

# Association between generalized joint hypermobility and temporomandibular joint clicking

*Asociación entre Hiperlaxitud Articular Generalizada y Chasquidos de la Articulación Témporo Mandibular*

*Associação entre Hiperlaxidade Articular Generalizada e Cliques de Articulação Temporomandibular*

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## Abstract

This study analyzes whether generalized joint hypermobility (GJH) is a risk factor for temporomandibular joint disorders (TMD). Therefore, we evaluated the potential association between TMD clicking and GJH diagnosis. We worked with the following hypothesis: patients with GJH would have a higher prevalence of TMJ clicking than those without it, making GJH a risk factor for joint disorders.

Two hundred and fourteen students from the School of Dentistry of Universidad de la República del Uruguay (UdelaR) were examined: 161 female and 53 male, aged 18 to 30 (average age: 23.8 years, SD=2.7). Each participant was given a questionnaire, and a clinical examination was performed to diagnose GJH using the Beighton score (BS), clicking, history of maxillofacial trauma, orthodontics, full dentition, open lock, and shift. A calibrated blind researcher (kappa inter-rater click calibration = 0.68; intra-rater BS score=0.82, click=1) performed all the examinations. The Ethics Committee approved the study, and all the participants signed an informed consent. A multiple logistic regression model was used to analyze the data statistically.

GJH prevalence was 34.16% in women and 7.55% in men; clicking prevalence was 24.22% in women and 11.32% in men. There was a significant association between sex (OR=3.244, p-value 0.018) and history of trauma (OR=2.478, p-value 0.041) and the presence of clicking. No association was found between clicking and GJH.

Female sex and history of trauma could be risk factors for TMJ disorders. The lack of association between GJH and clicking in this age group (18-30) suggests that GJH may not be a risk factor for developing these pathologies.

**Keywords:** Temporomandibular Articulation, Joint Instability.

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## Resumen

El objetivo del estudio fue analizar si la hiperlaxitud articular generalizada (HAG) es un factor de riesgo para los trastornos de la articulación temporomandibular (ATM). Para ello se evaluó la posible asociación entre chasquido de la ATM y el diagnóstico de HAG. Se trabajó con la siguiente hipótesis: el paciente con HAG, tendría mayor prevalencia de chasquido a nivel de la ATM que los que no la presentan, constituyendo la HAG un factor de riesgo para padecer un desorden articular (DA).

Se examinaron 214 estudiantes de facultad de odontología (FO) de la Universidad de la República (Udelar) de Uruguay, 161 participantes del sexo femenino y 53 masculino, de entre 18 y 30 años (edad media 23.8 años, DE=2.7). A cada participante se le realizó un cuestionario y un examen clínico para diagnóstico de HAG utilizando el índice de Beighton (IB), chasquido, antecedente de trauma maxilofacial, ortodondia, dentición completa, bloqueo abierto y turno. Todos los exámenes fueron realizados por un investigador ciego calibrado (calibración interoperador chasquido kappa= 0.68; intraoperador Beighton=0.82, chasquido=1). El estudio fue aprobado por el Comité de Ética y todos los participantes firmaron un consentimiento informado. El análisis estadístico de los datos fue realizado en base a un modelo de regresión logística múltiple.

La prevalencia de HAG fue 34.16% en el género femenino y 7.55% en el masculino, de chasquido 24.22% para el femenino y 11.32% en el masculino. Las variables género (OR=3,244, p-valor 0,018) y antecedente de traumatismo (OR=2,478, p-valor 0,041) se asociaron significativamente a la presencia de chasquido. No se encontró asociación entre chasquido e HAG. El género femenino y los antecedentes de traumatismo podrían ser factores de riesgo para desórdenes a nivel de la ATM. La ausencia de asociación entre HAG y chasquido en dicho grupo etario (18-30 años), sugiere que dicho factor podría no ser de riesgo para el desarrollo de dichas patologías.

**Palabras clave:** Articulación Temporomandibular, Inestabilidad de la Articulación.

## Resumo

O objetivo do estudo foi examinar se a hiperlaxidade articular generalizada (HAG) é um fator de risco para disfunção articulação temporomandibular (DAT). Isso foi feito avaliando a possível associação entre cliques atm e diagnóstico hag. O trabalho foi feito com a seguinte hipótese: o paciente com HAG, teria maior prevalência de clique no nível atm do que aqueles que não o fazem, tornando a HAG um fator de risco para DAT.

