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INVESTIGATION OF THE RELATIONSHIP AMONG TEMPOROMANDIBULAR JOINT SUBLUXATION, JOINT SPACE AND ARTICULAR EMINENCE INCLINATION IN BENIGN JOINT HYPERMOBILITY SYNDROME

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ABSTRACT

Objective: *The objective of this study was a radiological examination of the articular eminence inclination and space of the temporomandibular joint in benign joint hypermobility syndrome (BJHS) patient who has temporomandibular joint subluxation (TMJS) and not subluxation.*

Material and Method: *A hundred patients (50 females, 50 males) with BJHS who admitted to our clinic with the complaint of TMJ were included in the study. The eminence inclination and the joint space were measured on the cone-beam computed tomography (CBCT).*

Results: *In our study, there was no statistically significant difference among subluxation and all measurements and age. However, there was the statistically significant difference only between gender and superior joint space (SS). The SS was higher in males (mean 3.60 mm) than females (mean 2.89 mm). There was found a statistically significant correlation between the SS and the age, bf (best-fit) angle, tr (top-roof) angle, posterior joint space (PS) (p<0.05).*

Conclusion: *In patients with BJHS with and without subluxation, joint spaces [anterior joint space (AS), SS, PS and articular eminence inclination (determined by bf and tr angle) do not differ when the mouth is in the closed position.*

Keywords: *Joint space, benign joint hypermobility syndrome, subluxation, temporomandibular joint.*

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BENİGN EKLEM HİPERMOBİLİTESİ SENDROMUNDA TEMPOROMANDİBULAR EKLEM SUBLÜKSASYONU İLE EKLEM ARALIĞI VE ARTİKÜLER EMİNENS EĞİMİ ARASI İLİŞKİNİN İNCELENMESİ

ÖZET

Amaç: Bu çalışmanın amacı, temporomandibular eklem sublüksasyonu (TMES) olan ve olmayan benign eklem hipermobilitesi sendromu (BEHS) hastalarında artiküler eminens eğiminin ve eklem aralığının radyolojik olarak incelenmesidir.

Materyal ve Metot: Çalışmaya, TME şikâyeti ile kliniğimize başvuran BEHS'li 100 hasta (50 kadın, 50 erkek) dâhil edildi. Artiküler eminens eğimi ve eklem aralığı, konik-ışınlı bilgisayarlı tomografi (KIBT) ile ölçüldü.

Bulgular: Çalışmamızda sublüksasyon ile tüm ölçümler ve yaş arasında istatistiksel olarak anlamlı bir fark yoktu. Ancak, yalnızca cinsiyet ve üst eklem boşluğu (SS) arasında istatistiksel olarak anlamlı bir fark vardı. SS, erkeklerde (ortalama 3,60 mm) kadınlardan (ortalama 2,89 mm) daha yüksekti. SS ile yaş, bf (en iyi) açı, tr (en üst) açı, posterior eklem boşluğu (PS) arasında istatistiksel olarak anlamlı bir ilişki bulundu (p<0,05).

Sonuç: BEHS ile sublüksasyonu olan ve olmayan hastalarda, ağız kapalı konumdayken, eklem aralıkları [anterior eklem aralığı (AS), SS, PS] ve artiküler eminens eğimi (bf ve tr açısı ile belirlenir) farklılık göstermez.

Anahtar kelimeler: Eklem aralığı, benign eklem hipermobilitesi sendromu, sublüksasyon, temporomandibular eklem.

