Temporomandibular Involvement in Juvenile Idiopathic Arthritis

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ABSTRACT. ***Objective.*** To study occurrence as well as clinical signs and symptoms of temporomandibular joint (TMJ) involvement in juvenile idiopathic arthritis (JIA) in a population representing all subtypes of JIA.

Methods. Ninety-seven consecutive children with JIA underwent orthodontic evaluation including an orthopantomogram (OPG). Further evaluation included patient characteristics, disease onset, course, and medical treatment.

Results. Forty-five percent of all children had TMJ involvement. Frequencies according to JIA subtypes: systemic 67%, oligoarticular (persistent and extended) 39%, rheumatoid factor (RF) negative polyarticular 59%, RF positive polyarticular 33%, enthesitis related arthritis 13%, psoriatic arthritis 33%, and other arthritis 50%. In children with a polyarticular course, irrespective of their disease onset, TMJ involvement was more frequent (55% vs 31% in oligoarticular course). In children with disease onset at a young age and/or an extended course of the disease, TMJ involvement was also more frequent. Pain during jaw excursion, absence of translation, asymmetry during maximal opening and protrusion, as well as crepitation during evaluation are predictors for TMJ involvement with a good specificity but a low sensitivity. Not all patients with TMJ involvement have clinical signs.

Conclusion. Because of the high prevalence and discrepancy between clinical signs and presence of arthritis of the TMJ, regular orthodontic evaluation and OPG is recommended to recognize TMJ involvement and enable early intervention. (J Rheumatol 2004;31:1418–22)

Key Indexing Terms:
JUVENILE IDIOPATHIC ARTHRITIS TEMPOROMANDIBULAR JOINT

Juvenile idiopathic arthritis (JIA), formerly known as juvenile rheumatoid arthritis (JRA) and juvenile chronic arthritis (JCA), is a chronic inflammatory disease present for longer than 6 weeks starting before the age of 16 years[1]. The criteria for JIA were established by the International League of Associations for Rheumatology (ILAR) in 1994 in Santiago, Chile, and revised in 1997 in Durban. JIA is divided into 7 subtypes based on clinical symptoms during the first 6 months of the disease[1].

Underdevelopment of the mandibula is a characteristic feature of patients with JIA, recognized as early as 1890[2]. Involvement of the temporomandibular joint (TMJ) was first reported by Still when he described chronic arthritis in childhood in 1897[3]. The reported frequency of TMJ involvement varies from 17% to 87% in the literature depending on the population investigated, the subtypes of JIA represented, and the methods by which involvement is diagnosed[4-6]. In all subtypes of JIA, one or both TMJ can be involved and may even be the initial joints involved[4,7,8].

The most important site of growth of the mandible is the chondrogenic zone on the articular surface of the condylar head[9,10]. As a consequence of this superficial position, the growth center of the mandibula is more vulnerable to damage of the surface, due to arthritis[8]. Early destruction of the chondral part of the condyle due to arthritis may subsequently affect mandibular development and growth[11-13]. A well known example of these complications is micrognathia, the so-called bird-face, a small backward-positioned jaw[5].

Unfortunately, in many cases TMJ involvement evolves without any symptoms, causing a delay in detection[7]. Clinical symptoms (pain, local morning stiffness, and impaired function) alone have been shown to be quite unreliable as indication of arthritis[5]. Decreased opening of the mouth, earache, and pain during eating, chewing or yawning can also point at TMJ involvement[4,6-8]. The severity of TMJ symptoms is directly related to the inflammatory variables

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Submitted July 4, 2003; revision accepted January 27, 2004.

of JIA. As the disease progresses it may lead to severe destructive changes in the TMJ.

It is therefore important to recognize clinical symptoms that indicate TMJ involvement in an early stage. Combining these symptoms with radiological abnormalities may lead to early detection of imminent condylar destruction. When clinical symptoms are detected by the pediatrician, this should lead to referral for an extra orthodontic evaluation.

