

Dento-alveolar Changes and Need of Orthodontic Treatment in JIA Patients Followed from Childhood to Adulthood

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Abstract

Objectives: To determine the occlusal traits and the objective orthodontic treatment need in a group of JIA children, with and without radiographic TMJ involvement, and compare with the objective orthodontic treatment need in adulthood.

Methods: Fifty-three JIA children (mean age 11.2 years) were studied; 30 had radiographic TMJ involvement (TMJ+ group) and 23 had no involvement (TMJ- group). Measurements were performed on dental study casts using the Need of Orthodontic Treatment Index, with 106 gender and age matched controls. Of the 53 JIA patients, 41 had available dental study casts in adulthood that were compared to childhood study casts.

Results: In childhood, TMJ+ subjects had an overjet ≥ 9 mm more frequently than controls ($p < 0.037$). More TMJ+ subjects were categorized with "great treatment need" than controls, 23.5% versus 9.5% ($p < 0.042$), respectively. As adults, 34% had a history of orthodontic treatment; 7.3% with orthognathic surgery. Only 5% could be categorized with "great treatment need" compared to 24% in childhood ($p = 0.012$).

Conclusions: JIA children with TMJ involvement were more susceptible to a large overjet, leading to "great treatment need" more frequently than controls. In adulthood the occlusion had improved due to orthodontic treatment and orthognathic surgery and probably, due to craniofacial growth and development. In growing JIA patients it is important to monitor the occlusal development.

Keywords: Dental occlusion; Longitudinal JIA study; Rheumatic disease; Temporomandibular joints

Abbreviations:

TMJ: Temporomandibular Joint; JIA: Juvenile Idiopathic Arthritis

Introduction

Juvenile idiopathic arthritis (JIA) is one of the most common rheumatic diseases of childhood [1]; approximately one out of 1000 children in the Scandinavian countries below 16 years of age are diagnosed with this disease [2]. Arthritis in the temporomandibular joint (TMJ) during childhood and youth may affect condylar growth. This is considered the main reason for the altered facial structure and changes in the dental occlusion in JIA patients [3-5], as expressed from the rather extensive literature on the topic. However, the number of longitudinal studies is very limited [6-9]. In a series of investigations, we have shown the outcome of the disease in a group of 60 patients followed for almost 30 years. In adulthood the mandibular dimensions are smaller (micrognathia), the mandible more retrognathic and the mandibular plane angle steeper in JIA patients than in healthy controls [10]. As many as 27% of the patients had micrognathia, all in the group with bilateral TMJ involvement. Growth disturbances were found in 70% of these patients [11]. Craniofacial growth disturbances were found in 57% of the entire series of patients, including all

subtypes of JIA, except one group with only one patient. In another study, we found that a progressing JIA TMJ disease course was correlated to a posterior mandibular growth rotation, and an improving disease course to an anterior mandibular growth rotation [12].

In JIA patients, dento-alveolar adaptations may compensate for abnormal skeletal relationship between maxilla and mandibula. Proclination of the lower incisors is frequently seen to adjust for sagittal Class II relationship. An anterior open bite may develop with posterior mandibular growth rotation with insufficient dento-alveolar compensation [13,14]. However, Larheim and Haanaes 1981 [3], Rønning et al. 1994 [15] and Fjeld et al. 2009 [10] did not find a difference in the lower anterior facial height and the prevalence of anterior open bite in JIA patients compared to healthy subjects.

The present study focused on the dento-alveolar development and need of orthodontic treatment in JIA patients followed longitudinally until adulthood. This has, to our knowledge, not previously been reported. Most published longitudinal studies in JIA patients investigate craniofacial development. Therefore, the objectives of the present study were to (1) assess occlusal traits as well as (2) the objective orthodontic treatment need in a group of JIA children, related to presence or absence of radiographic TMJ involvement, compared with a group of healthy individuals, and (3) compare the objective orthodontic treatment need in the same patients in childhood and adulthood, related to TMJ involvement.

Material and methods

The Regional Committee for Medical Research Ethics, Southern Norway approved the study protocol (Ref. nr. 2012/2120/REK sør-øst A).

JIA study population

The JIA group studied originated from 103 JIA patients (71 girls, 32 boys) participating in a longitudinal study initiated in 1976-79, with a mean age of 9.0 years (range 2.5 - 16.4 years) and a mean age at disease onset of 5.5 years (0.8 - 14.5 years). The patients have previously been thoroughly described [4]. None of the patients had received steroid injections in their TMJs or any surgical TMJ treatment during childhood and youth.

