Resorption of the Temporomandibular Condylar Bone According to Subtypes of Juvenile Chronic Arthritis

THOMAS KLIT PEDERSEN, JENS JØRGEN JENSEN, BIRTE MELSEN, and TROELS HERLIN

ABSTRACT. ***Objective.*** To evaluate the relative impact of sex, type of onset, course of disease, age at onset, duration of disease and status of HLA-B27, antinuclear antibodies (ANA), and rheumatoid factor on the risk of developing a condylar erosion.

Methods. Condylar changes of the temporomandibular joint (TMJ) were diagnosed on orthopantomograms from 169 consecutive patients with juvenile chronic arthritis (JCA). A multiple regression analysis was applied to establish the relative weight of the independent variables affecting the severity of the condylar erosion.

Results. It was found that 62.1% of the patients exhibited condylar resorption. The highest prevalence was seen in children with a polyarticular onset or course of disease and early age at onset and severe resorption was also frequent in these groups. Patients with positive ANA also had a high prevalence but with a mild degree of resorption. In contrast, HLA-B27 positive patients had a lower risk of TMJ involvement and resorptive changes of the condyle.

Conclusion. Polyarticular and early onset arthritis are associated with a high risk for TMJ involvement and a severe condylar bone loss can be expected. ANA positive patients have a high prevalence, and B27 positive patients have a low prevalence of TMJ arthritis but in both subgroups, the outcome of the bone resorptive process is mild. (J Rheumatol 2001;28:2109–15)

Key Indexing Terms:
TEMPOROMANDIBULAR JOINT JUVENILE CHRONIC ARTHRITIS SUBTYPES

Temporomandibular joint (TMJ) involvement and subsequently mandibular growth disturbances are common complications in juvenile chronic arthritis (JCA)[1-4]. Based on clinical examination, the prevalence of arthritis in this particular joint varies between 42 to 65%[5,6] and radiological hard tissue changes show similar figures varying between 41 to 62% of the patients with JCA[7,8]. The consequences of TMJ involvement with resorption of condylar and temporal bone are frequently disturbances in the development of the craniofacial complex[3,9-12] (Figure 1) and secondary disturbances in occlusal development[3,9,13,14].

Clinical observations indicate a marked variation in the detrimental effect of arthritis on the condylar area both with regard to type of destruction and velocity of the resorptive process. Due to relatively few symptoms and modest clinical findings, the accurate onset of TMJ arthritis can be difficult to assess and treatment therefore often aims at the correction of already developed severe growth disturbances instead of supporting normal growth[15]. Since orthopantomograms are used routinely in most orthodontic clinics, they should be used for the establishment of the recognition of hard tissue destruction. Our aim was to study the prevalence and degree of condylar resorption diagnosed on orthopantomograms in relation to gender, subtypes, age at onset, duration of disease and serological findings in juvenile chronic arthritis patients. Secondly, it was the purpose to evaluate whether the potential risk and grade of destruction of the mandibular condyle can be predicted by variables related to type and characteristics of the disease.

MATERIALS AND METHODS

Patients. One hundred and sixty-nine patients with JCA (114 females and 55 males with a mean age at onset of the disease of 5.4 years ± 3.8 and at radiographic examination 9.8 ± 3.5 years of age) consecutively referred from the Pediatric Rheumatology Clinic to the Department of Orthodontics between 1990 and 1996 were included in our study. The patients were diagnosed according to the EULAR criteria[16]. Data on sex, subtypes, disease course, age at onset, duration of disease, HLA-B27, antinuclear antibodies (ANA), and rheumatoid factor (RF) were obtained from the hospital records (Table 1). The 23 independent variables are shown in Figure 2. Duration of disease was the time from onset of JCA to the diagnosis of the TMJ involvement.