As symptoms of TMJ involvement coincide with flares of disease activity, the primary goal is to control the synovitis with medication. Further, guidelines for conservative treatment that follow basic principles in rheumatology like heat and cold therapy and exercises to improve range of motion are added. After initial general medical care, treatment is aimed at normalization of mandibular growth in anterior and vertical directions[10]. In most patients orthodontic treatment can be carried out satisfactorily during growth by using both functional and fixed appliances[11,14].

Our aim was to study occurrence of TMJ involvement in a population representing all subtypes of JIA according to the Durban criteria and to determine which clinical signs are indicative of TMJ involvement.

MATERIALS AND METHODS

Patients. In this survey patients who visited the pediatric rheumatology clinic of the Sophia Children's Hospital over a period of 6 months were investigated. All children with JIA according to the Durban criteria were routinely referred for orthodontic evaluation, even in the absence of complaints of the TMJ region.

Data collection. Data were obtained from medical records and through clinical investigation. The following data were collected: patient characteristics (including sex, age at onset, and age at orthodontic examination), type of disease onset and course, disease activity, and laboratory data. Therapeutic interventions during the course of the disease were used as an indicator for the assessment of the disease activity and severity since onset of the disease. Drug treatments considered were nonsteroidal antiinflammatory drugs (NSAID), disease modifying antirheumatic drugs (DMARD), and immunosuppressive drugs such as corticosteroids and methotrexate.

The data collected by the orthodontist included: mandibular function estimated by measuring (in mm) the interincisal distance at maximal mouth opening adjusted for overbite, protrusion adjusted for overjet, side deviations at maximal opening and protrusion, and lateral movements between the midlines adjusting for midline discrepancy at intercuspal position. Other symptoms, such as masticatory problems and difficulty in opening of the mouth, were recorded as present or absent. Full-face photography was performed for assessment of facial asymmetry and thorough documentation, which makes comparison during followup or reevaluation possible.

Radiological examination consisted of an orthopantomogram (OPG) taken after clinical examination and scored according to Rohlin[15] by 2 blinded examiners (MT and MLMM). The scoring system of Rohlin for TMJ involvement consists of 6 grades: 0 normal, 1 slight abnormality, 2 definite early abnormality, 3 moderate destructive abnormality, 4 severe destructive abnormality, and 5 mutilating abnormality[15]. We defined TMJ involvement as present when the score was grade 1–5.

Although OPG is not the most sensitive method in detecting resorption and change in the cranial area, it can be used to show joint involvement[11,16]. Magnetic resonance imaging (MRI) or lateral tomograms may be more efficient in diagnosing early TMJ involvement; however, OPG is less expensive and patient exposure to radiation is less. OPG is widely used in most dental and orthodontic clinics and therefore the best tool for routine investigation[4,15].

Statistic analysis. Continuous group data were summarized as medians and interquartile ranges. Comparisons of continuous variables between the groups of children with and without TMJ involvement were performed by a t test for normally distributed variables, by a Mann-Whitney test for variables not normally distributed, and by a Pearson chi-square for categorical variables. To determine independent prognostic factors of TMJ involvement, simple and multiple logistic regression analyses were performed. The following variables were tested univariately as well as in a multivariate procedure: swelling, pain, pain during jaw excursion, morning stiffness, clicking and crepitation noticed by the patient and asymmetric opening and protrusion, or crepitation, absence of translation, limited rotation of the head, limited head extension and/or flexion, and clicking noted by the orthodontist during examination.

All analyses were performed with the SPSS version 10.0 package (SPSS-PC, Chicago, IL, USA).

RESULTS

Patients. Over 6 months, 110 children with JIA according to ILAR criteria visited the pediatric rheumatology clinic. Of these patients, 97 were evaluated by the orthodontist and 13 patients did not show up at their appointment, or refused to make one. The orthodontist had evaluated 19 of the 97 children earlier in the course of their disease. To include these 19 patients, orthodontic measurements and anamnestic data of the initial visit were obtained from medical records.