Foram examinados 214 alunos de Odontologia da Universidad de la República (Udelar) do Uruguai, 161 do sexo feminino e 53 do sexo masculino, com idade entre 18 e 30 anos (idade média de 23, 8 anos, DE: 2, 7). Cada participante recebeu questionário e exame clínico para diagnóstico de HAG utilizando o índice Beighton (IB), clique, antecedente de trauma maxilofacial, ortodontia, dentição completa, bloqueio aberto e turno. Todos os testes foram realizados por um pesquisador cego calibrado (calibração interoperadora kappa-click-0, 68; intraoperador Beighton-0.82, clique-1). O estudo foi aprovado pelo Comitê de Ética e todos os participantes assinaram consentimento informado. A análise estatística dos dados foi realizada com base em um modelo de regressão logística múltipla.

A prevalência de HAG foi de 34, 16% no sexo feminino e 7, 55% no masculino, de clique 24, 22% para o feminino e 11, 32% para o masculino. As variáveis de gênero (OR-3.244, valor p 0, 018) e antecedente do trauma (OR-2.478, valor p 0, 041) estiveram significativamente associadas à presença de clique. Não foi encontrada associação entre clique e HAG.

O sexo feminino e o histórico de trauma podem ser fatores de risco para distúrbios no nível do articulação temporomandibular. A ausência de associação entre HAG e click sugere que esse fator pode não estar em risco para o desenvolvimento de tais patologias.

**Palavras-chave:** Articulção Temporomandibular, Inestabilidade Articular.

## Introduction

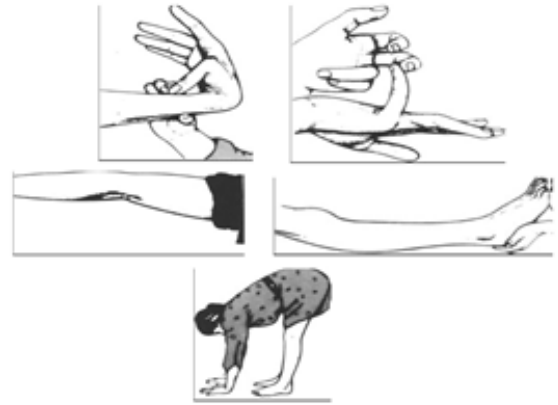
Temporomandibular disorders (TMD) have been defined as a heterogeneous group of conditions affecting the temporomandibular joint (TMJ), masticatory muscles, and related structures. It has recently been considered of multifactorial etiology, where several systems (psychological, genetic, hormonal, neurological) interact with anatomical factors (2). Within TMDs, we find TMJ joint disorders. Joint disorders often arise as mild pathological conditions and can evolve into more severe conditions (4). Clicking noises and pain are clinical manifestations associated with early TMJ disorders (5). A nationwide survey conducted in Uruguay reported a 29.77% clicking prevalence on the clinical examination in Montevideo and a 23.01% prevalence in the rest of the country. Approximately 19% of the Swedish population show joint disorder symptoms, 7%, reciprocal clicking, and chronic 12%, closed locking (7). Joint clicking is considered a predictive variable for joint disorder and has been considered its main clinical manifestation (8). Disc displacement with reduction (DDwR) has been associated with condylar flattening, and disc displacement without reduction (DDwoR) has been associated with degenerative bone changes (9). The prevalence of TMDs and joint disorders is higher in women than in men (6,10). General and local risk factors could explain this difference (8). Knowing these predisposing and triggering factors could help prevent TMDs while understanding perpetuating factors increases the chances of treatment success (11).

Joint hypermobility (JH) has been studied as a risk factor for joint disorders and is defined as an increased joint mobility range compared to the general population. It may be observed as local joint hypermobility (LJH) or generalized joint hypermobility (GJH) (12). GJH may result from a collagen defect, such as in Ehlers-Danlos and Marfan syndromes, although it may also occur without an underlying collagen defect. These could be cases of benign GJH (13).

The Beighton score (Fig. 1) has gained international acceptance and appears to be the most widely used index in the scientific literature to diagnose GJH (14).

The Beighton score has been validated for use in dentistry (15). Several studies have been conducted to analyze the possible association between joint disorders and GJH.

**Fig.1**



Extracted from Carter & Wilkinson (20).

