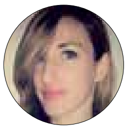
Correlation between generalised joint hypermobility and temporomandibular joint disc displacement in adolescent patients: Magnetic Resonance Imaging study



A. M. Boboc, A. De Stefano, A. Impellizzeri, E. Barbato, G. Galluccio

Sapienza University of Rome, Department of Oral and Maxillofacial Sciences, Rome, Italy

e-mail: any_bok@msn.com

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Abstract

Aim Temporomandibular disorders (TMD), in particular disc displacement, are recognised to have a multifactorial aetiology. Ligamentous laxity has been suggested as a potential risk factor for TMD. Ligamentous laxity can lead to generalised joint hypermobility (GJH) involving multiple joints, including the temporomandibular joint (TMJ). The aim of this work is to evaluate the correlation between GJH and disc displacement (DD) assessed on magnetic resonance images (MRI) of the TMJ in adolescent patients.

Materials and methods The study was included 40 adolescent patients (10–16 years), divided into two groups, a Study Group (SG), composed of 20 subjects with GJH, and a Control Group (CG), composed of 20 subjects without GJH. The GJH was assessed by the Beighton test with a threshold value of ≥ 4. The severity of the TMD was determined using the Fonseca Questionnaire and a clinical evaluation of the type of TMD. The condylar-discal relationship and the condylar mobility of the TMJ were evaluated by MRI. Pearson's χ^2 Test was performed for the analysis of the statistical correlation.

Results A statistically significant correlation emerged between GJH and DD ($p = 0.006$) and in the SG the most frequent type of disc displacement was the disc displacement without reduction. According to the Fonseca Test the most frequent TMD severity grade in patients with GJH was the mild degree ($p = 0.019$). There was also a correlation between GJH and type of TMD and the articular type was the most frequent (60%) in the SG ($p = 0.038$). No correlation was observed between GJH and the joint mobility of the TMJ.

Conclusion This study suggests that adolescents with GJH have a greater risk of developing TMJ disc displacement, especially disc displacement without reduction.

KEYWORDS Temporomandibular disorders, Temporomandibular joint disorders, Joint instability, Joint laxity, Temporomandibular joint disc, Magnetic resonance imaging.

Introduction

Temporomandibular disorders (TMD) is a collective term that includes different clinical conditions affecting the masticatory muscles, the temporomandibular joints (TMJ) and associated structures, or both [Okeson, 1996]. The origin of TMD is multifactorial and occurs frequently in the paediatric and adolescent population; many adults with TMD report that their symptoms developed mainly during adolescence [de Leeuw, 2008]. The prevalence of TMD in the paediatric and adolescent population varies widely in the literature, ranging from 16% to 68%, due to differences in the studied populations, the diagnostic criteria and the evaluation methods used [Barbosa et al., 2008; Romani et al., 2018]. The most common signs and symptoms of TMD are joint noises, joint or muscular pain, impaired or limited mandibular movement and headache. In the paediatric population signs and symptoms are usually mild or moderate, and can fluctuate, while progression to severe pain and dysfunction is rare [Barbosa et al., 2008].

Assessing the presence and severity of TMD can be done by using indices and questionnaires such as the Fonseca questionnaire, which classifies the severity of TMD as mild, moderate or severe [Bevilaqua-Grossi et al., 2006].

Disc displacement (DD) is one of the intra-articular TMD that can occur in the paediatric and adolescent population with click noise as the most frequent sign [da Silva et al., 2016].

Ligamentous laxity is a relatively frequent constitutional condition characterised by increased joint mobility beyond the normal physiological limits, and has been reported as a possible predisposing risk factor for the development of signs and symptoms of TMD [Westling and Mattiasson, 1992; Dijkstra et al., 1993; Kavuncu et al., 2006; Hirsch et al., 2008]. It is considered a non-pathological phenomenon and may affect only some or all joints and the excessive joint movement is indicated with the term joint hypermobility (JH). Generalised joint hypermobility (GJH) is present when multiple joints are involved and its prevalence varies widely in the literature, ranging from 10% to 30% in the adult population and from 2% to 65% in children and adolescents. Hypermobility decreases with age and is more frequent in females and in the Asian and African populations than in Caucasians [Remvig et al., 2007].