The survey consisted of 60 girls and 37 boys, with a mean age of 10 years, 8 months (range 2 yrs, 10 mo, to 18 yrs), mean age at onset 5 years, 10 months, and mean duration of the disease of 4 years, 9 months.

TMJ changes. OPG was performed for all children. In 44 of 97 patients (45%), TMJ involvement (grade 1–5) was found. The percentage of TMJ involvement in relation to the different subtypes of JIA and the course of the disease are summarized in Table 1. The frequency of TMJ involvement increased if the arthritis had progressed to a polyarticular course (55% compared to 31% in oligoarticular course). Of the 44 patients with TMJ involvement, 22 had unilateral

Table 1. JIA subtype and frequency of TMJ involvement (n = 97).

JIA Type and Course	n	% TMJ Involvement
Systemic	15	67
Oligo	1	0
Poly	14	71
Oligoarticular	41	39
Persistent	37	34
Extended	4	75
Polyarticular RF positive	9	33
Polyarticular RF negative	17	59
Enthesitis related arthritis (ERA)	8	13
Oligo	4	0
Poly	4	25
Psoriatic arthritis	3	33
Oligo	2	50
Poly	1	0
Other arthritis	4	50
Oligo	3	33
Poly	1	100
Total	97	45

involvement of the TMJ and 22 patients had bilateral TMJ involvement. In total, 66 condyles were involved: 28 right-side and 38 left-side. The Rohlin score for severity yielded 30 condyles with grade 1, 3 condyles with grade 2, 8 condyles with grade 3, 22 condyles with grade 4, and 3 condyles with grade 5. TMJ arthritis was associated with an earlier onset of the disease compared to JIA without TMJ arthritis (5 years vs 6 years, 7 months). The children with TMJ arthritis also had a longer duration of the disease than children without (5 yrs, 2 mo, vs 4 yrs, 5 mo).

Disease severity and activity in the past were assessed by means of drug intervention used as therapy. Of the patients treated with local corticosteroids, NSAID, and DMARD (n = 69), 29% had TMJ involvement. Of the patients treated with NSAID, DMARD, and immunosuppressive therapy (n = 28), 68% had TMJ involvement. Treatment as a sign of disease activity showed a linear by linear association with TMJ involvement ($p = 0.013$). If therapy included NSAID, DMARD, and immune suppressive drugs, chances of TMJ involvement increased 5-fold (odds ratio, OR, 4.75) compared with no medication.

Clinical signs of TMJ changes. Signs and symptoms that could indicate TMJ involvement are summarized in Table 2. Of the symptoms noticed by the patient during the course of the disease, only pain during jaw excursion ($p < 0.05$) was a valid predictor. If the other signs were present, the possibility of TMJ involvement increased, but not significantly ($p > 0.05$). Of the signs noted by the orthodontist during orthodontic examination, absence or impaired translation during maximal mouth opening, crepitation during jaw movement, asymmetric opening, and protrusion seem to be important predictors ($p < 0.05$). Clicking appeared to be a borderline sign ($p < 0.06$). Through a backwards selection procedure absence of translation during maximal opening ($p < 0.009$) and asymmetry during maximal opening and protrusion ($p < 0.021$) appeared to be the most important factors, as the other factors in Table 2 did not increase risk of TMJ involvement significantly ($p > 0.05$).

Orthodontists' observations are summarized in Table 3. Of the measurements performed by the orthodontist only maximal opening of the mouth and protrusion were significantly decreased in patients with TMJ involvement compared to those without.