A single systematic review helped us establish the association, although GJH could not be shown as a risk factor for joint disorders (12). A higher prevalence of GJH and TMD was found in a case-control study in the 15-24 age group. Additionally, there was an association between GJH and clicking. The authors concluded that joint disorder risk is higher in individuals with GJH (16). A study of 701 young patients (77% female) found that 63% of joint disorder patients were diagnosed with GJH, which shows a strong connection (17). A study of 1,600 individuals reported a GJH prevalence of 6.9% and a joint disorder prevalence of 14.8%. The authors concluded that GJH was associated with DDwR TMD (18). Ting-Han et al. conducted a retrospective study (19) on 975,788 randomly selected individuals. They concluded that confirming the association between joint disorder and GJH suggests that a multidisciplinary team should treat individuals with TMD and GJH.

In a recent case-control study<sup>(20)</sup> applying the BS and RDC/TMD criteria, the authors concluded that the risk of developing a joint disorder is higher in patients with specific systemic pathologies such as GJH.

Barrerara Mora et al.<sup>(21)</sup> had opposite results in a study of 140 patients (male and female) needing orthodontic treatment. They reported a GJH prevalence of 28.%, finding no association between different types of TMD and GJH. The authors of a case-control design study including 60 cases and 60 control cases<sup>(22)</sup> (mean age=25) concluded that GJH does not contribute to TMD onset, as it was a common finding in asymptomatic patients. Sáez-Yuguero et al. analyzed 66 young women being treated for TMD. They reported a BS of 51% and a DDwR of 40%, concluding that the BS and an MRI could not establish that GJH is a risk factor for TMJ<sup>(23)</sup>.

A cross-sectional and prospective study<sup>(24)</sup> evaluated 34 young women diagnosed with joint disorder. GJH prevalence was 65%, but it was impossible to establish GJH as a risk factor for TMD.

A case-control study that included 42 surgical patients and 20 control cases found they were 9.6 times more likely to develop reciprocal clicking with GJH. They concluded that GJH is a significant etiological factor for developing reciprocal clicking and chronic closed lock<sup>(25)</sup>. Another control case design<sup>(26)</sup> used MRI technology for joint diagnosis. The authors concluded that GJH is not a predictor of joint disorder based on the Bighton score.

The results of these studies are controversial. Some studies have found an association between benign GJH and joint disorders, while others have been unable to prove such an association.

This study aimed to:

- assess the prevalence difference between GJH sex and clicking;
- study the potential association between joint clicks and GJH.

We worked with the following hypothesis: GJH patients would have a higher prevalence of TMJ clicking than those without it, making GJH a risk factor for joint disorders.

## Materials and methods

### Participants

A cross-sectional study was designed. Students at the School of Dentistry, UdelaR, aged between 18 and 30 were surveyed. They were selected consecutively, Monday through Friday, in the two shifts—from 8:00 am to 12:00 pm and from 7:30 pm to 11:30—pm during October, November, and December 2017, and March 2018. The study protocol was approved by the Ethics Committee of the School of Dentistry, UdelaR (Uruguay). Each participant was interviewed and clinically examined in a single session, including BS application.

### Sample size

The prevalence rates previously reported were considered to calculate the sample size<sup>(21)</sup>. Considering a level of 5% and a power of 80%, the resulting sample size was 200 (+25 to cover non-responses): 150 women and 50 men to find differences in GJH prevalence between sexes. Everyone agreed to participate.

The following selection criteria were considered:

Inclusion criteria:

- School of dentistry (UdelaR) students
- Men and women aged between 18 and 30
- Participants agreed to take part in the clinical study and expressed their consent in writing.

Exclusion criteria:

- History of TMJ surgery
- Any general disease affecting connective tissue or joints
- Injury with permanent joint mobility sequelae
- Joint prosthesis
- Extreme hypermobility (e.g., dancers, professional athletes)

## Variables

Each participant was given a questionnaire, and a clinical examination was conducted in a single interview, which included the BS. The rater was blind to the study objectives. The rater is a member of the Department of Rehabilitation, Fixed Prosthodontics, and TMD, with field experience. The rater was calibrated and measured the variables studied (kappa inter-rater click calibration = 0.68; intra-rater BS =0.82, click=1). Table 1 shows the results of each calibration.