INTRODUCTION

Benign hypermobile joint syndrome or benign joint hypermobility syndrome (BJHS) (or is known as generalized joint hypermobility) is a hereditary disorder.[1-5] BJHS is noted a benign, non-pathologic phenomenon.[6,7] Increased joint mobility during active and passive movements are described in BJHS. It consists due to variations in the conjunctive tissues of the body such as collagen, fibrils, elastin, and proteoglycans.[7] BJHS is found a higher incidence in women in percentage 5-10 of white people.[3,8] Benign BJHS consists in the Asians a greater prevalence than Europeans.[8,9] The first system to measure BJHS was introduced by Carter and Wilkinson in 1964, and since then has been repeatedly changed by various authors.[8,10-12] The evaluation test and criteria developed by Beighton have been used for many studies of hypermobility.[8,13] Taking into account the properties of BJHS, this representation is thought to affect all joints including the temporomandibular joint (TMJ). Many studies report that BJHS is a risk factor for the development of symptoms and signs of temporomandibular joint disorders (TMD).[14-16] It is believed that the TMJ is overloaded, depending on the loosening of the ligament in BJHS.[15,16] Thus, TMD is predisposed to orthopedic disorders such as degenerative joint disease, spontaneous dislocations, joint effusion, and myalgia.[4] The relationship between degenerative diseases of the temporomandibular joint (TMJ) and generalize joint hypermobility (GJH) was first described by Annandale.[17] Since then, several researchers have noticed a correlation between BJHS and TMD.[18-21] TMJ disease has not been known to be a complication of BJHS. One case of chronic TMJ subluxation (TMJS) in a 19-year-old man with a recognized diagnosis of Ehlers-Danlos syndrome has been noticed.[22] In 1981, Solberg suggested the possibility of a relationship between TMJ disease and systemic joint laxity, and Bates *et al*, in a preliminary study, suggested the presence of joint laxity in 11 of 15 women with internal derangements of the TMJ.[18,23] In the literature, temporomandibular joint diseases have been evaluated clinically, to the best of our knowledge, but no studies evaluating temporomandibular joint morphology radiologically have been found by cone-beam computed tomography (CBCT) in BJHS patients.

The objective of the present study was a radiological examination of the articular eminence inclination and space of the temporomandibular joint in BJHS patient who has TMJS and not subluxation by means of cone beam computed tomography.

MATERIAL AND METHOD

Patients

We designed study using the CBCT images from 100 patients (50 females; 50 males) with BJHS diagnosed by Beighton score[8] who suffered from TMJ to our clinic between June 2016 and September 2018. The study group consisted of 25 females and 25 males with subluxation, and the control group consisted of 25 females and 25 males without subluxation. In total, 200 TMJ was evaluated. The mean age of the study group was 27.64±9.95 years. Pregnant women or patients suffering from systemic rheumatoid complaints were excluded. Besides patients who