As adults, the patients were invited to participate in a follow-up study. Sixty were willing to participate, on average 27 years (range 23 -31 years) after the initial registration. For details, see Arvidsson et al. [16]. At adulthood, the patients had a clinical rheumatological examination and were assessed, according to the International League of Associations for Rheumatology (ILAR) classification [17] by one rheumatologist at the Department of Rheumatology, Oslo University Hospital, Rikshospitalet, with old and new patient files at hand [16]. They also had radiological examinations and a clinical dental examination, including plaster models of the dentition. Previous orthodontic- and orthognathic surgery treatment was recorded.

Individuals with study models of good quality, eruption of all permanent incisors and no history of orthodontic treatment at the childhood registration (T1) were included in the present study. Fifty-three JIA individuals (38 females and 15 males), mean age 11.1 years (range 8.0 - 14.8 years), met these criteria and their plaster models were evaluated for occlusal traits.

In the second part of the study, plaster models from adult age (T2) were evaluated. Forty-one (32 women, nine men) of the 53 patients from T1 had available plaster models. The remaining 12 were not willing to have dental impressions taken. To evaluate the long-term changes, plaster model assessments from the two stages (T1, T2) were compared.

Radiographic TMJ examination

JIA TMJ involvement at childhood registrations was evaluated by two maxillofacial radiologists (LZA, TAL) using a combination of radiographic methods, performed at the Department of Maxillofacial Radiology, University of Oslo [16].

At the follow-up examinations, the imaging methods were supplemented with CT and MRI, performed at the Department of Maxillofacial Radiology, University of Oslo and at the Department of Radiology, Oslo University Hospital, Rikshospitalet, and assessed by one general and two maxillofacial radiologists [18].

Healthy controls

Each of the 53 JIA patients was matched with two controls for gender and age at T1, which gave 106 controls with dental study models, from "The University of Oslo Craniofacial Growth Archives" [19]. The control subjects had no history of orthodontic treatment and no known joint diseases. The mean age of the controls was 11.2 years (range 8.9 - 15.0 years). There was no control material available for the T2 registrations.

Malocclusion traits and need for orthodontic treatment

The various occlusal traits were assessed using plaster models from the JIA patients and healthy controls at T1, and from JIA patients at T2. To measure overjet, overbite and space conditions to the nearest 0.5 mm, a ruler was used. Two of the authors (KB, MF) recorded the traits according to the Need for Orthodontic Treatment Index (NOTI) [20]. The index consists of four categories (A-D), depending on the severity of malocclusion. Category A indicates very great need for orthodontic treatment, category B great need; category C obvious need and category D little or no need of treatment. In this study, total treatment need calculated from the NOTI was the sum of individuals in category B and C only. No patients or control subjects were categorized to group A. Patients with category D traits only, were not included in the total treatment need group because of the minor need of orthodontic treatment. Twenty plaster models were examined twice by two observers (MF, KB) with an interval of at least four weeks.

Statistical analysis

Kappa statistics were used to analyse inter- and intra-examiner agreement of the NOTI. To analyse differences in traits between the JIA subjects and the healthy controls a chi-square test was used. Fischers exact test was used to test distribution of JIA subtypes according to the ILAR classification. A Student's t-test was used to analyse differences in overjet, overbite and crowding between the three groups. When comparing percentages at two different time points in the same patient group, McNemar's test was used for significance testing. The level of significance was set at $p \leq 0.05$. The SPSS 20 program (SPSS Inc., Chicago, IL) was used for the analysis.

Results

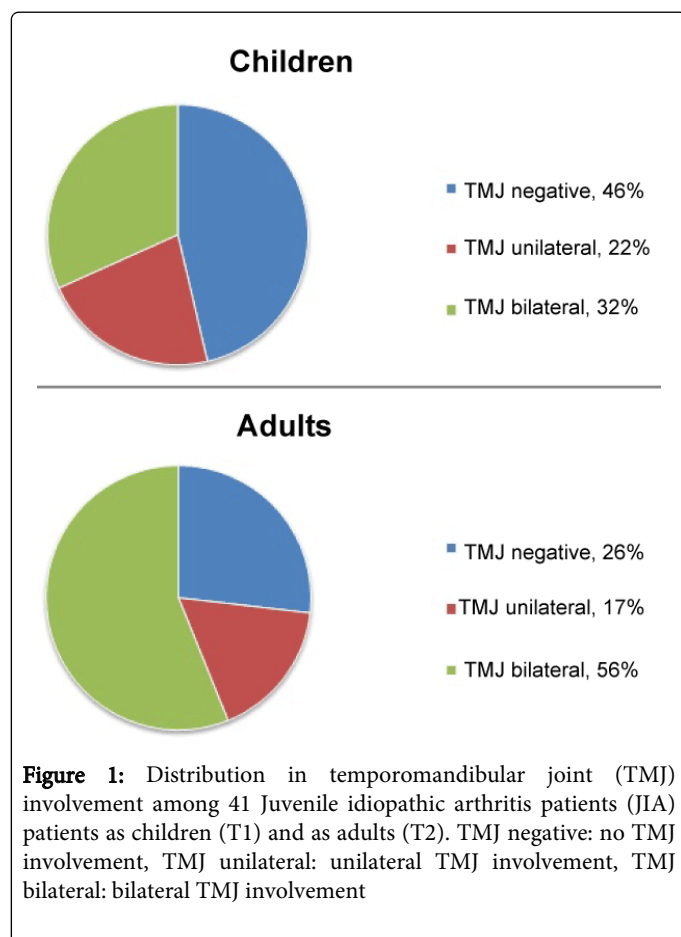
The method error study regarding the NOTI categories showed a Kappa value of 0.95 for inter- and intra-examiner agreement.

