disorder, or any other known genetic disorder were excluded from the study.

The sociodemographic and clinical characteristics of the individuals with joint pain (neck, shoulder, elbow, hand, low back, back, hip, knee, foot-ankle) and the accompanying pain severity in the last month were recorded. Pain severity was assessed with the Faces Pain Scale-Revised (FPS-R) scale and the Likert scale. The FPS-R is a scale consisting of six faces without smiles or tears. It is easy for children between the ages of six and 14 years to use and the results are correlated on a numerical scale from zero to 10.^[25]

Joint hypermobility was evaluated by using Beighton scores for diagnosing BJHS. The Beighton scores were: (i) Passive apposition of the thumbs to the flexor aspects of the forearm, (ii) passive dorsiflexion of the little fingers beyond 90° , (iii) hyperextension of the elbows beyond 10° , (iv) hyperextension of the knees beyond 10° , (v) forward flexion of the trunk with

Table 1. Demographic characteristics of the patients with attention deficit/hyperactivity disorder and the control group

	ADHD (n=54)			Control (n=36)			<i>p</i>
	n	%	Mean±SD	n	%	Mean±SD	
Gender							
Female	9	16.7		17	47.2		0.002
Male	45	83.3		19	52.8		
Age (year)			9.70±2.18			9.75±2.35	0.924
Education (year)			3.85±2.28			3.92±2.62	0.901

SD: Standard deviation; ADHD: Attention deficit/hyperactivity disorder.

the knees straight. The tests given are bilateral, and the scoring is on a scale ranging from zero to nine. Subjects who scored four or higher were considered to have BJHS.^[26]

The Ufuk University ethics committee approved the study, and informed consent forms were signed by the parents of the children.

The SPSS for Windows Release 15.0 version (SPSS Inc., Chicago, Illinois, USA) software was used for the statistical analysis. Descriptive statistics (mean ± standard deviation or median-minimum-maximum values) were used for determining the distribution of the age, sex, education, and joint pain of the patients. The Mann-Whitney U-test was used for comparing the numerical variables and the chi-square test for the categorical variables. A *p* value of 0.05 or less was considered significant.

RESULTS

The demographic and clinical characteristics of the subjects are given in tables 1 and 2. No significant differences were observed between the groups regarding age, gender, or education.

Benign joint hypermobility syndrome was found in a ratio of 31.5% patients with ADHD and 13.9% of the control group. The difference between ratios was found to be statistically significant (*p*=0.05). Girls comprised 33.3% of the cases with BJHS while 66.6% were boys.

There were no statistically significant differences in the FPS-R and Likert scales between the children with ADHD and the controls (*p*>0.05). The *p* values are given in tables 3 and 4. A statistically significant increase was observed in the Beighton total score in ADHD patients compared with the control group (*p*=0.012).

DISCUSSION

Attention deficit hyperactivity disorder is a psychiatric disorder characterized by attention deficit, hyperactivity, distractibility, and impulsivity leading to functional impairment in social, school, and occupational life.^[27] Joint hypermobility syndrome is a connective tissue disorder condition which includes the increased distensibility of joints in the absence of rheumatological disease.^[8] The signs of ADHD, for example hyperactivity or attention deficit, have been reported in diseases

Table 2. Clinical characteristics and Beighton scores of the patients with ADHD and the control group

	ADHD (n=54)			Control (n=36)			<i>p</i>
	n	%	Mean±SD	n	%	Mean±SD	
Neck pain	9	16.7		5	13.9		0.72
Shoulder pain	7	13		3	8.3		0.49
Elbow pain	2	3.7		2	5.6		0.67
Hand pain	14	25.9		5	13.9		0.17
Low back pain	13	24.1		6	16.7		0.39
Back pain	9	16.7		3	8.3		0.25
Hip pain	3	5.6		0	0		0.15
Knee pain	16	29.6		7	19.4		0.27
Foot-ankle pain	14	25.9		6	16.7		0.30
Beighton score			2.41±1.82			1.39±1.87	0.004

SD: Standard deviation; ADHD: Attention deficit/hyperactivity disorder.

Table 3. Mean and median values of the Faces Pain Scale-Revised of the patients with attention deficit/hyperactivity disorder and the controls

	ADHD (n=54)			Control (n=36)			p
	Mean±SD	Median	(min.-max.)	Mean±SD	Median	(min.-max.)	
Faces Pain Scale-Revised							
Neck pain	0.52±1.35	0	(0-6)	0.75±2.19	0	(0-10)	0.814
Shoulder pain	0.63±1.93	0	(0-10)	0.44±1.79	0	(0-10)	0.502
Elbow pain	0.15±0.76	0	(0-4)	0.22±1.04	0	(0-6)	0.678
Hand pain	1.11±2.25	0	(0-10)	0.56±1.55	0	(0-6)	0.173
Low back pain	1.19±2.47	0	(0-10)	1±2.46	0	(0-10)	0.474
Back pain	0.81±2.03	0	(0-8)	0.33±1.21	0	(0-6)	0.242
Hip pain	0.87±4.53	0	(0-30)	0±0	0	(0-0)	0.153
Knee pain	1.59±2.94	0	(0-10)	1.17±2.80	0	(0-10)	0.322
Foot-ankle pain	1.30±2.58	0	(0-10)	0.56±1.40	0	(0-6)	0.234

ADHD: Attention deficit/hyperactivity disorder; SD: Standard deviation.

characterized by joint laxity in case reports.^[19,20] In literature, there are several publications about musculoskeletal disorders, which accompany ADHD, but no study was found evaluating the frequency of joint hypermobility in patients with ADHD. To our knowledge, this is the first report on the relationship between ADHD and BJHS.