In the female group, pauciarticular onset was most frequent (68.5%), polyarticular onset was seen in 21.9% and systemic onset was seen only in 9.6%. In the male group 58.2% could be characterized as pauciarticular, 23.6% as polyarticular, and 18.2% as systemic (Table 1). Due to the low number of patients with psoriatic arthritis (5.9%, F/M:7/3) the males and females were pooled in the regression analysis with the pauciarticular type,

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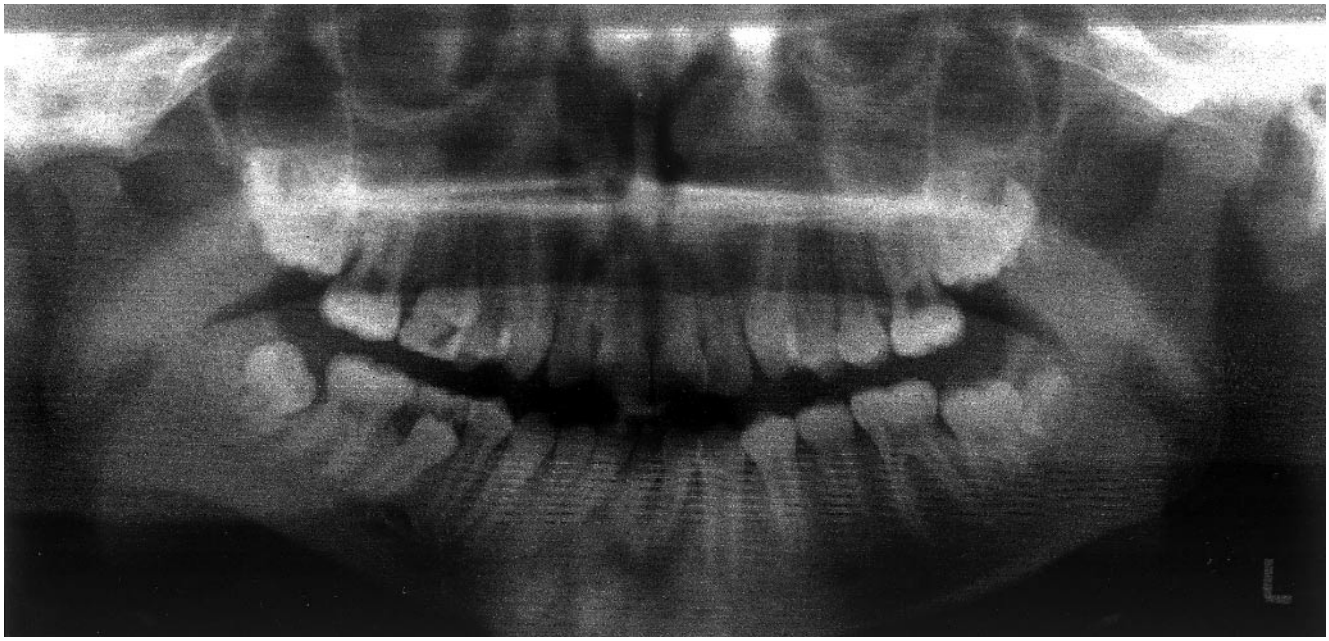

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as they all had pauciarticular onset. Age at onset and the time the radiographs taken were compared for girls and boys (Table 1). Mean duration of disease at time of investigation was 4.5 years (± 3.2) and did not differ for boys and girls. We found no significant difference between age at onset among the patients with systemic, pauci and polyarticular onset.

Laboratory variables. Of the 165 patients analyzed for HLA-B27, 15.8% were found positive with a higher representation of boys than in the HLA-B27 negative group ($p < 0.025$). Age at onset for the B27 positive group was 8.2 ± 4.0 years, and for the B27 negative group, 4.8 ± 3.5 years ($p < 0.0005$). Out of 161 patients, 54.7% were ANA positive (titers of 1:40 or above using a HEp2 cell assay). The M/F ratio did not differ in the 2 groups, but the mean age at onset was significantly lower for the ANA positive (4.4 ± 3.6 years) than for the negative patients (6.4 ± 3.7 years)($p < 0.001$). The ANA positive patients comprised 71.6% pauciarticular, 22.7% polyarticular, and 5.7% of the systemic onset type. Among the ANA negative patients, 54.8% were of the pauciarticular type while 23.3% were polyarticular and 21.9% were of systemic onset type. IgM RF (> 20 IU/ml) was found in only 5 (3.1%) patients: 2 with systemic onset, 2 with polyarticular and one with oligoarticular onset arthritis, all with a polyarticular course.