Different methods are used for the assessment of GJH, but the most used in clinical screening is the Beighton Scale, which evaluates nine joint sites [Beighton et al., 1973].

GJH is a frequent condition in children because the connective tissue is not completely developed with the possibility of presenting ligamentous laxity and consequently a relaxation of the tissues that hold the joint. The tension of the joint capsule and the restriction due to ligaments play a crucial role in the stability of joints, and ligamentous laxity is often the main cause of joint hypermobility. The joint laxity and the consequent JH can also affect the TMJ and cause an overload, resulting in degenerative changes such as intra-articular disorders and/or joint inflammation [Dijkstra et al., 1993].

The aim of this research was to investigate the correlation between GJH and the presence of disc displacement, evaluated on magnetic resonance imaging (MRI) of TMJs in adolescent patients.

Materials and methods

Patients on first visit at the Orthodontics Operational Unit of the Department of Odontostomatological and Maxillofacial Sciences of the University "La Sapienza" of Rome were evaluated in a period of 12 months (from december 2018 to december 2019). The final sample was composed of 40 adolescent patients divided into two groups: Control Group (CG) of 20 subjects without GJH and Study Group (SG) of 20 subjects with GJH, identified with Beighton Test with a threshold value of ≥ 4 [Beighton et al., 1973].

The inclusion criteria were: patients in good general health, males and females, aged between 10 and 16 years, informed consent to perform orthodontic diagnostic evaluation, GJH assessment and MRI of TMJs. The exclusion criteria were: concomitant systemic pathologies, clinical history of craniofacial traumas, syndromic patients, previous orthodontic or gnathological treatment, previous TMJ or maxillofacial surgeries and inflammatory TMJ conditions.

After patient's data collection and assessment of the presence of GJH by the Beighton test, the Fonseca questionnaire for TMD was filled by the patients and the evaluation of signs and symptoms was carried out to determine the type of TMD [Bevilaqua-Grossi et al., 2006].

The intra-articular relationships of the condylar-discal complex and the condylar mobility were evaluated on MRI images (Table 1). The GJH was assessed using the Beighton test and the procedures used were as follows.

- Passive abduction of the thumb towards the front of the forearm.
- Passive dorsiflexion of the metacarpophalangeal joint of the 5th finger >90 °.
- Hyperextension of the elbow joint >10°.
- Hyperextension of the knee joint >10°.
- Contact of the palms of the hands on the floor with the extended lower limbs.

One point was assigned for each positive test (1 for the right side and 1 for the left side) considering as threshold value for the diagnosis of GJH a score of ≥4 positive tests.

The Fonseca questionnaire, which classifies patients into categories with different degrees of severity of TMD, is composed of 10 questions and, based on the sum of the points, the patients are classified into 4 categories: without TMD (score 0–15 points), mild TMD (score 20–40), moderate TMD (score 45–65) and severe TMD (score 70–100) [Bevilaqua-Grossi et al., 2006].

The type of TMD was assessed by clinical examination and, based on the presence of signs and symptoms, was classified into: articular type if the patient presented pain only upon palpation of the TMJ and/or joint click; muscular type if the patient had pain only upon palpation of the masticatory muscles; and articular and muscular type if both conditions were present.

Beighton test	Presence of GJH (cutoff ≥ 4)
Fonseca Questionnaire (for TMD severity)	Mild Moderate Severe
Clinical evaluation of the type of TMD based on • Palpation pain in the masticatory muscles • Palpation pain of the TMJ • Presence of clicks during mandibular movements	Articular Muscular Articular and muscular
Disc displacement evaluated on MRI	Normal DDwR DDwoR
Condilar mobility evaluated on MRI	Hypomobility Normal mobility Hypermobility

TABLE 1 Evaluations for each patient.