Maximal opening capacity of the mouth in our population was also compared with the normal ranges for age described in different surveys, summarized in Table 4[17,18]. Maximal opening capacity of patients with TMJ involvement was in the lower range, but within the normal standard deviation. For protrusion, normal ranges are described in the literature for 10 and 15-year-old children (10-year-old mean protrusion 10 mm, 15-year-old mean protrusion 9.7 mm)[7,17]. In our survey, children aged 9 to 11 years with TMJ involvement (n = 4) had a decreased mean protrusion (7.3 mm) compared to those without (n = 7, mean protrusion 9.5 mm). In children aged 14 to 16 years there was no difference in mean protrusion in those with (n = 6, protrusion 9.9 mm) and without (n = 6, protrusion 9.25 mm) TMJ involvement.

DISCUSSION

In this cross-sectional survey of pediatric patients including all JIA subtypes according to the Durban criteria, the mean frequency of TMJ involvement was 45%. A considerable variation depending on subtype was seen, with the highest prevalence in children with a polyarticular course. A wide variability in the frequency of TMJ involvement is reported in the literature (17% and 87%); however, most surveys do not include all subtypes of JIA and in some, only older children and adolescents are evaluated[4,7,8,12,13]. The frequency of

Table 2. Symptoms of TMJ involvement in patients with JIA (n = 97).

Signs	Patients with Signs (%)	Patients with TMJ Involvement (%)	Odds Ratio	95% Confidence Interval	p
Noticed by the patient					
Swelling	5 (5)	4 (80)	5.6	0.6–5.2	0.17
Pain	11 (12)	6 (55)	1.6	0.67–12.1	0.53
Pain during jaw excursion	9 (9)	6 (67)	5.2	1.02–26.5	0.04*
Morning stiffness	8 (8)	4 (50)	1.4	0.32–5.8	0.72
Clicking	12 (13)	8 (67)	2.9	0.82–10.5	0.087
Crepitation	9 (9)	6 (67)	2.8	0.67–12.1	0.17
Orthodontic examination					
Asymmetric opening and/or protrusion	24 (28)	17 (71)	4.9	1.74–13.5	0.002*
Crepitation	8 (8)	7 (88)	10.1	1.2–85.5	0.011*
Absence of translation	22 (23)	16 (73)	4.9	1.7–14.1	0.002*
Limited rotation of the head	25 (28)	14 (56)	2.1	0.81–5.3	0.13
Limited head extension and/or flexion	36 (41)	17 (41)	1.3	0.56–3.1	0.52
Clicking	13 (13)	9 (69)	3.3	0.94–11.6	0.052

* $p < 0.05$.

Table 3. Jaw mobility and TMJ involvement in children with JIA (n = 97).

Symptoms	Patients Without TMJ Involvement		Patients with TMJ Involvement		p
	n	Mean, mm	n	Mean, mm	(t-test)
Protrusion	48	8.7	35	8.3	0.05*
Maximal opening	54	50.2	42	44.9	0.001*
Overjet	54	4.0	43	5.4	0.81
Overbite	53	2.4	43	1.4	0.54
Lateral movement					
Left	54	9.8	42	9.4	0.41
Right	54	10.5	42	8.9	0.77

* $p < 0.05$.

Table 4. Maximal opening in JIA by age category compared to normal mobility. Ranges are shown in brackets.

Age	0–6 years	6–11 years	11–16 years	16–21 years
Ingervall[17]		49 mm (37–66 mm)		51 mm (33–64 mm)
Sheppard[18]	42 mm (35–49 mm)	46 mm (37–53 mm)	51 mm (40–72 mm)	49 mm (38–59 mm)
This study	n = 9	n = 15	n = 24	n = 6
Patients without OPG alterations	43 mm (37–51 mm)	48 mm (38–56 mm)	53 mm (38–71 mm)	53 mm (50–56 mm)
	n = 12	n = 13	n = 13	n = 4
Patients with OPG alterations	42 mm (33–50 mm)	43 mm (20–51 mm)	47 mm (40–56 mm)	57 mm (45–57 mm)

TMJ involvement also depends on the radiographic tool used to diagnose TMJ involvement. In our survey OPG was used to study the occurrence of TMJ abnormalities in JIA.