Table 1

variable	kappa
<b>inter</b>	
clicking	0.679
beighton 1	0.800
beighton 2	0.824
beighton 3	0.800
beighton 4	0.800
beighton 5	0.750
beighton 6	1.000
beighton 7	-
beighton 8	-
beighton 9	0.800
<b>intra</b>	
clicking	1.000

**Clicking:** We followed the DC/RDC concepts<sup>(3)</sup> and calibration protocol used by Riva et al. in 2011<sup>(6)</sup>. The rater explained that TMJs would be examined for joint noise during opening and closing. The rater placed their index and middle fingers on the skin covering the sides of both TMJs simultaneously while the participant made three maximal opening and closing movements, starting and ending these movements in the maximum intercuspatation position. When the noise occurred in one of three cycles, in one or both TMJs, both when opening and when opening and closing, the participant was diagnosed with click-type joint noise.

A differential diagnosis was made concerning shape alterations, discarding reciprocal joint noises in the same range in opening and closing; therefore, the result was negative for clicking.

The presence of full dentition (**FDent**) and the absence of molars 6, 7, and 8 (**Molars**) in the same quadrant were evaluated in an intra-oral clinical examination with examination instruments, mirror, clamp, probe, periodontal probe, and dental light.

**FDent** was positive when all incisors, premolars, and molars were present, considering the absence or presence of some or all of the third molars.

**Molars** was positive when the rater detected the clinical absence of first, second, and third molars in the same quadrant.

**Open lock and history of trauma** were positive when the participant reported them in their medical history.

The **Beighton score** was applied as proposed by the author and expanded using a goniometer as per Juul-Kristensen et al.'s 2007 description<sup>(14)</sup>.

### Beighton score

- 1.Passive dorsiflexion of the thumb with flexed wrist
- 2.Passive dorsiflexion of little finger beyond 90° with extended wrist
- 3.Active hyperextension of elbows beyond 10°
- 4.Active hyperextension of knees beyond 10°
- 5.Forward flexion of the trunk with the knees fully extended so that the palms of the hands rest flat on the floor

The score range was 0-9. The higher scores indicated higher hypermobility. A score of 4 or more points was considered Beighton positive. Patients were not classified based on hypermobile joints.

## Statistical analysis

First, a descriptive analysis of the variables was conducted with frequency distribution for the qualitative variables. For quantitative data, however, the mean and standard deviations were calculated. Second, bivariate analysis of clicking and potential risk factors was conducted. Finally, these factors were assessed with multiple logistic regression (MLR) analysis, and the relevant odd ratios (OR) and intervals were calculated with a 95% confidence level.

## Results

A total of 214 participants with an average age of 23.8 (SD=2.7) were included, of whom 161 were women and 53 men. Total GJH prevalence was 27.6%, clicking, 21.0%, and history of trauma, 15.0%. The frequency distribution of the other variables is presented in Table 2.

The prevalence of GJH by sex was higher in women (34.16%; SD=3.74%) than in men (7.55%; SD=3.66%) (Graph. 1). Clicking prevalence was also higher in women (24.22%; SD=3.39%) than in men (11.32%; SD=4.39%) (Table 3).

Table 2

	Frequency	%
<b>Sex</b>		
men	53	24,8%
women	161	75,2%
<b>History of trauma</b>		
yes	32	15,0%
no	182	85,0%
<b>Open lock</b>		
yes	4	1,9%
no	210	98,1%
<b>Orthodontics</b>		
yes	144	67,3%
no	70	32,7%
<b>Clicking</b>		
yes	45	21,0%
no	169	79,0%
<b>Full dentition</b>		
yes	7	3,3%
no	207	96,7%
<b>Hypermobility</b>		
yes	59	27,6%
no	155	72,4%
<b>Shift</b>		
morning	140	65,4%
evening	74	34,6%