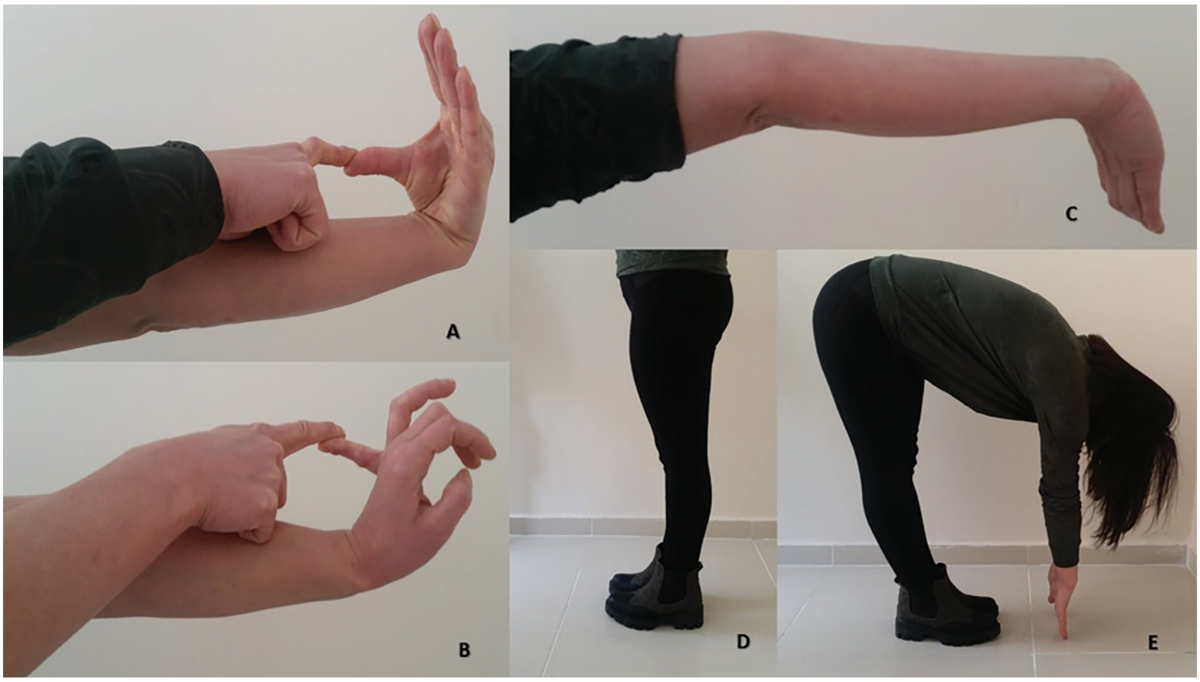

Figure 1. Assessment using the Beighton hypermobility scoring system: (A) passive apposition of the thumb to the flexor aspects of the forearm, (B) passive dorsiflexion of the fifth fingers beyond 90°, (C) hyperextension of the elbow more than 10°, (D) hyperextension of the knee more than 10°, (E) forward trunk flexion with the knees straight so that the palms of the hands rest easily on the floor. (A), (B), (C), and (D) were scored 2 points on both sides and (E) was scored 1 point.

have systemic/local bone disease or degenerative joint disease were not included in the study. All of the participants and their parents were informed of the nature of the investigation, and their acceptance was acquired. This study was approved by the Atatürk University Ethics Committee (Protocol no: 39/ 2016).

Assessment of TMJS

TMJS evaluation was performed both clinically according to the Research Diagnostic Criteria for temporomandibular disorders (RDC/TMD) and radiologically according to CBCT.[24,25] (The distance between the condyle-articular eminence was 4 mm or more).[26]

Assessment of BJHS

Using the criteria in the mobility index and the Carter-Wilkinson Scale, as modified by Beighton *et al.* to include: [8,10]

1. Passive apposition of the thumb to the volar aspect of the forearm: 2 points for both sides (Figure 1A)
2. Passive hyperextension more than 90° of the fifth finger: 2 points for both sides (Figure 1B)
3. Hyperextension of the elbow more than 10°: 2 points for each elbow (Figure 1C)
4. Hyperextension of the knee more than 10°: 2 points for each knee (Figure 1D)
5. Trunk flexion, so that the palms rest easily on the floor with the knees straight: 1 point (Figure 1E)

The scoring range was 0-9, with the higher scores denoting greater joint laxity. Each individual item was scored using a nominal scale in which “1” offers a positive mark and “0” shows a negative mark. All measurements were acquired bilaterally using a goniometer, with the exception of trunk flexion. Individuals who carried out four or more of the above maneuvers were considered to be hypermobile.[19,20,27,28]

Imaging Procedures

All patients underwent CBCT imaging with a NewTom 3G System (Quantitative Radiology, Verona, Italy). The 9-inch field of view included both TMJs. The X-ray parameters (kV, mA) were automatically detected from scout views by the NewTom 3G. The mean axial diameter was 1 mm, mean sagittal diameter was 2 mm, mean coronal diameter was 2 mm, scanning time was 36 s, and voxel size was 0.16 mm. All CBCT scans were performed with the patients resting in the supine position. The positioning of the patient's head was performed using two-light-beam markers. The vertical positioning light was aligned with the patient’s mid-sagittal line, which helped to keep the head centered relative to the rotational axis. The lateral positioning light was centered at the level of the condyles, indicating the optimized center of the reconstruction area. In addition, the head position was set in such a manner that the hard palate was perpendicular to the floor. The primary reconstruction was restricted to a region of approximately 1 cm in diameter with the TMJ at the center, and a series of axial views were automatically generated. On the axial TMJ images, a line perpendicular to the anteroposterior plane of the examined condyle was drawn, and parasagittal images were created by NNT Software (version 2.21; Quantitative Radiology). The thickness of the image slices was 1 mm, and the distance between the parasagittal slices was 1 mm.