Radiographic evaluation. Two orthopantomograms, one in occlusion and one with maximal opening were taken to assess the presence of condylar resorption. Condylar resorption was graded into one of 4 categories (Figure 3) according to severity[13,17]: Grade 0: no radiological abnormalities; Grade 1: defects in the condylar border, minor erosions; Grade 2: flattening of the condyle; Grade 3: total destruction of the condyle[3,9-12].

Figure 1. A: Asymmetric growth disturbance of the craniomandibular complex deviating to the right of an 11 year old boy with pauciarticular JCA. B: Resorption of the right condyle with failure of development of the ramus height in the same patient.

Table 1. Mean age at onset and for radiographic examination.

	Age at Onset	Radiographic Exam.	Systemic		Poly	Pauci		HLA-B27		ANA		Rheumatoid Factor	
			PC	NPC		PC	NPC	Pos	Neg	Pos	Neg	Pos	Neg
Female	5.0 ± 3.7	9.6 ± 3.4	7	4	25	25	53	12	98	63	44	4	104
Male	6.3 ± 3.9	10.1 ± 3.8	8	2	13	8	24	14	41	25	29	1	53
Total	—	—	21		38	110		26	139	88	73	5	157

Systemic: systemic onset, Poly: polyarticular onset, Pauci: pauciarticular onset, PC: polyarticular course, NPC: non-polyarticular course.

Statistical analysis. The prevalence and severity of TMJ involvement in the different groups were compared with Pearson's chi-square test. In order to evaluate the impact of variables characterizing the general disease on the prevalence and the severity of TMJ involvement, a backward stepwise logistic regression analysis was applied. As this analysis requires the variables to be dichotomized, the groups with Grades 2 and 3 were pooled and tested against the group with Grade 1 resorption. Grades 1 and 2 were likewise pooled and tested against Grade 3.

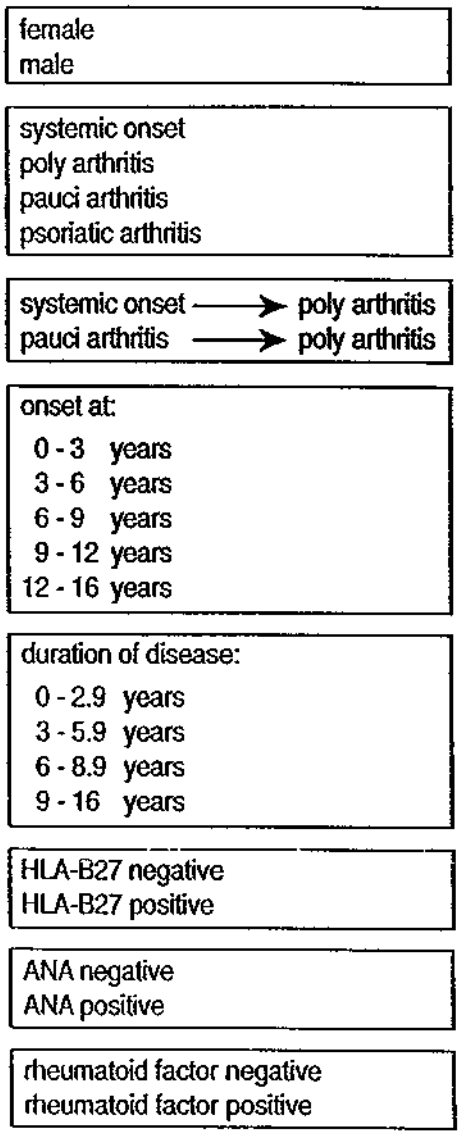

Figure 2. Independent variables tested for relation to prevalence and severity of condylar resorption of the TMJ.

