

The Relationship Between Generalized Joint Hypermobility and Cervical Disc Degeneration, Neck Pain: A Multidisciplinary Clinical Study

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ABSTRACT

Objective: Generalized joint hypermobility (GJH) is a condition of the connective tissue, which has movement ability beyond the normal limit of synovial joints. Its effects on disc degeneration and neck pain are not fully known. The aim of the present study was to determine the relationship between GJH and cervical disc degeneration that is detected in magnetic resonance imaging (MRI) and also neck pain.

Methods: Cases aged between 20 and 50 years who were admitted to outpatient clinics with neck and arm pain were included in the study. Their cervical MRIs were evaluated. Beighton score was used to evaluate these cases for GJH, and they were also evaluated prospectively using the Visual Analog Scale (VAS) for pain and Neck Disability Index (NDI) for disability.

Results: Of the 75 cases, 59 (78.7%) were female, 16 (21.3%) were male, and GJH was found in 15 (20%). There was no statistically significant difference in the values of Miyazaki grade parameters in all cervical disc levels and VAS and NDI values between the patients with and without GJH ($p>0.05$).

Conclusion: This result suggests that GJH may not be a single risk factor for cervical disc degeneration, and VAS and NDI values increase in patients aged between 20 and 50 years.

INTRODUCTION

Generalized joint hypermobility (GJH) is an observed phenomenon that may be inherited or very rarely acquired, which is defined as the ability to move the synovial joints beyond their normal range of motion (ROM).^[1-3] Although its prevalence varies according to gender, age, and racial characteristics, it reaches to 15% in adults and 30% in childhood.^[1,4] The Beighton scoring system (Annex 1) is used to detect GJH.^[5,6] Cases with a score of ≥ 4 according to this scoring system are classified as hypermobile, and these criteria can be determined by

five simple maneuvers that take a maximum of 1 min for physicians to perform.^[7,8] Hypermobility (HM) plays an important role because of comorbid chronic pain complaints, premature osteoarthritis, osteoporosis, and vascular and spinal disorders (scoliosis, spondylosis, and spondylolisthesis), and it may be overlooked especially when not searched for.^[8,9]

The prevalence of chronic neck pain, which is a serious public health problem, has been reported to be between 30% and 50%.^[10-12] The effects of HM on cervical disc degeneration and neck pain, of which the prevalence is more than expected, are not known exactly. Nowadays,

magnetic resonance imaging (MRI) is the most valid tool to evaluate disc pathology clinically.^[13] In this evaluation method, it is very important to use standard and reliable terminology in the evaluation of cervical disc degeneration, and the classification developed by Miyazaki et al.^[14] fulfills this requirement. To the best of our knowledge, there are not enough studies investigating the effect of HM on cervical intervertebral disc (IVD) degeneration. Therefore, the effect of GJH on the severity of disc degeneration and cervical pain was investigated based on cervical MRI findings by including cases aged between 20 and 50 years.

MATERIALS AND METHODS

The study was conducted with the approval of the ethics committee of İstanbul Fatih Sultan Mehmet Training and Research Hospital. The aim of the present study was to determine the effect of joint HM on cervical disc degeneration in patients aged between 20 and 50 years. Patients who had neck and/or arm pain were evaluated prospectively in the present study. The collected data were compared with a case-control study. Patients who had previously experienced trauma, neck surgeries, tumors, rheumatic diseases or infection, stroke, dementia, or intensive care treatment and those who were in the menopausal period were excluded from the study.

Table 1. Beighton scoring system*

	Right	Left
Passive dorsiflexion of five metacarpal joints beyond 90°	1	1
Passive apposition of the thumb to the flexor aspect of the forearm	1	1
Passive hyperextension of the elbow beyond 10°	1	1
Passive hyperextension of the knee beyond 10°	1	1
Hands rest flat on the floor when standing upright with knees in extension	1	
Total	9	

*In adult patients, the cut-off value of Beighton score is 4 points from a total of 9 points, and cases whose scores were ≥ 4 points were considered as patients with HM.