Radiologic Measurements

The measurements belonging to the articular eminence were performed on the slices defined above. The points and planes used in this study were as follows:[29]

Points

- The point which the F line cut the eminence posterior surface: Ce
- The highest point of the condylar process: Cu
- Porion (the highest point of auditory meatus): Po
- The highest point of the fossa: R
- The lowest point of the articular eminence: T

Planes

- The best-fit plane of the articular eminence inclination connecting the Ce: Ebf (eminence inclination best-fit line) plane
- The plane passing through the points Cu and R: Etr (eminence inclination top-roof line) plane
- Frankfort horizontal: F
- The parallel line to the F passing through the point Cu: F1
- The parallel line to the F passing through the point R: F2

Measurements made according to the reference points described above are:

1. Angular Measurements

Using these planes, the eminence inclination was measured in two methods.

Method 1: The best-fit line method was the angle between Ebf and Frankfort horizontal: best-fit angle (bf angle) (Figure 2);

Method 2: The top-roof line method was the angle between Etr and Frankfort horizontal: top roof angle (tr angle) (Figure 3).

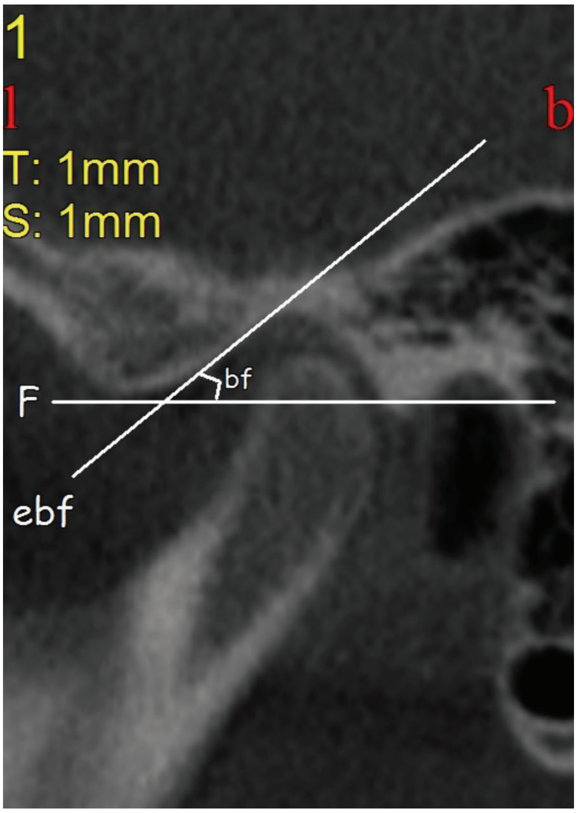

Figure 2. bf angle; the measurement of eminence inclination with the best-fit line method

Ebf: Eminence inclination best-fit line

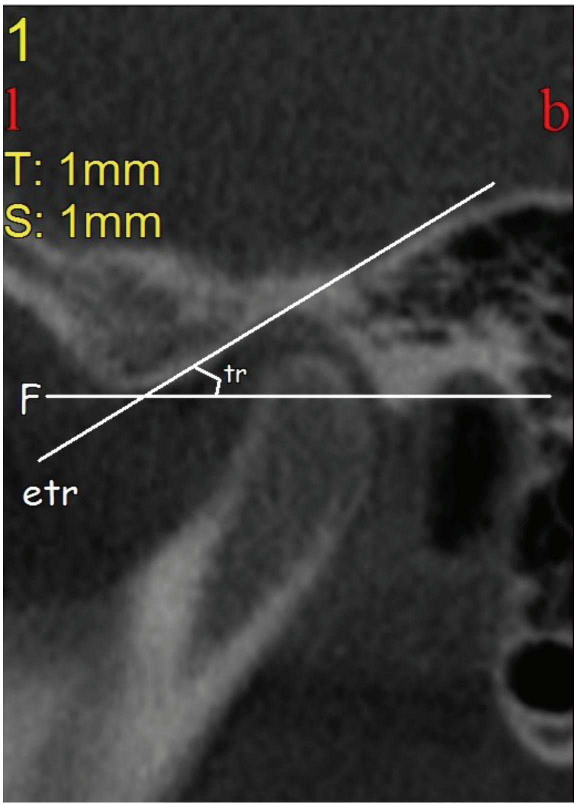

Figure 3. tr angle; the measurement of eminence inclination with the top-roof line method