A multiple regression analysis was applied to establish the relative weight of the independent variables describing the general disease on the severity of the condylar resorption expressed as a score. This made it possible to express severity as a continuous variable. The score was found in the following way: the grades of resorption in the right and left joints were added and the combination 2 and 0 was considered worse than 1 and 1, as was 3 and 0 compared to 2 and 1. Due to a strong correlation between the 2 joints of the same individual with respect to the grade of resorption in spite of the differences in appearance of involvement of the right and left joint, this was considered acceptable.

Within each of the subgroups, the distribution of different TMJ involvement and degrees of condylar involvement was compared by a Mantel-Haenzel test and Pearson's chi-square test[18-22] where possible. P values < 0.05 were considered significant.

RESULTS

Intraarticular hard tissue changes of one or both TMJ were found on radiographs in 105 (62.1%) of the 169 patients corresponding to an involvement of 49.1% of the total number of joints. Forty-two (40.0%) of the patients with affected TMJ had involvement of one joint, 22 of the right and 20 of the left joint. The distribution of unilateral involvement was equal in all the different subgroups. In the case of bilateral involvement, the affect and severity of the left and right joints were highly correlated ($r = 0.57$). The prevalence of affected TMJ was higher in girls (53.1%) than in boys (40.9%, $p < 0.05$). Due to a higher number with Grade 3 resorption, 6.4 vs 3.9%, bone resorption was slightly more severe in boys.

Patients with polyarticular onset had affected TMJ more frequently (65.8%) than patients with pauciarticular onset (44.5%, $p < 0.01$) and systemic onset type (42.9%, $p < 0.025$). Patients with polyarticular onset had more severe resorption ($p < 0.01$) than patients with pauciarticular type. There was no significant difference in severity between the systemic and the polyarticular type. The TMJ involvement was significantly more frequent in children with systemic or pauciarticular onset having a polyarticular course than in the patients without.

TMJ involvement was more frequent in the case of early age of disease onset and more severe resorption was observed in the age group 3-6 years compared to 6-9 years ($p < 0.05$). Longer duration increased the risk of involvement and also the severity was related to duration ($p < 0.05$).

HLA-B27 positive patients had TMJ involvement less frequently (38.5%) than patients negative for B27 (54.0%, $p < 0.05$). In addition, severity of bone destruction was higher

