Association of generalized joint hypermobility with history, signs, and symptoms of temporomandibular joint dysfunction in children

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Abstract

This study determined the prevalence of signs and symptoms of temporomandibular joint dysfunction (TMD) in children with and without generalized joint hypermobility (GJH). Twenty children with GJH, ages 4–19, and 20 age- and sex-matched control children completed a TMD signs/symptoms history and underwent an examination consisting of palpation of the joints and associated musculature for tenderness, clicking, or crepitation. Children with historical or clinical findings were designated positive for TMD signs/symptoms. Maximum vertical opening, expressed as a percentage of lower facial height, and maximum mandibular lateral excursion (in mm) were recorded. Fifteen (75%) of the GJH participants and ten (50%) of the controls were positive for TMD findings. There were statistically significant differences between the groups for the presence of total positive findings ($P < 0.001$) and for responses to palpation of muscle or joint ($P = 0.03$). There were no significant differences in positive responses to the history alone, joint palpation alone, or muscle palpation alone. There were no significant differences between the groups in jaw excursions. This study suggests that children with GJH may be more likely to demonstrate some signs and symptoms of TMD than children with normal joint mobility. (Pediatr Dent 15:323–26, 1993)

Introduction

Joint hypermobility has been considered a clinical problem since the end of the 19th century. It was not until 1967, however, that the syndrome of generalized joint hypermobility (GJH) was described by Kirk et al.[1] as "joint laxity associated with musculoskeletal complaints," but without demonstrable systemic rheumatology. This definition excludes heritable disorders of connective tissue or bone, such as Ehlers-Danlos syndrome, Marfan syndrome, osteogenesis imperfecta, and others. GJH in otherwise healthy individuals has been estimated to affect 5–7% of school-age children and 4–5% of adults,[2,3] though these figures underestimate its prevalence because many individuals with joint laxity are asymptomatic.[4,5] Also, physicians are generally trained to look for restriction of joint movement, rather than increased range. GJH affects females 4–8 times more often than males.[6] The condition diminishes with age, though perhaps not until the mid-20s in men and mid-40s in women.[6]

GJH is diagnosed by a series of five joint maneuvers designed by Carter and Wilkinson[7] or their modifications by Beighton.[8] Patients able to achieve three or more of the maneuvers are considered to have GJH. Pauciarticular involvement has been estimated by one study to affect 47% of young adult males and 78% of young adult females, prompting the suggestion that the definition of hypermobility be extended to those who can complete even one of the Carter and Wilkinson maneuvers.[6]

Individuals with GJH frequently display signs and symptoms of joint overuse trauma. Traction injuries at the insertion sites of ligaments and tendons are readily identifiable, as are synovitis, disc prolapse, and lesions. Arroyo et al.[9] found that 50% of GJH children ages 5–19 years had a history of arthralgia, compared to only 20% of a control group. Gedalia et al.[10] identified juvenile episodic arthritis in 66% of a group of 5- to 17-year-olds with GJH, compared to 12% of a group of normal schoolchildren. Joint instability, recurrent dislocation, and recurrent subluxation also are common in GJH. Among the joints so affected is the temporomandibular joint (TMJ).[11]

In recent years a relationship between GJH and TMJ dysfunction (TMD) has been suggested.[12,13] Bates et al.[14] found a significant relationship between wrist and elbow laxity and internal derangements of the TMJ in adult females, but not in adult males. Greenwood[15] found no relationship between hypermobility of the wrist and maximum mandibular opening or a TMD index in young adults. She stated, however, that there is no reason to assume that the TMJ is not affected in GJH and therefore not at risk for musculoskeletal problems similar to other joints. McCarroll et al.[16] demonstrated a significant correlation between a peripheral joint laxity score and mandibular opening in adult males, but not in females.

Harnistein et al.[17] found a 52% prevalence of GJH among 40 adult patients with severe TMD in a prospective study. They suggested a cause and effect relationship between joint laxity and TMD. Buckingham et al.[18] identified a similar percentage of GJH individuals in a prospective series of 70 patients with TMD. Westling[19] found a significantly higher prevalence of TMD (83%) among young adult females with GJH than those without GJH (41%).