The joint click was marked positive when it occurred at least once during three consecutive opening and closing movements and muscle palpation pain was marked positive if the patient referred pain upon palpation of the masticatory muscles under a pressure of 1 gram for 3 seconds. The joint pain upon palpation was marked as positive if it was present under a pressure of 0.5 grams in the TMJ area.

Intra-articular relationships of the condyle-disc complex of both TMJs, with open and closed mouth, both in the sagittal and coronal projection, were assessed by MRI. Condylar mobility with open mouth also was assessed in the sagittal projection.

MRI images of the TMJ were obtained with a dedicated bilateral antenna. On the basis of an axial image, sections 3 mm thick were obtained on the oblique sagittal plane (perpendicular to the longitudinal axis of the anteroposterior condyle) and on the oblique coronal plane (parallel to the lateromedial condyle axis). For each TMJ (right and left) the following sequences were obtained: T1 sagittal oblique -w, T2 sagittal oblique -w, oblique sagittal proton density (PD) and oblique T1-w coronal in closed mouth and oblique sagittal PD with the mouth open.

Disc position in each TMJ was identified as follows [Bertram et al., 2001].

- Normal position (N) if, with a closed mouth, in the sagittal projection the posterior band of the articular disc is located above the apex of the condylar head (at 12 o'clock position), while with the open mouth the thinnest intermediate area is interposed between the condyle and the articular eminence; in coronal projection the disc is positioned over the head of the mandibular condyle. With the open mouth the disc remains interposed between the articular surface of the condyle and the articular surface of the articular eminence.
- Disc displacement with reduction (DDwR) if, with a closed mouth, the posterior band is located anteriorly to the condylar head in sagittal projection and coronally can appear normal and/or displaced medially or laterally. When the mouth is open, the correct relationship of the condyle-disc complex is restored.
- Disc displacement without reduction (DDwoR) the disc is displaced from the normal position in sagittal and/or coronal

projection both with closed and open mouth.

In the present investigation, the discal position of each patient was classified into: normal (N) if the position was normal in both TMJs; DDwR if at least one of the TMJs had a DDwR and the contralateral was similar or normal; DDwoR if at least one of the TMJs had a DDwoR (Fig. 1).

Condylar mobility of both TMJs was also analysed by MRI based on the translation of the condylar head compared to the articular eminence of the temporal bone in sagittal projection. In maximum opening, normal mobility corresponds to a condyle located in the vertex of the articular eminence. Movement beyond the articular eminence by the condyle was considered condylar hypermobility, while the failure to reach the articular eminence was marked as hypomobility [Kalaykova et al., 2006].

Condylar mobility was classified as normal if both TMJs were normal, hypermobility if at least one TMJ presented hypermobility and hypomobility if one of the TMJs was hypomobile and the other was similar or normal (Fig. 2).

Statistical analysis

The statistical analysis was carried out using the IBM SPSS software version 20.0 and application of Pearson's χ^2 test to compare the frequency of events of the two groups. The Chi-Square analysis conducted in SPSS indicates the level of significance for the difference between the samples. The statistical significance level of the correlations was set at 5%, i.e. with $p \leq 0.05$.

Results

Of a total of 560 patients, 61 patients who met the inclusion criteria were initially selected, of whom 21 were excluded for different reasons (9 did not complete the evaluation, 4 showed inflammatory diseases during the evaluation of the TMJ, 2 did not give consent, 6 refused treatment). The remaining 40 patients completed all the required evaluations and performed the diagnostic investigations necessary to conduct the present research and formed the final sample, which was composed of 27 females (67.5%) and 13 males (32.5%) aged between 10 and 16 years with an average age of 12.95 (SD 1.99 years).