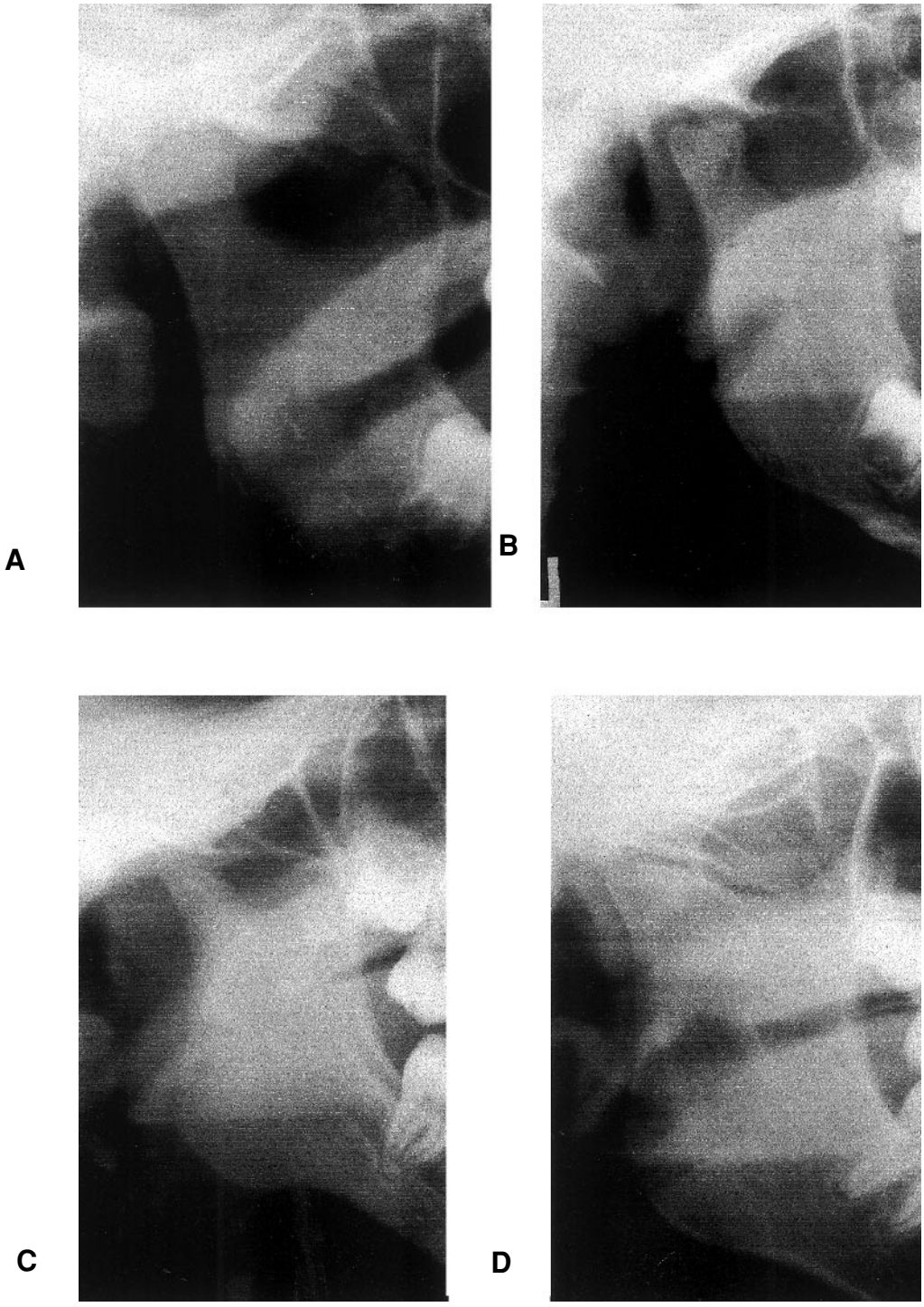

Figure 3. Orthopantomograms showing the images of the condylar head used for classification of the different grades of resorption. A: Grade 0, no radiological signs. B: Grade 1, defects in the condylar border, minor erosions. C: Grade 2, flattening of the condyle. D: Grade 3, total destruction of the condyle.

in B27 negative patients ($p < 0.025$). No Grade 3 involvement in the B27 positive patients was observed. Patients with positive ANA had a higher frequency of TMJ arthritis (55.1%) than the ANA negative group (40.4%, $p < 0.01$). The prevalence of Grade 3 resorption on the other hand was higher in the ANA negative group ($p < 0.01$). In the RF positive group, severe TMJ resorption was seen in the patients with early disease onset.

Analysis of all independent variables. Among the independent variables, polyarticular onset type and early onset age had the highest impact on the involvement of the TMJ. A polyarticular course and ANA positivity also had a significant influence. The female patients tended to have more joints involved. This relationship disappeared if onset age was included in the equation instead of duration of disease (Table 2).

Polyarticular onset with long disease duration increased the severity of resorption. As expected, the variable early disease onset was similarly responsible for TMJ involvement as duration of disease. Patients with negative ANA were also at risk for severe TMJ involvement. Patients with positive HLA-B27 have significantly less severe bone resorption (Table 3). This was confirmed by the backward stepwise analysis which found the duration of disease and polyarthritic type responsible for extended resorption while ANA positive and HLA-B27 positive patients were not at risk for severe resorption.