**Graph 1: Prevalence of hypermobility by sex**

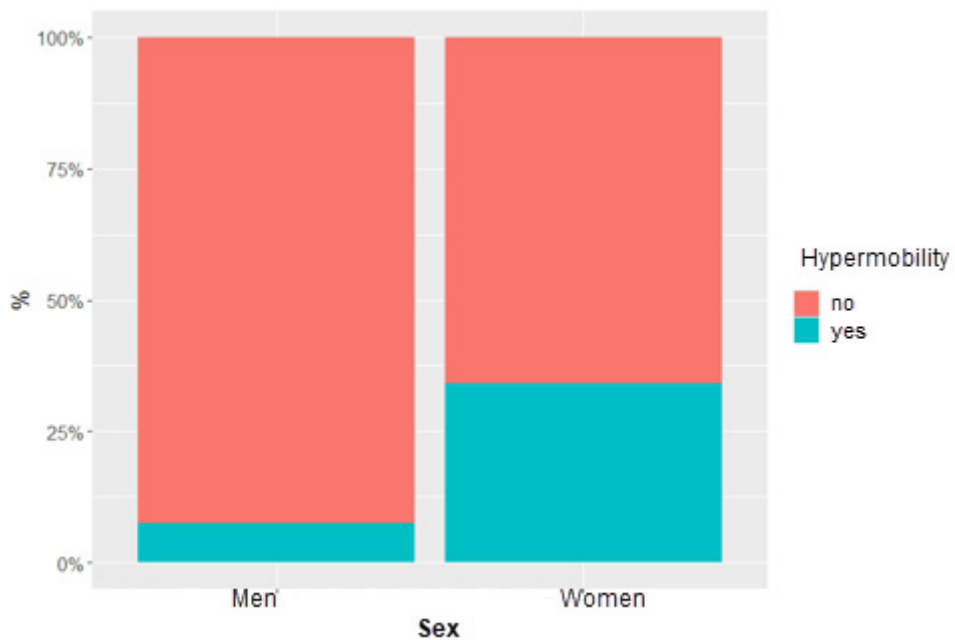


Table 3 shows the results of the first bivariate analysis.

**Table 3**

	No clicking	Clicking	p-value
<b>Sex</b>			
men	88,7	11,3	0,071
women	75,8	24,2	
<b>History of trauma</b>			
yes	81,3	18,7	0,076
no	65,5	34,5	
<b>Orthodontics</b>			
yes	77,1	22,9	0,780
no	77,9	22,1	
<b>Hypermobility</b>			
yes	78,7	21,3	0,999
no	79,7	20,3	
<b>Shift</b>			
morning	82,1	17,9	0,165
evening	73,0	27,0	

Molars, FDent, and lock were not included in the bivariate analysis or logistic regression model because they lacked sufficient variability to evaluate the association with the response variable. The independent variables used for clicking were age, sex, trauma history, orthodontics, shift, and BS.

The prevalence of clicking in participants with and without GJH was similar: 21.3% and 20.3% (Graph. 2).

**Graph 2: Prevalence of clicking according to hypermobility**

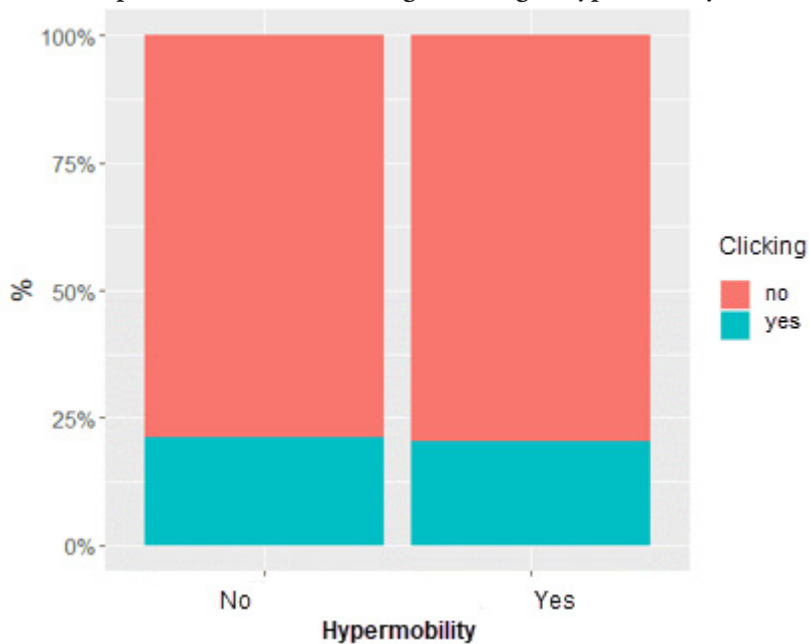


Table 4 describes the results obtained for each BS component.

**Table 4: Prevalence of each Beighton score component according to sex**

	n	Right elbow	Left elbow	Right thumb	Left thumb	Right little finger	Left little finger	Right knee	Left knee	Flexion
Men	53	5,7%	5,7%	17,0%	26,4%	17,0%	22,6%	0,0%	1,9%	7,6%
Women	161	20,5%	26,1%	45,3%	47,2%	34,8%	39,8%	10,6%	11,2%	27,3%
Total	214	16,8%	21,0%	38,3%	42,1%	30,4%	35,5%	7,9%	8,9%	22,4%

**Table 5: Logistic regression for risk factors vs. clicking**

	Estimation	OR	p-value	95% CI
Sex	1,177	3,244	0,018	(1,298 -9,422)
Age	-0,004	0,996	0,949	(0,872 -1,134)
Trauma	0,907	2,478	0,041	(1,016 -5,871)
Orthodontics	-0,378	0,685	0,308	(0,333 -1,433)
Hypermobility	-0,331	0,718	0,408	(0,318 -1,542)
Shift	0,606	1,833	0,093	(0,899 -3,721)

The proposed model attempted to measure the potential association between possible risk factors and joint clicking as a sign of joint disorder.