Patients were divided into a study and control group, SG and CG respectively, according to the presence or absence of GJH, evaluated with the Beighton Test (≥4 / 9). The SG was composed of 20 subjects with GJH, including 14 females (70%) and 6 males (30%), and the CG was composed of 20 subjects without GJH, including 13 females (65%) and 7 males (35%). There was no statistically significant correlation between GJH and gender, but in both groups there was a greater percentage of females than males.

From the analysis of the results of the Fonseca questionnaire it emerged that mild TMD was the most frequent form in the total sample (35%). In the comparison between groups, a statistical correlation emerged between GJH and the severity of the TMD ($p = 0.019$). The SG patients presented different degrees of TMD with the mild grade being the most frequent (50%), while the CG patients presented a more even distribution and did not present severe TMD.

A statistical relationship was found between the GJH and the TMD type ($p = 0.038$). The SG presented a higher frequency of articular TMD (60%), while the CG presented a more even distribution of TMD types.

Articular clicks were recorded during the clinical evaluation and the statistical analysis showed that 85% of the SG presented articular clicks with a statistically significant correlation (p =

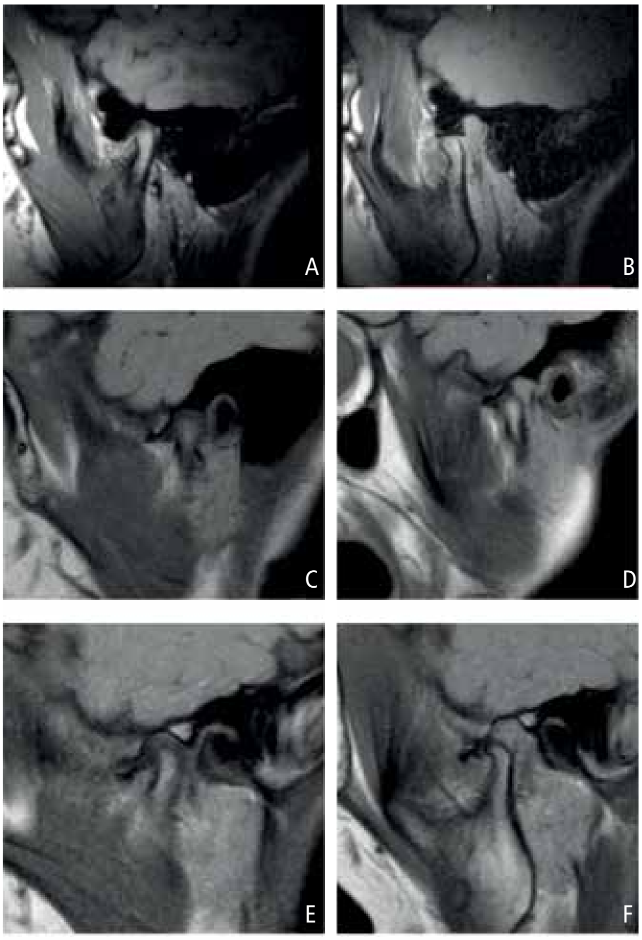

FIG. 1 Disc position. Normal disc position with closed (a) and open (b) mouth. Disc displacement with reduction with closed (c) and open (d) mouth. Disc displacement without reduction with closed (e) and open (f) mouth