In the study period, 75 of the 278 patients who presented to the BS and FTR outpatient clinics with neck and/or arm pain were in compliance with our criteria. Demographic data, waist circumference values, and fingertip-to-floor (FTF) distance were recorded. Body mass index (BMI) was calculated, and cervical MRIs obtained within the last 6 months were evaluated in patients who were eligible for the study and filled in the survey questionnaire forms in outpatient clinics. The Beighton scale scores (Table 1) were used for the HM study in these cases.^[5] Cases with a score of ≥ 4 according to the Beighton HM scoring system were classified as hypermobile. The severity of neck and/or arm pain was evaluated by Visual Analog Scale (VAS), and clinical results were evaluated by the Turkish version of the Neck Disability Index (NDI).^[15-18]

The evaluations of the MRIs obtained within the last 6 months were performed by an expert radiologist (EDC MD) blinded to the study. In these cervical MRIs of the cases, IVD degeneration at C2-3, C3-4, C4-5, C5-C6, C6-C7, and C7-Th1 levels was classified using the Miyazaki grading system.^[14] In this grading system, disc degeneration is evaluated into five grades (Grades I-V) considering disc density and height (Table 2). Mean \pm standard deviations related to these five grades were calculated for each case, and the values were compared between the two groups.

Statistical analysis

Data were evaluated using IBM SPSS Statistics 22 (SPSS IBM, Turkey) programs for statistical analysis. Shapiro-Wilk test was used to study parameters with normal distribution. Descriptive statistical methods (mean, standard deviation, and frequency) were used for the evaluation of data. Student's t test was used for the intergroup comparison of quantitative data with normal distribution. Mann-Whitney U test was used for the intergroup comparison of parameters without normal distribution. Fisher's exact test and Yates continuity correction test were used for the comparison of qualitative data. A p value < 0.05 was accepted as statistically significant.

RESULTS

Of the 75 cases who met the inclusion and exclusion criteria, 16 (21.3%) were male, and 59 (78.7%) were female. The mean age of the study group was 37.61 ± 7.89 years. Of the 75 cases, 15 had HM (Group A), and 60 had no HM

Table 2. Miyazaki classification of disc degeneration

Grade	Nucleus signal intensity	Nucleus structure	Distinction of nucleus and annulus	Disc height
I	Hyperintense	Homogenous white	Clear	Normal
II		Inhomogeneous with horizontal band, white		
III	Intermediate	Inhomogeneous gray to black	Unclear	Normal to decreased
IV		Inhomogeneous gray to black	Lost	
V	Hypointense	Inhomogeneous gray to black		Collapsed

Table 3. Evaluation of general characteristics according to the presence of generalized joint hypermobility

	Group A	Group B	Total	p
	Mean±SD	Mean±SD	Mean±SD	
Age (year)	32.87±8.53	37.37±7.95	36.47±8.21	¹ 0.057
Gender, n (%)				
Male	1 (6.7)	15 (25)	16 (21.3)	³ 0.168
Female	14 (93.3)	45 (75)	59 (78.7)	
Waist circumference (cm)	83.07±11.83	91.77±11.29	90.03±11.85	¹ 0.010*
Body mass index	22.66±3.83	26.59±4.52	25.81±4.64	¹ 0.003*
Fingertip-to-floor distance test, n (%)				
Yes	9 (60)	11 (18.3)	20 (26.7)	³ 0.002*
No	6 (40)	49 (81.7)	55 (73.3)	
Employment status, n (%)				
Employed	8 (53.3)	33 (55)	41 (54.7)	² 1.000
Unemployed	7 (46.7)	27 (45)	34 (45.3)	
Smoking status				
Smoker	4 (26.7)	19 (31.7)	23 (30.7)	³ 1.000
Non-smoker	11 (73.3)	41 (68.3)	52 (69.3)	
Additional disease, n (%)				
Yes	6 (40)	19 (31.7)	25 (33.3)	² 0.759
No	9 (60)	41 (68.3)	50 (66.7)	

¹Student's t-test. ²Yates continuity correction. ³Fisher's exact test. *p<0.05. SD: Standard deviation.

Table 4. Evaluation of study parameters according to the presence of generalized joint hypermobility

	Group A	Group B	Total	p
	Mean±SD	Mean±SD	Mean±SD	
Visual Analog Scale (median)	5.87±1.36 (6)	6.4±1.77 (6.5)	6.29±1.7 (6)	¹ 0.343
Neck Disability Index	49.53±18.55	54.62±17.84	53.6±17.98	² 0.331

¹Mann-Whitney U test. ²Student's t-test. ³Fisher's exact test. *p<0.05. SD: Standard deviation.

Table 5. Evaluation of Miyazaki grading parameters according to the presence of generalized joint hypermobility

Miyazaki grade	Grup A	Grup B	Total	p
	Mean±SD (median)	Mean±SD (median)	Mean±SD (median)	
C2-3	3.31±1.11 (4)	3.18±0.96 (3)	3.2±0.98 (3)	0.373
C3-4	3.62±0.65 (4)	3.13±0.97 (3)	3.22±0.94 (3)	0.113
C4-5	3.69±0.75 (4)	3.23±1.1 (3)	3.32±1.05 (3)	0.207
C5-6	3.77±0.6 (4)	3.54±1.35 (4)	3.58±1.24 (4)	0.892
C6-7	3.23±0.83 (3)	2.91±1.21 (3)	2.97±1.15 (3)	0.444
C7-T1	2.25±0.5 (2)	2±0.82 (2)	2.03±0.78 (2)	0.634

Mann-Whitney U test. SD: Standard deviation.