	TMJ+ n=30	TMJ- n=23	p-values
Disease onset age (years)	4.8 ± 2.8	6.8 ± 3.0	0.016
Mean age at initial registration (years)	11.3 ± 1.5	10.8 ± 1.4	0.32
ILAR classification			
Oligoarthritis	8 (27%)	4(17%)	
Extended oligoarthritis	10(33%)	4(17%)	
Polyarthritis, rheumatoid factor negative	6(20%)	7(30%)	
Polyarthritis, rheumatoid factor positive		1(4%)	
Systemic arthritis	4(13%)	3(13%)	
Enthesitis related arthritis	2(7%)	4(17%)	
Values are the mean ± SD, or the number of patients (%)			

Table 1: Age and JIA subtypes according to the International League of Associations for Rheumatology (ILAR) classification in 53 juvenile idiopathic arthritis (JIA) children, with (TMJ+) and without (TMJ-) temporomandibular joint involvement

Table 1 describes the disease onset age and distribution of ILAR JIA subtypes related to TMJ involvement. The disease onset age was 2 years lower in the group with TMJ involvement compared to those without ($p = 0.016$). No further significant differences were found, in subtypes, related to presence or absence of TMJ involvement.

At T1, in the total group of 53 subjects, 23 children (43%) had no TMJ involvement. Eleven (21%) had unilateral and 19 (36%) had bilateral TMJ involvement. In the 41 subjects with a follow-up examination, the distribution of TMJ involvement at T1 was 19 (46%) without TMJ involvement, nine (22%) with unilateral and 13 (32%) with bilateral TMJ involvement. At T2, the figures in the same 41 patients were eleven (27%), seven (17%) and 23 (56%), respectively. Figure 1 describes the development of TMJ involvement in the 41 JIA patients followed from T1 to T2. The frequency of TMJ involvement increased with 19% from T1 to T2. At the follow-up examination, 30 (73%) of the patients had either unilateral or bilateral involvement. Bilateral TMJ involvement increased significantly ($p = 0.026$), from 32% at T1 to 56% at T2.



At T1, in the total group of 53 subjects, the TMJ+ group had significantly more subjects in category B (great treatment need) than the healthy controls, 23.5% and 9.5% ($p = 0.042$), respectively. The only trait with a statistically significant difference was an overjet ≥ 9 mm found more frequently in the TMJ+ group than in healthy controls ($p = 0.037$).

Table 2 describes the distribution of NOTI categories in childhood and in adulthood, and the development of occlusion, showed by

change in NOTI categories, for the 41 patients followed longitudinally. Eighty-five per cent had the same or improved occlusion at T2. Only 2 (5%) of all the JIA patients could be categorized in group B (great need of orthodontic treatment) at T2 compared to 10 (24%) at T1 ($p = 0.012$). Total treatment need was reduced for the entire group of 41 patients, from 68% at T1 to 34% at T2 ($p = 0.002$).

	Total (n)	Adults, NOTI classification			
		B	C	D	
Children, NOTI classification	B 10		4 (3)	6 (4)	Total treatment need (B+C) in childhood was 68%
	C 18	2	4 (2)	12 (3)	
	D 13		4 (1)	9 (1)	
Total (n)	41	2	12 (6)	27 (8)	
		Total treatment need (B+C) in adulthood was 34%			

Numbers in parenthesis are the patients that received orthodontic treatment/orthognathic surgery (n = 14)

Table 2: Distribution of Need of Orthodontic Treatment Index (NOTI) categories in childhood and in adulthood in 41 patients with juvenile idiopathic arthritis (JIA) followed longitudinally. Change in occlusion, illustrated by altered NOTI category are showed by colours green (improvement in NOTI category from childhood to adulthood, $n = 22$ (53.5%)), yellow (same NOTI category in childhood and adulthood, $n = 13$ (31.5%)) and red (worsening in NOTI category from childhood to adulthood, $n = 6$ (15%)). Same or improved NOTI category (yellow + green) in $n = 35$ (85%)

	n	NOTI category at T2			Orthodontically treated patients	
		B (great need)	C (obvious need)	D (little/no need)	n	percent
TMJ+ at T1 and T2	22		9 (5)	13 (5)	10	45.5%
TMJ- at T1 and TMJ+ at T2	8	1	1	6 (1)	1	12.5%
TMJ- at T1 and T2	11	1	2 (1)	8 (2)	3	27%
Total	41	2	12 (6)	27 (8)	14	34%