Few studies have evaluated this relationship in children. Westling and Mattiasson[20,21] evaluated correlations between TMD and GJH in 193 adolescents ages 16–19. Twenty-two per cent of the females and 3% of the males

were classified as extremely hypermobile. No significant differences in symptoms between males and females with GJH were found. However, among GJH adolescents significantly more symptoms occurred in those who reported oral parafunctions (nail biting, gum chewing, tooth clenching). GJH was deemed an unfavorable factor that seemed to predispose adolescents with a history of trauma and oral parafunctions to TMD. Agerberg[22] found that active mandibular movements were significantly related to maximal fingerspread in 13-year-old children.

The purpose of this study was to assess the prevalence of signs or symptoms of TMD in children and adolescents with and without GJH.

Methods and materials

A total of 40 children, ages 4–19, were evaluated in this study. Twenty children diagnosed with generalized joint hypermobility (GJH) were compared to 20 age- and sex-matched controls. The examiner was blind as to each subject's joint status until the oral/TMJ examination was completed and the GJH screening maneuvers were tested.

The study was approved by the Institutional Review Board for studies involving human participants. After obtaining informed parental consent for participation, the parents completed a brief medical history and a TMD signs/symptoms questionnaire that gathered information on pain from the joint and related structures, joint sounds, TMJ trauma, juvenile arthritis, and previous orthodontic treatment. Positive answers to TMJ trauma, juvenile arthritis, or previous orthodontic treatment disqualified children from the study.

All children were examined by one of the authors (CH) who was trained in the TMJ examination criteria of Gross and Gale[23] and the modified Beighton maneuvers. The maximum interincisal distance was measured by having the patient open as wide as possible. A millimeter ruler was placed on the maxillary and mandibular incisal edges adjacent to the dental midline, the patient was asked to open wider if possible, and the measurement was recorded. The millimeter measurement of overbite (or openbite) was added to (or subtracted from) the maximum interincisal distance to achieve a measurement of maximum incisal opening. The distance from soft tissue point A (A') to soft tissue pogonion (Pg') was measured, and the ratio of maximum incisal opening to A' – Pg' was calculated in order to express maximum incisal opening as a percentage of lower facial height. This allowed comparison of maximum mandibular opening across patients of different sizes. Extreme lateral excursive movements were measured from the maxillary dental midline to the corresponding point in the mandibular arch. A measurement of lateral mobility (mm) was obtained by adding the values for left and right lateral excursions.

Joint clicks (single irregularity) and crepitus (multiple irregularities) were recorded by palpation and auscultation. The temporalis, masseter, medial pterygoid, and sternocleidomastoid were palpated bilaterally; the lateral pterygoids were palpated unilaterally. The mastoid processes and orbital ridges were palpated bilaterally as controls to qualify the results of sensitivity to palpation. Responses to either palpation or auscultation were recorded as present or absent. Participants were classified as positive for TMD signs/symptoms if there was clinical or historical evidence of joint pain, muscle pain, or joint sounds.

Following the oral/TMJ examination, each subject underwent a hypermobility examination of the wrist, thumb and fingers, elbow, knee, and trunk, as modified from Beighton.[2,8] Participants were asked to attempt: 1) passive hyperextension of the fingers parallel to the extension aspect of the forearm; 2) passive apposition of the thumb to the flexor aspect of the forearm; 3) hyperextension of the elbow ≥ 10°; 4) hyperextension of the knees ≥ 10°; and 5) flexion of the trunk with knees straight and palms resting on the floor. Maneuvers 3 and 4 were measured with a goniometer. Children able to accomplish three or more of these maneuvers were classified as hypermobile.

Results

The mean age of the GJH and control groups was 9.7 (± 1.02 SD) years. The ratio of females to males was 4:1. Fifteen (75%) of the GJH patients and 10 (50%) of the controls were positive for one or more characteristics of possible TMD. This difference was not statistically significant. However, the GJH group exhibited a greater number of positive responses in each of the dichotomous areas studied (Table 1). We elicited 32 positive responses from the GJH patients for historical or clinical evidence of sounds or pain in the TMJ or associated musculature. From the controls we elicited 16 positive responses. There was a significant difference in the distribution of positive responses to palpation of muscle or joint ($P = 0.03$), and for all positive findings combined ($P < 0.001$, Table 1). However, a series of Mantel-Haenzel[24] tests found no significant differences in the distribution of positive responses to joint palpation only ($P = 0.11$), muscle palpation only ($P =$

Table 1. Comparison of positive clinical and/or historical findings for TMD in children with GJH and controls, with one-tailed *P*-values for Mantel-Haenzel chi-square tests of differences in distribution