To analyze the influence of the variables describing the general disease on the severity of resorption, it was necessary to change from the dichotomous analysis to a multiple regression analysis. Thus, the dependent variables changed from the number of TMJ to the number of patients due to the strong agreement between right and left joints ($p < 0.001$).

First, the total group was studied including the patients without TMJ involvement. Three of the variables turned out to influence the TMJ score (Table 4). Increasing score of condylar resorption was strongly related to early age of onset or duration of disease and polyarticular type. Patients with a polyarticular course showed significant severity in bone destruction compared to the other patients.

Applying the multiple regression analysis to the group of patients with affected TMJ, the duration of disease or age at onset, patients with a polyarticular onset and a polyarticular course were still responsible for extended resorption (Table 5). HLA-B27 and ANA positive patients with TMJ involvement had significantly less severe condylar destruction.

DISCUSSION

Our study related arthritis of the TMJ to the subtypes of juvenile chronic arthritis. To optimize therapeutic strategy, it is important to identify patients at risk for a TMJ involvement since the use of a functional appliance to reduce the load on the joint during active arthritis can minimize the detrimental effect of the disease[15,23]. If, on the other hand, destruction of the condyle and the subsequent abnormal growth is already present, the simple orthopedic principles can no longer be used. Repetitive functional and structural analysis of the TMJ and facial skeleton combined with radiographic examination are recommended for patients with JCA. Although orthopantomograms are not optimal in detecting resorptions and changes in the cranial area[17], they can show joint involvement and thereby help define the subgroups of JCA. In this context, it is an available method in most orthodontic clinics.

Our findings support previous studies that consider TMJ involvement as resorption of the articular surfaces[7,8,24,25]. We found that 62.1% of our patients had radiological changes as a consequence of TMJ arthritis, which is comparable to the prevalence in other studies[7,8,24,25], but it is likely that the resorption has been preceded by synovial inflammation usually associated with discrete symptoms and clinical signs[26].

Among children with TMJ involvement, the higher representation of girls (73.3%) corroborated previous findings[13,25], but the influence of sex disappeared when age at onset was used in the equation. Also, the early onset[27] and duration of the disease significantly influenced the frequency and severity of the bony lesions[24,28,29]. The higher representation of girls with TMJ involvement may have actually been due to the fact that a large group of girls in our study had disease onset from 0-3 years of age.

Without distinguishing between different subtypes, Rönning and Väliaho[24] found that minor lesions were seldom seen. We were not able to confirm this. If a minor lesion (Grade 1) can be considered a stage in a process leading to a flattening or destruction of the condyle (Grades 2 and 3), we can conclude from our results that the pathological process either develops slowly in the pauciarticular type or resorption stops without serious lesions. Evolving a polyarticular course in the systemic and oligoarticular onset subtypes is perceived as an aggravation of the disease[30]. The severity of resorption is more pronounced in the systemic and polyarticular onset type, confirming earlier descriptions of increased severity of resorption following an increase in the number of other peripheral joints involved[7,8,10,29].

Table 2. Variables with a significant influence on the risk of developing TMJ involvement.

	Age at Onset Used in Equation		Duration of Disease Used in Equation	
	Regression Coefficient	p	Regression Coefficient	p
Poly onset	1.6839 ± 0.3484	< 0.0001	1.4799 ± 0.3294	< 0.0001
Poly course	0.7602 ± 0.287	< 0.01	0.8208 ± 0.2866	< 0.005
ANA pos	0.5234 ± 0.2613	< 0.05	0.6705 ± 0.252	< 0.01
Onset age	–0.1978 ± 0.0464	< 0.0001	—	—
Duration	—	—	0.1273 ± 0.252	< 0.002
Female	—	—	0.4928 ± 0.2648	= 0.0627

Table 3. Variables with a significant influence on the resorption grade (Grade 1 vs Grades 2 and 3). Patients with a polyarticular onset and long duration of disease are at higher risk for Grades 2 and 3 resorption while patients with positive HLA-B27 or ANA are at a lower risk.