The association of BJHS with psychological diseases such as anxiety and depression has been reported in some publications.^[15-17] Bulbena et al.^[16] reported that the risk of encountering anxiety disorders, like panic disorder, agoraphobia, and simple phobia, increases in individuals with BJHS. In another study, 526 patients were evaluated by Bulbena et al.,^[17] and a significant increase in anxiety scores in patients with joint hypermobility; therefore, joint hypermobility was determined to be a predisposing factor for the anxiety. In contrast, Martín-Santos et al.^[28] concluded that patients with panic disorder and agoraphobia

were 16 times more likely to have joint hypermobility syndrome than healthy subjects. The frequency of joint hypermobility was found to be 67.7%.

In the study by Hofman et al.^[18] 13% of 30 school-age children with Marfan syndrome had learning disabilities, and 17% were determined to have ADHD. It is reported that learning disabilities are related to joint laxity. Hyperactivity and autistic retardation were reported with Lujan-Fryns syndrome, which is an X-linked mental retardation syndrome progressing with characteristics such as marfanoid habitus, joint laxity, pectus excavatum, and kyphosis.^[20]

In our study, we aimed to determine the BJHS frequency in children with ADHD and we found BJHS in 31.5% of the patients with ADHD and 13.9% in the control group. The difference was found to be significant. Also, there was a statistically significant increase in the Beighton total score in ADHD patients

Table 4. Mean and median values of the Likert Pain Scale of the patients with attention deficit/hyperactivity disorder and the controls

	ADHD (n=54)			Control (n=36)			p
	Mean±SD	Median	(min.-max.)	Mean±SD	Median	(min.-max.)	
Likert Pain Scale							
Neck pain	0.22±0.57	0	(0-3)	0.25±0.69	0	(0-3)	0.804
Shoulder pain	0.24±0.67	0	(0-3)	0.19±0.74	0	(0-4)	0.511
Elbow pain	0.06±0.30	0	(0-2)	0.11±0.52	0	(0-3)	0.670
Hand pain	0.43±0.81	0	(0-3)	0.19±0.52	0	(0-2)	0.157
Low back pain	0.52±1.04	0	(0-4)	0.36±0.93	0	(0-4)	0.402
Back pain	0.31±0.77	0	(0-3)	0.14±0.48	0	(0-2)	0.247
Hip pain	0.07±0.26	0	(0-1)	0±0	0	(0-0)	0.097
Knee pain	0.63±1.17	0	(0-4)	0.50±1.15	0	(0-4)	0.425
Foot-ankle pain	0.44±0.90	0	(0-6)	0.25±0.64	0	(0-3)	0.352

ADHD: Attention deficit/hyperactivity disorder; SD: Standard deviation.

compared with the control group. These results lead to the conclusion that there is a pathogenic relationship between the development of the central nervous system and connective tissue diseases.

Benign joint hypermobility syndrome is more frequent in girls.^[29] However, in this study, most of the BJHS cases were found in boys. A higher frequency of ADHD in males is the reason for that result.^[1]

It is known that psychiatric disorders increase pain prevalence and severity.^[30] The recurrent and chronic pain prevalence in children is reported as 15-20%.^[31] It is known that in children, repeating pain can give rise to not only psychiatric problems, including anxiety and depression, but also to restrictions in daily living, such as school activities, social relations, and sleep patterns.^[30,32] The observation of pain-related health complaints in children with ADHD has been shown in some studies.^[32-34] It has been reported that repeating abdominal pain is observed twice as much in children with ADHD.^[32] Cuffe et al.^[35] reported that headaches were more prevalent in children with ADHD. In a study by Kaplan et al.^[34] headaches and leg cramps were observed more frequently in children with ADHD than in healthy children. There is no doubt that a child with this kind of repeating pain should be evaluated on the basis of ADHD. Young and Redmond^[5] reported that somatic disorders, like chronic fatigue syndrome and fibromyalgia, can develop in children with ADHD when they become adults. However, we could not find any study associated with pain complaints of the musculoskeletal system of children with ADHD. In our study, we investigated whether there was any difference in joint pain between children with ADHD and healthy children. There was no statistically significant difference in the FPS-R and Likert scales between children with ADHD and the controls. This result may be due to the inability of children with ADHD to express their pain correctly because of inattention and the inability to concentrate. We could reach more objective results if the parents would use a diary while questioning the child about the pain. On the other hand, no difference was reported in acute or in chronic pain severity between children with ADHD and healthy children in the study of Scherder et al.^[36] This finding supports our study.

There are no accepted diagnostic standards for evaluating BJHS, and there is not enough literature showing the reliability of the tests used for diagnosis.^[37] In this study, Beighton scores were used for diagnosing

BJHS. If another test was used, maybe the results would be different. Another limitation of this study is the relatively small sample size for evaluating the incidence of BJHS.

In conclusion, our study results support that joint hypermobility seems to be associated with patients with ADHD, and this should be taken into consideration in assessing patients' musculoskeletal complaints. However, more studies with a higher number of children are needed to determine the association between musculoskeletal pain and ADHD.

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