Group	*History*	*Joint Sound*	*Palpation of Muscle or Joint*	*Total Positive Findings*
GJH	10	9	13	32
Control	6	5	5	16
x^2 *P*-value	0.17	0.11	0.03	<0.001

Table 2. Comparison of lateral jaw excursions in mm (mean, SE) and opening ratio for children with GJH and controls

Excursion	*GJH Group*	*Controls*	*P-value*
Opening ratio	0.98 (0.028)	0.96 (0.025)	0.69
Lateral excursion	21.1 (0.93)	20.2 (1.04)	0.80

With *P*-values of paired *t*-tests; Opening ratio is calculated as (maximum interincisal opening: A' – Pg').

0.11), history of TMJ signs/symptoms ($P = 0.17$), and joint sounds ($P = 0.11$). No control subject responded positively to muscle palpation.

Table 2 gives the mean measurements of the opening ratios (maximum incisal opening: A' – Pg ') and lateral excursions of the GJH and control groups. Paired *t*-tests found no significant differences between the two groups in mandibular movement.

Discussion

This study suggests that children with GJH may demonstrate a higher prevalence of signs and symptoms of TMD. Fifty per cent more GJH children (15) than controls (10) were positive for palpation of muscles or joints, historical evidence of TMJ pain, or joint sounds. This distribution would have become significant for even a slightly larger sample. Twice as many positive responses indicative of possible TMD were found among GJH subjects. However, significant differences could not be demonstrated for every individual indicator of possible TMD.

This study may have overestimated the prevalence of TMD in the two groups, though the prevalence of positive findings in both groups was consistent with previously reported figures for children.[25] The history, in particular, may have identified a number of children in whom TMD signs and symptoms were episodic. It is unlikely that all those with positive histories would have progressed to the development of TMD. The transient nature of TMD signs and symptoms in children has been documented previously.[25]

Fifteen of the GJH children were not selected randomly, but represented a self-selected sample of GJH patients from a larger group previously diagnosed by a rheumatologist. Five additional GJH participants were examined as presumed controls, but were diagnosed as having GJH after completing the modified Beighton maneuvers. The ratio of females to males in the final GJH group was 4:1, consistent with ratios reported in other studies.[6,9]

Vertical and lateral jaw excursions were not significantly different between the two groups. This may be explained by several factors. First, females generally exhibit greater peripheral joint mobility than males. Males, however, by virtue of their greater size and larger masticatory muscle mass generally can open wider than females.[26,27] Thus, unaffected males in the control group may have been able to open as wide as some females with GJH. Second, the age range of children in the study also may have affected TMJ mobility findings. Increasing age is associated with decreased peripheral joint mobility, but also with greater mandibular excursions. Older children, who might be expected to demonstrate less joint mobility because of increasing age, may offset any age-related reduction in jaw opening by their greater size. Other studies[21,28] have noted significant correlations between body size or facial morphology and mandibular opening. This study concurs with those [15,29] that found no relationship between maximum mandibular opening and peripheral joint laxity. Others, however, have demonstrated such a relationship in adolescents[22] and in adult males.[16] Third, a linear measurement does not fully describe the ginglymoarthrodial nature of mandibular opening. The combination of hinge and sliding movements, however, precludes the use of a goniometer to measure its movement, as is done with other joints. Finally, mandibular excursions may not be consistent indicators of TMJ dysfunction except when noticeably restricted.

Two of the maneuvers used to diagnose GJH are passive extensions of joints. It may be the case that GJH children could demonstrate greater passive jaw opening with a mouth prop, for example, but potential damage to the joint by over-opening precludes such a test in a study of this nature.

The modified Beighton maneuvers are simple and reliable diagnostic tests for GJH.[2,6,7,8] When evaluating a child for possible TMD, this diagnostic screening should be used to determine the presence of GJH. Children with GJH and their parents should be counseled regarding the possible effects of joint laxity on the TMJ. Referral to a rheumatologist for further consultation should be considered.

Conclusions

1. Children ages 4–19 with GJH exhibited a higher prevalence of positive responses to muscle or joint palpation and total combined positive historical or clinical responses than did an age- and sex-matched group with normal joint flexibility.
2. Vertical and lateral jaw excursions may not be reliable indicators of TMD in children unless those movements are restricted.

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