Our study population was relatively young compared to other studies, and all subtypes of JIA were represented. The highest risk of developing TMJ involvement was in systemic and polyarticular JIA (67% and 50%). Children with an oligoarticular JIA with a polyarticular course also had an increased risk of TMJ involvement (oligoarticular persistent 34%, oligoarticular extended 75%). The high frequency of TMJ involvement in polyarticular JIA was accounted for by the polyarticular RF negative group in which the frequency of TMJ arthritis was 59%. Surprisingly, the polyarticular RF positive group, known for an erosive destructive character, had a relatively low frequency of TMJ destruction on OPG (33%). The RF positive patients had late disease onset compared with the other subtypes. We postulate that older age may make the condyle less vulnerable to damage by arthritis than in young children. When most of the growth of the mandibula is completed and the growth center is closed, arthritis of the TMJ will have less devastating consequences.

In contrast with our expectations, unilateral and bilateral involvement were equally divided in our patients, while in other reports bilateral involvement was more frequent than unilateral involvement[7].

Of all symptoms noted by the patient, only pain during jaw excursion was a (weak) predictor of TMJ involvement (Table 2). Abnormalities on orthodontic examination that were significant and good predictors for TMJ involvement included absence of translation during maximal opening of the mouth, asymmetry of the maximal opening, and protrusion and crepitation (Table 2).

A trained pediatrician/rheumatologist can detect these predictors. Translation is the second part of the normal jaw movement during opening, the frontal movement of the condyle after the initial rotation. TMJ involvement should be suspected when only rotation is observed during lateral palpation of the TMJ. During this lateral palpation, asymmetry at maximal opening can also be observed. However, the fact remains that the sensitivity of these predictors is less than the specificity. Absence of translation, for example, having a sensitivity of 37% and a specificity of 89%, is a good predictor for involvement: if translation is present, TMJ involvement remains a possibility. Even when different tests were combined, no conclusive predictors for TMJ involvement were found.

We agree with Stabrun, *et al*[19] that impaired lateral rotation of the head is not an independent clinical predictor for TMJ involvement. In patients with cervical spine involvement, pain during movement of the head, reduced rotation, extension, and flexion are experienced because of interaction of symptoms. Patients with systemic or polyarticular onset have an increased risk of developing cervical spine involvement.

Measurements analyzed in Table 3 are more limited than those of Table 2 because they depend on normal growth and mobility of the mandibula. These predictors vary according

to age and should therefore be compared with healthy controls. The literature provides only normal values for maximal mouth opening and for protrusion[7,17,18]. There is a significantly smaller maximal opening capacity in patients with TMJ involvement compared to those without[7,10,12,13,19]. This difference may even be underestimated, since patients with initial stages of TMJ involvement with impaired opening but no visible OPG alterations are classified as having no TMJ involvement. Values for maximal opening in our patients were within the normal range (Table 4), although if TMJ involvement was present, the maximal opening was just in the outer limits of normal. In summary, impaired opening of the mouth is an important indicator of TMJ involvement that should be checked regularly.

There was a significantly decreased protrusion capacity in patients with TMJ involvement compared to those without. We were able to compare protrusion measurements of only our 10 and 15-year-old patients with controls because data on normal protrusion values are not available for other age groups. Since we had only a small number of children in these age categories, no definite conclusions can be drawn.

For the clinician it is important to realize that involvement of the TMJ may be present without signs and symptoms. Therefore every physical examination of a patient with JIA should include the TMJ region. In case of doubt about possible involvement of the TMJ, the patient should immediately be referred to an orthodontist. Even if there is no clinical suspicion of TMJ involvement, periodic evaluation with OPG is recommended for all children with JIA. In our hospital it is standard policy to perform an orthodontic examination in every patient at least once a year; the optimal interval between examinations has not yet been defined.

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