Etr: Eminence inclination top-roof line

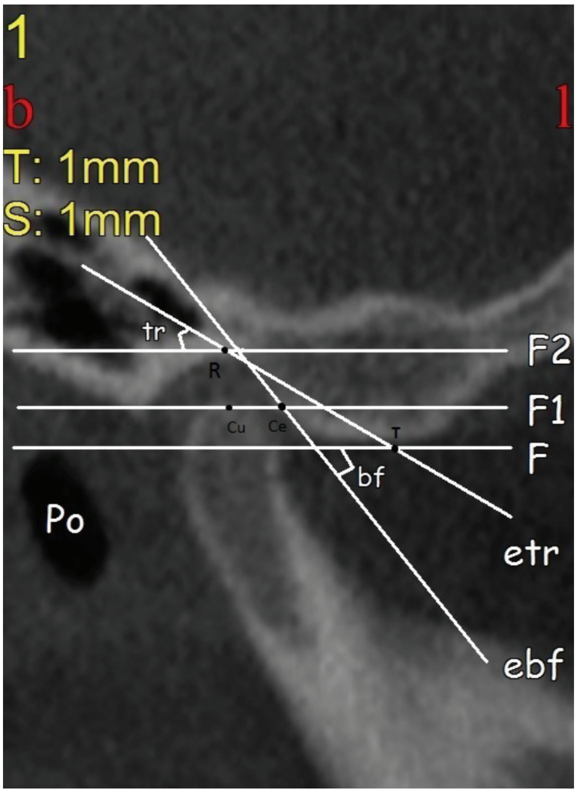

Figure 4. Lines and angles used in the study.
Ebf: Eminence inclination best-fit line; **Etr:** eminence inclination top-roof line

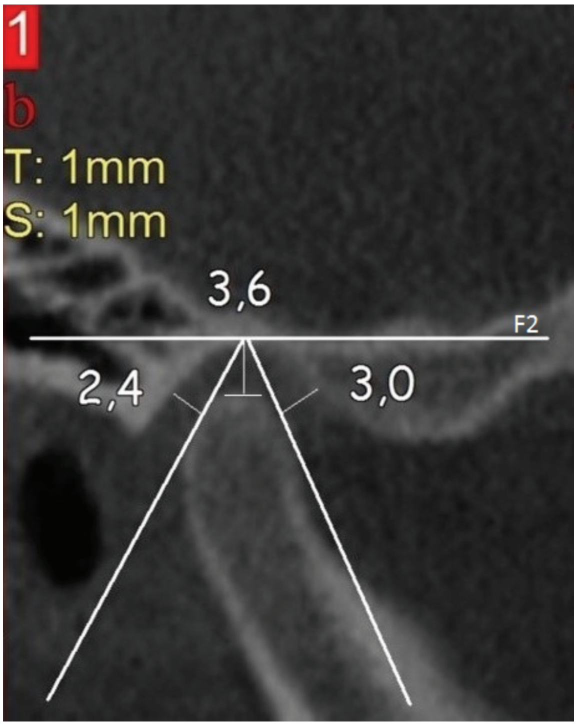

Figure 5. A case that showed measured posterior, superior and anterior joint spaces in the study.

These angles that we use in our work show the perpendicularity of the articular eminence and the depth of the mandibular fossa. In those who have upright articulation eminens and deep mandibular fossa, the angles are bigger.

All measurements were performed on the parasagittal slice of the TMJ (Figure 4).

2. The Measurements of the Joint Space

Linear measurements of optimal joint space between the condyle and fossa were made on the central sagittal slices of the TMJ in the software. The following measurements were assessed according to a study conducted by Ikeda and Kawamura (Figure 5).[30,31]

Anterior joint space (AS): Expressed by the shortest distance between the most anterior point of the condyle and the posterior wall of the articular tubercle (Figure 5).