(Group B) (Table 3). In addition, 1:4 equivalence design was used in comparison of cases with and without HM. There was no difference between these groups with respect to age, gender, smoking, previous diseases, and study status; however, a difference with respect to BC, BMI, and FTF data was remarkable (Table 3). The mean VAS scores were 5.87±1.36 in Group A and 6.4±1.77 in Group B.

However, no statistically significant difference was found between the two groups with respect to VAS density and NDI values (Table 4). In both groups, there was no difference in Miyazaki grades between cervical disc levels (Table 5). The degree of disc degeneration in Group A was higher than that in Group B in all examined disc levels, but this difference was not statistically significant.

DISCUSSION

Despite the prevalence of HM in the community, its clinical effects are not fully understood, and there are not enough studies investigating the effect of cervical IVD degeneration and neck pain. The findings of our study on cases aged between 20 and 50 years revealed that HM did not cause neck pain and cervical disc degeneration in this age range (Tables 4 and 5). Since aging may complicate the evaluation of neck pain that may be directly or indirectly caused by HM, cases in the premenopausal period aged <50 years were included in our study.

However, to the best of our knowledge, there is only one study investigating the effect of HM on cervical disc degeneration and neck pain in patients who were not operated using the Miyazaki scoring system in the literature. Although the results of Lee et al.^[19] differed from our study, they included only cases in the 20–30 age group. With aging, the joint ROM decreases, which is true for hypermobile joints.^[20] In the literature, it is stated that disc degeneration is a progressing process with age. Although the majority of adults aged >30 years have a structural degeneration on one or more discs, degeneration in these cases is not always accompanied by pain.^[13,21–24]

Furthermore, cervical disc herniation is usually seen in cases aged >40 years, different from lumbar disc herniation.^[25,26] Adams et al.^[27] showed that cadaveric discs aged >50 years are damaged even by minor traumas.

It has been suggested that impaired proprioception in HM has increased the tendency for sports injuries and thus joint injuries.^[8,28] In a meta-analysis by Pacey et al.,^[29] it has been shown that knee injuries are more common than ankle injuries in hypermobile cases. In fact, knee joint is more susceptible to contact injury, and the joint ROM is greater than the ankle and remains between the relatively stable and mobile regions.

Decreased range of movement in one region and increased mobility of the other end due to HM may lead to faster and more severe degeneration of the in-between joint. As a matter of fact, it has been reported that the restriction of motion by posterior lumbar interbody fusion in HM cases leads to overload and movement in the neighboring segment, and as a result, it is reported that adjacent segment disease (ASD) occurs more easily and frequently in these cases.^[28,30] The study also confirmed this hypothesis, and it was shown that HM did not adversely affect fusion rate or clinical outcome (pain intensity or functional status) but had a negative effect on ASD.^[31] The same problem occurs after cervical stabilization. As a matter of fact, it has been reported that ASD can develop after anterior cervical discectomy and fusion (ACDF), and that male patients have higher risks for secondary surgery.^[32–35]

In the meta-analyses, the incidence was reported to be lower for the second surgery in the cervical disc arthroplasty group than that in the ACDF group, and cervical artificial discs were preferred to reduce or delay the oc-

currence of ASD.^[36–39] Currently, MRI is used as the most accurate non-invasive diagnostic tool that can detect the pathology of IVD at the earliest stage of degeneration.^[14,23,37,40] Degenerative changes in the disc are more frequently related to the reduction of water content.^[14,41] In the Miyazaki^[14] scoring system, for the evaluation of the degree of disc degeneration, the classification was based on both disk signal intensity and disc height categories.

Considering that the low signal intensity observed in disc degeneration in MRI has been shown to be mostly related to dehydration, disc dehydration in normal hypermobile cases will not increase under normal conditions; therefore, detection of increased degeneration in cervical MRI of these cases should not be expected. In addition, it has been shown that HM has a protective effect on osteoarthritis in hand joints where the joint movement is intense. It is also suggested that increased joint motion in traumatic joints may not be the cause of degeneration alone.^[42] Although HM is not referred to as a disease, the predominant complaint is usually widespread and long-lasting pain, and it is also the most common cause of unexplained joint pains.^[2,8] However, in our study, we could not find any statistically significant difference not only between disc degeneration and HM but also between VAS and NDI values (Table 4).