Table 5 shows the MLR results. Female sex was associated with a potential risk factor for joint disorder with an OR=3.2 (p-value 0.018).



A history of maxillofacial trauma had a clear association with clicking with an OR=2.5 (p-value 0.041).

Age, undergoing or having undergone orthodontic treatment, and shift were not associated with clicking prevalence. GJH was not significantly associated with clicking as a clinical sign (p-value: 0.408 95% CI (0.318 -1.542))

## Discussion

Previously published studies generally applied various methodological designs, thus making it difficult to conduct a comparative analysis. The association between TMD and GJH has been reported in several studies, although results are not decisive. Some authors have proved the association, while others have not been able to prove it.

The studies that do not show a connection, as did our results, are bivariate statistical analysis, whose methodology poses statistical inference limitations<sup>(22-26)</sup>. Creating an MLR model allowed us to control the potential risk factors for clicking most commonly reported in the literature. In contrast, two prospective design studies in general population samples concluded that GJH is a risk factor for TMDs<sup>(15,20)</sup>.

Although the BS is the most accepted criteria in the literature<sup>(27)</sup>, other tests have been used: Carter and Wilkinson's criteria, Rocabado's Temporomandibular Pain Analysis for TMD, Brighton criteria. This could make it difficult to compare our results and theirs. Additionally, the studies surveyed that applied the BS showed that GJH ranged from 3 to 6 positive points. Deodato et al.<sup>(17)</sup> had a cut-off point of 3 and reached a GJH prevalence of 66%, while Pasinato et al.<sup>(24)</sup> and Sáez Yuguero et al.<sup>(23)</sup> used a cut-off point of 4 and reached 51% and 65% prevalences. This might question the validity of the BS as a diagnostic element. GJH prevalences in patient samples range from 4%<sup>(26)</sup> (Wang et al., 2012) to 65%<sup>(23,17)</sup>.

Our sample had a reduced age range (18-30), so age variability could not be studied compared to other studies with broader age ranges in non-patient samples<sup>(18)</sup>. A GJH prevalence of 43% has been reported in adolescents without TMD<sup>(28)</sup>. Our clicking prevalence results are similar to those reported in a national survey<sup>(6)</sup> and studies with general population samples<sup>(18)</sup>. Our study distinguished GJH prevalence by sex. However, most studies have been conducted exclusively in women<sup>(24,26,28)</sup>, reporting other ratios that reach up to 5:1 in their favor<sup>(17)</sup>. Primarily, there are no prevalence reports by sex. Hirsch et al.<sup>(18)</sup> reported a GJH prevalence of 6.9% in the general population, without age discrimination. This is lower than our results but with differences by sex.

Only one study<sup>(25)</sup> included a history of trauma as a variable in bivariate analysis and with a negative association result. Our results showed that this control variable impacted joint clicking prevalence (p-value: 0.041 - 95% CI (1.016 -5.871)).

None of the studies surveyed included racial diversity in their samples. Some focus on populations of European descent<sup>(12,17-18,21-25,28)</sup> and others on Asian people<sup>(16,19,26)</sup>, which makes it impossible to generalize results.

Only three studies<sup>(18,20,26)</sup> described intra- and inter-rater calibration for both diagnostic methods. Calibration is necessary to ensure the methodological rigor that guarantees result reliability.

## Conclusions

Generalized joint hypermobility diagnosed with the BS was equally prevalent in participants with and without signs of joint disorders. The study results suggest that GJH should not be considered a risk factor for TMJ disorders among young people (18-30 years). Female sex and history of maxillofacial trauma could be regarded as risk factors for developing TMJ pathologies.

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### **Authorship contribution:**

1. Conception and design of the study
2. Acquisition of data
3. Data analysis
4. Discussion of results
5. Drafting of the manuscript
6. Approval of the final version of the manuscript

MS has contributed in: 1, 3, 4, 5, 6.

MM has contributed in: 2, 6.

RR has contributed in: 1, 4, 5, 6.

### **Editor's opinion:**

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