Superior joint space (SS): Measured from the shortest distance between the most superior point of the condyle and the most superior point of the mandibular fossa (Figure 5).

Posterior joint space (PS): Represented by the shortest distance between the most posterior point of the condyle and the posterior wall of the condylar fossa. (Figure 5).

Statistical Analysis

Statistical analyses were performed using SPSS for Microsoft Windows software (version 20.0; SPSS, Chicago, IL, USA). Correlations among the variables (age, articular eminence inclination, and joint spaces) were established using Pearson's correlation coefficient with the significance set at $p<0.05$. The t-test was used to compare mean values in sex and subluxation.

To assess the intra-observer reliability, the reconstructions were assessed twice by an expert oral and maxillofacial radiologist with 8 years of experience in diagnosing TMJ on CBCT images, using a computer with a 17-inch LCD monitor in a darkened room. The minimum interval between evaluations of the same patient was 30 days. The right and left TMJs were evaluated separately, resulting in a total of 200 TMJs. Age, sex, the presence of subluxation in TMJ, joint spaces and articular angles were recorded. To avoid misinterpretation, measures had to be found in at least two consecutive slices.

Kappa statistics for agreement were used to assess the intra-observer reliability, and interpreted as follows: <0, poor agreement; 0.00-0.20, slight agreement; 0.21-0.40, fair agreement; 0.41-0.60, moderate agreement; 0.61-0.80, substantial agreement; 0.81-0.99, almost perfect agreement; and 1.00, perfect agreement. In our study, the intraobserver reliability was defined using an intraclass correlation coefficient and was found almost perfect agreement (ICC, ICC> 0.90).

RESULTS

Descriptive statistics of the joint spaces ve angles are shown in Table 1.

Relation of subluxation to joint distance and articular inclination

No statistically significant differences were found between subluxation, joint space and articular angles (*p*>0.05). So subluxation has no effect on joint space and angle.

Relation of sex to joint distance and articular inclination

No statistically significant differences were found between gender and PS, AS and articular angles (bf and tr) (*p*>0.05). However, there was a statistically significant difference only between gender and SS (*p*<0.05). The SS was higher in males (mean 3.60) than females (mean 2.89).

The relationship between articular distance, articular inclination, and age

Only SS correlated with age (r:0.254, *p*<0.001) and bf angle (r:0.215, *p*:0.002). SS was also associated only with PS from other joint distances (r:0.368, *p*< 0.001) and tr angle (r:0.335, *p*<0.001). That is, as the age progresses, the condyle is positioned further downward and forward. We found a statistically significant correlation between bf and tr angles (r:0.747, *p*<0.001 (Table 2).

Table 1. Descriptive statistics of age, the joint spaces (AS, SS, PS) and the articular eminence inclination (bf and tr angles).

	N	Minimum	Maximum	Mean	SD
Age	200	15	60	27.64	52,5
bf angle	200	22.5°	90°	58.46°	42,5
tr angle	200	19.2°	59.3°	38°	37,5
PS	200	1 mm	7.2 mm	2.3 mm	40
SS	200	0.9 mm	7.1 mm	3.3 mm	30
AS	200	1 mm	5 mm	2.4 mm	12,5

AS: Anterior joint space; **SS:** superior joint space; **PS:** posterior joint space, **bf angle:** best fit angle; **tr angle:** top roof angle; **N:** total number of TMEs examined in 100 patients, **TME:** temporomandibular joint, **°:** degrees, **SD:** standard deviation.

Table 2. The results of correlation analysis among joint space (AS, SS, PS), articular eminence inclination (bf and tr angles) and age.

	Age	tr angle	bf angle	AS	SS	PS
Age	1.000 .	0.215** *p*:0.002	0.111 *p*:0.118	-0.004 *p*:0.955	0.254** *p*<0.001	0.011 *p*:0.873
tr angle	0.111 *p*:0.118	1.000 .	0.747** *p*<0.001	0.001 *p*:0.984	0.335** *p*<0.001	0.085 *p*:0.229
bf angle	0.215** *p*:0.002	0.747** p<0.001	1.000 .	-0.009 *p*:0.284	0.381** *p*<0.001	0.100 *p*:0.159
AS	-0.004 *p*:0.955	0.001 *p*:0.984	-0.009 *p*:0.902	1.000 .	0.064 *p*:0.372	-0.034 *p*:0.632
SS	0.254** p<0.001	0.335** p<0.001	0.381** *p*<0.001	0.064 *p*:0.372	1.000 .	0.368** *p*<0.001
PS	0.011 *p*:0.873	0.085 *p*:0.229	0.100 *p*:0.159	-0.034 *p*:0.632	0.368** *p*<0.001	1.000 .