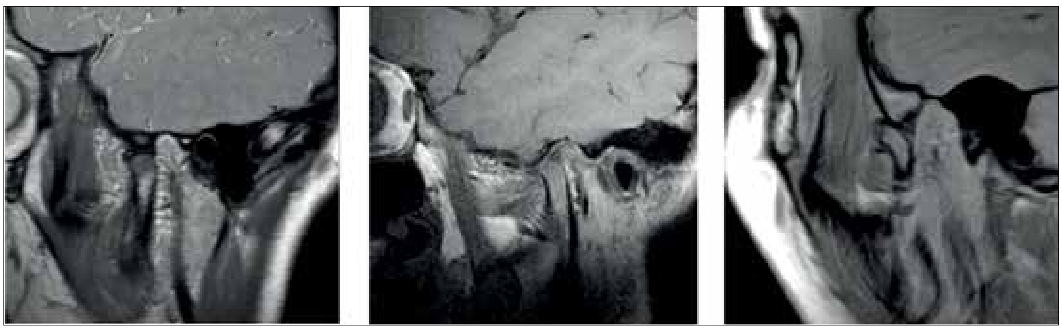

FIG. 2 Condylar mobility with open mouth.

	Disc position		
	Normal	DD	
Control Group	Count	10	10
	% in GJH	50.0%	50.0%
Study Group	Count	3	17
	% in GJH	15.0%	85.0%
Total	Count	13	27
	% in GJH	32.5%	67.5%

GJH: Generalised joint hypermobility DD: Disc displacement in at least one of the two joints

TABLE 2 Gjh and disc displacement.

	Disc Displacement type			
		Normal	DDwR	DDwoR
Control Group	Count	10	7	3
	% GJH	50.0%	35.0%	15.0%
Study Group	Count	3	7	10
	% in GJH	15.0%	35.0%	50.0%
Total	Count	13	14	13
	% in GJH	32.5%	35.0%	32.5%

GJH: Generalised joint hypermobility DDwR; Disc displacement with reduction. DDwoR: Disc displacement with reduction

TABLE 3 Gjh and disc displacement.

0.003). In the CG the joint click was absent in 60% of cases.

In the comparison between GJH and DD in at least one of the two TMJs emerged a statistical correlation (p = 0.018), and 85% of the SG presented disc displacement (Table 2).

In the SG the most common type of DD was DDwoR (50%), while in the CG 50% presented a normal intra-articular relationship and 35% presented a DDwR (p = 0.02) (Table 3).

The relationship between GJH and condylar mobility, evaluated by MRI, was also analysed, but there was no statistically significant relationship (p> 0.05). 40% of SG presented hypomobility, 30% hypermobility and 30% normal mobility, while the most frequent condylar mobility in the CG was normal mobility (50% of CG).

Discussion

In the present study a statistically significant association was observed between GJH and disc displacement and the most frequent type in the SG was disc displacement without reduction.

TMD are widespread, with about 10% of the world's population experiencing having pain and other dysfunctional symptoms that require treatment, and many of these subjects develop predisposing factors during childhood or adolescence [de Leeuw, 2008].

From the analysis of the data collected by the Fonseca questionnaire, the most frequent TMD were those of a mild grade, representing 35% of the total sample. This result is in agreement with other authors who report that TMD in growing patients occur mainly mild or moderate, with fluctuations and rare progressions to severe pain and dysfunction [Barbosa et al., 2008].

With regard to GJH and gender, there was a high percentage of females in both groups, but without a statistical correlation with GJH, unlike previous studies where there was a predominance of the female gender in the patients with GJH [Remvig et al., 2007].

GJH has been reported as a possible predisposing risk factor for the development of signs and symptoms of TMD [Westling and Mattiasson, 1992; Kavuncu et al., 2006]. In the literature there are numerous studies concerning the correlation between GJH and TMD with conflicting results and subsequent controversy on the subject over the years. The hypothesis concerning the influence of GJH in the development of TMD has not yet been completely confirmed.

From some studies it emerged that the TMJs of subjects with GJH are at increased risk of articular clicks as a manifestation of articular disc dislocation [Westling and Mattiasson, 1992; Kavuncu et al., 2006; Hirsch et al., 2008], and at lower risk of reduction in the ability to open the mouth but without an increased risk of TMD pain (myalgia/arthralgia).