The capsular ligaments of facet joints in the cervical spine are the main stabilizing structures. Steilen et al.^[12] reported that capsular ligament laxity is the main source of chronic neck pain, and that this pain reflects instability. In addition, Hogg-Johnson et al.^[11] reported that they could not find any evidence indicating that disc degeneration is a risk factor for neck pain. The results of all these studies show that neck pain is related to cervical pathologies caused by an overloading strain or trauma that may cause instability of capsular ligament rather than disc degeneration.

The fact that the cases we included in our study were selected among patients who were not exposed to trauma and lack of any difference between the workload of patients with and without HM (Table 3) may be the reason why any statistically significant difference was not found between the two groups with respect to neck pain.

One of the limitations of our study is that only disc degeneration was evaluated radiologically to reveal the relationship between HM and neck pain. Further studies should be performed on both the facet joints and capsular ligaments. Another limitation is that the study has been performed in a wide age range due to the small number of cases. Further studies on greater number of cases and separately in each decade may lead to more healthy outcomes.

CONCLUSION

In conclusion, there was no statistically significant relationship between GJH and cervical disc degeneration, VAS, and NDI. The tendency of patients with GJH to trauma will easily disrupt this harmony and will lead to faster and

more aggravated development of joint degeneration. For this reason, it is necessary to avoid rigid stabilization especially in patients with GJH before spinal surgery.

In addition, these cases should be informed about starting paravertebral muscle strengthening exercises at an early age to avoid all types of traumas and to reduce the tendency to trauma.

Ethics Committee Approval

Approved by the İstanbul Fatih Sultan Mehmet Training and Research Hospital Ethics Committee (date: 23.11.2017, number: 2017/ 35).

Peer-review

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Authorship Contributions

Concept: N.K., A.A. ??.; Design: N.K., A.A., P.A.; Data collection &/or processing: E.D.Ç., Ö.G.İ., N.K., A.E.S.; Analysis and/or interpretation: E.D.Ç., N.K.; Literature search: A.A., A.E.S., N.K., Ö.G.İ.; Writing: N.K., A.A.; Critical review: P.A., A.A., E.D.Ç.

Conflict of Interest

None declared.

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Generalize Eklem Hipermobilitesi ile Servikal Disk Dejenerasyonu ve Boyun Ağrısı İlişkisi: Bir Multidisipliner Klinik Çalışma

Amaç: Generalize eklem hipermobilitesi (GEH) sinovial eklemlerin normal sınırın ötesinde hareket yeteneğinin olduğu bir durum olup disk dejenerasyonu üzerine etkileri bilinmemektedir. Çalışmamızın amacı GEH ile manyetik rezonans görüntüleme (MRG) saptanılan servikal disk dejenerasyonu ve boyun ağrısı arasındaki ilişkiyi ortaya çıkarmaktır.

Gereç ve Yöntem: Beyin cerrahisi ve fizik tedavi ve rehabilitasyon polikliniklerine boyun ve/veya kol ağrısı yakınması ile baş vuran 20–50 yaş arasındaki olgular çalışmaya alındı. Kriterlere uyan olguların servikal MRG'leri değerlendirildi. Bu olguları GEH yönünden değerlendirmede Beighton skoru kullanıldı. Olgular ayrıca Vizüel Analog Skala (VAS) kullanılarak ağrı, Boyun Dizabilite İndeksi (BDİ) kullanılarak dizabilite yönünden ileriye dönük olarak değerlendirildiler.

Bulgular: Çalışma kriterlerine uyan 75 olgunun 59'u kadın (%78.7), 16'sı erkek (%21.3), yaş ortalamaları 37.61 ± 7.89 yıl idi. Olguların 15'inde GEH saptandı (%20), 60'ında GEH'ye rastlanılmadı (%80). GEH görülenlerle görülmeyenler arasında servikal disk düzeylerinde Miyazaki grade parametreleri açısından istatistiksel olarak anlamlı bir farklılık bulunmadı ($p > 0.05$). Aynı şekilde gruplar arasında VAS ve BDİ değerleri açısından da istatistiksel olarak anlamlı bir farklılık yoktu ($p > 0.05$).

Sonuç: Bu sonuç 20–50 yaş aralığındaki olgularda, normal şartlar altında, GEH'nin servikal disk dejenerasyonu ile VAS ve BDİ artışında tek başına bir risk faktörü olmayabileceğini düşündürmüştür.

Anahtar Sözcükler: Boyun ağrısı; boyun dizabilite indeksi; disk dejenerasyonu; generalize eklem hipermobilitesi; servikal omurga; Vizüel Analog Skala.