	Regression Coefficient	p
Poly onset	1.982 ± 0.8563	< 0.03
Duration	0.2231 ± 0.701	< 0.002
B27 Pos	–1.3617 ± 0.6489	< 0.04
ANA Pos	–0.9629 ± 0.4607	< 0.04

Logistic regression analysis of all independent variables, testing exclusively patients with TMJ involvement.

Table 4. Significant influence of polyarticular onset, polyarticular course, and early age at onset on TMJ involvement is confirmed.

	Regression Coefficient	p
Poly onset	1.32 ± 0.23	< 0.001
Poly course	0.64 ± 0.21	< 0.01
Onset age	–0.23 ± 0.03	< 0.0001

Multiple regression analysis of all variables, testing all patients.

Table 5. Severity of condylar resorption was confirmed for patients with polyarticular onset, polyarticular course, and long disease duration. B27 and ANA positive patients had less severe condylar resorptions.

	Regression Coefficient	p
Poly onset	0.86 ± 0.206	< 0.001
Poly course	0.497 ± 0.206	< 0.02
Duration	0.115 ± 0.27	< 0.001
B27 Pos	–0.569 ± 0.268	< 0.04
ANA Pos	–0.566 ± 0.181	< 0.003

Multiple regression analysis of all variables, testing exclusively patients with involvement.

The time for diagnosing TMJ involvement is influenced by the fact that resorption is believed to be present for a period before the bone destruction is radiographically detectable[31]. A preresorptive phase, in which the joint may be inflamed but without showing resorptive changes as observed on MRI scans, has been described[26]. This weakens the value of age as a predictor in preference to disease duration until the radiographs are taken. Thus, the resorptive changes increase with duration of the disease.

Since the polyarthritis subtype has a late onset age in our study, the influence of age at onset and subtype of JCA could not act synergistically. In the subgroup of pauciarticular onset with a polyarticular course, the number of girls was relatively high, which is also considered as a confounding effect regarding the high prevalence of TMJ destructions among girls.

The pathophysiology of TMJ involvement in psoriatic arthritis was similar to that of the pauciarticular type but the number of patients with psoriatic arthritis was low. Among the characteristics of psoriatic arthritis is asymmetric joint involvement[32]. This could not be confirmed in our study as the frequency of unilateral involvement of the TMJ did not differ from that of other subtypes. Neither could the difference in involvement of right and left joints[33] be confirmed.

Patients with positive ANA apparently had a higher frequency of TMJ involvement but lower severity than ANA negative patients. This is likely to be strongly influenced by other factors but significance was achieved in our logistic regression analysis. A positive ANA is mainly seen in girls with early onset[34] and in our study, the ANA positive group had a significantly lower age at onset, was associated with the pauciarticular group, and was dominated by girls although not to a significant level. On the other hand, the ANA negative group mainly consisted of patients with systemic and polyarticular subtypes, which might explain the higher frequency of Grade 3 lesions in this group. The number of children with positive RF in this study was small, only 3.1%, compared to other studies[35]. Children with RF positive JCA often have a severe, destructive arthritis with a bad prognosis[36].

The presence of positive HLA-B27 is mainly found in boys, representing 60% in our study, and the same sex ratio was found in B27 positive patients with TMJ involvement. B27-associated JCA is characterized by a late onset[37], which may have an influence on the low prevalence of TMJ arthritis in this subgroup. However, logistic regression analysis demonstrated B27 as a significant factor for avoiding severe resorption of the condyles.

In conclusion, polyarticular and early onset arthritis carry a high risk for TMJ involvement and a severe condylar bone loss can be expected. Patients with a systemic or pauciarticular onset arthritis may have a higher risk and increased severity of condylar resorptions if the course is polyarticular. ANA positive patients have a high prevalence and B27 positive a low prevalence of TMJ arthritis but in both subgroups the outcome of the bone resorptive process is mild.

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