*Correlation is significant at the 0.005 level.
AS: Anterior joint space; **SS:** superior joint space; **PS:** posterior joint space.
bf angle: best fit angle; **tr angle:** top roof angle

DISCUSSION

BJHS is related to a wide range of locomotor-system complaints, such as arthralgia, myalgia, synovitis, joint dislocations, and soft tissue lesions. A reason for this correlation might be that the locomotor system has a greater propensity to mechanical overload owing to the extensive range of movements.[24] BJHS is associated with localized joint pain, sport specific injuries, and rheumatic diseases in adults.[32,33] BJHS is often asymptomatic, but BJHS is believed to be associated with inherited collagen disorders in children, such as Marfan and Ehlers-Danlos syndromes. An effective assessment tool is required for proper screening and management of BJHS.[34,35] The Scale of Beighton, is widely used as a joint mobility index.[8] There is no agreement concerning the quantitative definition of generalize joint hipermobility (GJH) The cut-off point of a score of 4 out of 9 was selected in accordance with most other studies.[8,19,20,27,28,36]

The TMJ can be affected by BJHS. The joint takes an overload, and as a result, suffers degenerative changes which can produce internal derangements and/or inflammation. An association between TMD and BJHS has been reported in several studies; however, the results have not been conclusive. Although some authors have devised that an association exists, others have not reached the same conclusions. In a systematic review of the literature dealing with the association between BJHS and TMD, it was accomplished that the association remained unclear and that further, more rigorous, studies were needed.[16]

Another research of the relation between TMD and BJHS was realized on 248 girls of between 15 and 16 years of age and found a 43% prevalence of BJHS, with signs and symptoms of TMD in 27.3% of the girls.[5] This work is not comparable with the present study, because the subjects were healthy young women, whereas our study was performed on patients who presented with TMJ complaints to our clinic. Sáez-Yuguero *et al.* noted that BJHS is more common in women with TMD complaints than healthy women.[16]

In one study, BJHS and localized condylar hypermobility were found to lend to the predisposition to TMD; however, the association between hypermobility and disk displacement was based on clinical findings alone, with no imaging findings being used.[37] We believe that the clinical examination should be confirmed by imaging techniques.

By Hansson *et al*, the joint space were classified as reduced (less than 1.5 mm), normal (between 1.5 and 4.0mm), or increased (more than 4.0 mm).[38] According to our study data, the mean values of joint spaces were in the normal range.

This study has several limitations. The study participants were not evaluated by a medical specialist for BJHS diseases. The determining of BJHS patients were made by us used to Beighton score.[8] On the other hand, a third study group was want to conduct with patients with TMJS without BJHS. However, this group was excluded because the number of enough patients could not be reached. Beighton scores of more than 4 in patients with TMJS suggest that BJHS causes hypermobility to the temporomandibular joint. However, studies with a sufficient number of patients with BJHS (-) and TMJS (+) are needed to confirm this knowledge.

BJHS treatment mainly consists of patient education, drug therapy, physical therapy modalities and exercise therapy.[39] A similar protocol is also followed for the treatment of subluxation in patients with TMJ subluxation.

In the present research, we studied a group of patients with or without subluxation according to RDC/TMD, and it was for this reason that CBCT was carried out as a complementary examination to confirm the diagnosis.

CONCLUSION

In conclusion, in this study, it was concluded that the occurrence or absence of TMJS in a patient with BJHS did not have an association with anatomic hard tissues. In other words, the joint space and the articular eminence steepness have no effect on the occurrence of subluxation in patients with BJHS.

*The authors declare that there are no conflicts of interest.

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