During the evaluation of the TMJ both groups presented articular noises (during joint movements and lateral-deflection during the opening), but these were more frequent in subjects with JH. A statistically significant correlation between GJH and articular click emerged (p = 0.003), in agreement with previous studies [Chiodelli et al., 2016].

The presence of articular noise could suggest a possible reduction of proprioception that in patients with GJH can cause condylar hypertranslation, articular noises and possibly intra-articular disorders and joint inflammation [Sáez-Yuguero et al., 2009].

These results are in agreement with aepidemiological studies that report joint noise as the most common sign and symptom of TMD in the 10–15 age group, in addition to pain during chewing and muscle pain upon palpation [Köhler et al., 2009].

There was also a statistical relationship between GJH and the clinically evaluated type of TMD (p = 0.038). Subjects with GJH had a higher frequency of articular TMD (60% of the GS), while the remaining 40% presented articular and muscular TMD, either isolated or coexisting.

The greater frequency of articular TMD in the SG, which also coincides with a higher frequency of DD in the same group, determined a statistically significant correlation (p = 0.018) between GJH and disc dislocation. In fact 85% of the SG presented at least one TMJ with disc dislocation, while only 15% of this group presented a normal position of the articular disc, in agreement with studies by other authors [Bates et al., 1984; Westling and Mattiasson, 1992; Adair and Hecht, 1993; Perrini et al., 1997; De Coster et al., 2005; Kavuncu et al., 2006; Huddlestone Slater et al., 2007; Hirsch et al., 2008; Ögren et al., 2012]. However, unlike this study, they identified disc displacement clinically. Westling and Mattinson [1992] studied a group of adolescents and found a statistical correlation between DD and GJH. Other studies that have investigated the relationship between GJH and DD, clinically evaluated [Conti et al., 2000; Winocur et al., 2000; Dworkin and Huggins, 2010] or in MRI [Sáez-Yuguero et al., 2009], have not found a correlation. With this respect, the different age of the sample should be taken into account.

The most frequent type of DD in subjects with GJH was

DDwoR, while in the control group it was DDwR. DDwoR can induce greater mechanical stress than DDwR, causing greater joint pain [Emshoff, 2001; Sáez-Yuguero et al., 2009]. Some studies suggest a clinical evaluation of the articular disc before 10 years of age, and if necessary preventive measures to stop progression of disc dislocation, since they have observed progression of disc dislocation with age [Ikeda et al., 2014].

The type of condylar mobility most frequently encountered during MRI analysis was normal mobility in the CG and hypomobility in the SG, although without statistical significance ($p > 0.05$). This result is in line with previous studies that did not find a correlation between the GJH and an increased condylar translation [Conti et al., 2000].

GJH by definition is characterised by a greater excursion of normal joint movements. Hypomobility was found to be the most frequent type of condylar mobility in the SG (40%). This result could be explained by the high frequency of DD in these subjects (85% of the SG).

A possible limitation of this investigation was the use of a ≥4 cutoff for the Beighton test to evaluate and determine the presence of GJH. This cutoff was chosen in accordance with most studies. The laxity of ligaments decreases with age and is also influenced by gender and ethnicity. Considering that joint mobility decreases with age, a greater cutoff of the Beighton Sscore as a diagnostic criterion (≥6 positive joints) has been suggested in growing patients and the prevalence of ligamentous laxity has also decreased when applied in the studies [Remvig et al., 2011].

Conclusions

This study suggests that adolescent subjects with GJH are at greater risk of TMJ's disc displacement, especially discal displacement without reduction.

Early diagnosis of TMD signs and symptoms in growing patients is important to prevent or minimise worsening pain and dysfunction and to reduce the TMD impact on the quality of life.

Generalised hypermobility, being a congenital condition, is not a modifiable risk factor. However, growing subjects with TMD associated with GJH should be carefully evaluated as this can influence the therapeutic approach and could affect the prognosis of the TMD and consequently the normal craniofacial development.

Further studies with larger sample sizes are needed to confirm the results.

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