Number in parenthesis is patients that received orthodontic/orthognathic surgery treatment

Table 3: Distribution of Need for Orthodontic Treatment Index (NOTI) categories in 41 juvenile idiopathic arthritis patients in adulthood (T2), related to temporomandibular joint involvement (TMJ+, TMJ-) in childhood (T1) and in adulthood (T2)

The distribution of NOTI categories at T2, related to TMJ involvement in childhood and in adulthood, is described in Table 3. Fourteen (34%) of the 41 JIA patients had received orthodontic treatment, three of these (7.3%) had been subject to orthognathic surgery.

In the TMJ+ group, a significant reduction in the number of patients categorized in group B between T1 and T2 was found ($p = 0.016$).

Discussion

The longitudinal design of the present study allowed us to examine malocclusions in a group of young JIA patients and evaluate the outcome almost 30 years later. Arthritis in the TMJ is considered the main reason for craniofacial growth deficiency, influencing dento-alveolar development in JIA patients [5,8,21]. Radiological examination is crucial in TMJ diagnostics and made it possible in the present study to differentiate between JIA patients with presence or absence of uni- and bilateral TMJ involvement.

The NOTI was chosen since occlusion and changes in occlusion can be measured by using severity graded morphological categories in accordance with health rationale for orthodontic intervention. Despite the fact that the NOTI is a rather rough index, a change in category can provide information about improved dental occlusion following orthodontic treatment or normal occlusal development, as shown in the present study.

The frequency of radiographic TMJ involvement in our study in childhood was in accordance with other studies of children [5,22]. An increase in the number of patients with TMJ involvement from childhood to adulthood could be expected but not to what extent; there are no studies to compare with. With a frequency of 73% TMJ involvement in adulthood, of whom the majority of patients had bilateral abnormality, one would have expected more occlusal disturbances than what was observed. On the other hand, again there are no studies to compare with.

The patients with TMJ involvement at T1 had disease onset two years earlier than those without TMJ involvement, in accordance with the study by Cannizzaro et al. [23]. Early disease onset and long disease duration seem to have an impact on craniofacial growth [4]. Thus, this might increase the risk of developing malocclusions. In the present study, no significant differences were found in distribution of JIA subtypes between those with and without TMJ involvement. However, the subgroups were small.

Regarding the NOTI classification, the only significant finding was a higher prevalence of large overjet in the JIA patients with TMJ involvement compared to controls at childhood. This is in accordance with findings by Karhulahti et al. [13]. TMJ involvement and reduced mandibular growth, including a steep mandibular plane in JIA patients [10], may be an important contributing factor to the increased overjet. The relationship between bilateral TMJ involvement and micrognathia in these patients was recently shown by Arvidsson et al. [11]. Further, in the patients with TMJ involvement, a significant reduction in category B traits, from childhood to adulthood, was found. The majority of them had an improved NOTI category due to orthodontic treatment, and some from intervention such as extraction of teeth. For the remainder the NOTI category may have improved because of craniofacial growth or dento-alveolar compensatory growth and development. This is in agreement with the findings that destructive

changes in the TMJ may heal over time [16,24], and that the mandible may regain its growth potential when the arthritis is inactive [12,25]. However, a reduction in total treatment need in the group with TMJ involvement during follow-up was non-significant. Further, the JIA patients without TMJ involvement did not differ from healthy control subjects regarding occlusal traits and distribution to NOTI categories.

In the total group of JIA patients, orthodontic treatment and orthognathic surgery had significantly reduced the need of further treatment in adulthood. This may result from careful orthodontic treatment in childhood. Adequate orthodontic treatment may compensate for the adverse effects of growth disturbances in JIA patients. Still, there was a less positive response to treatment found in the group with TMJ involvement from early age, maybe because of possible dysplastic dento-alveolar mechanisms related to impaired mandibular growth.

About 85% (34 patients) of the JIA patients showed no change or improvement in NOTI category over time. From the 10 patients categorized with a great treatment need (category B) in childhood, seven (70%) received orthodontic treatment. This placed them in category C and D, with obvious or little/no need for orthodontic treatment in adulthood. Among patients in category C and D in childhood, five (28%) and two (15%) received orthodontic treatment, respectively. The category B traits were treated more frequently, as expected. They are visible aesthetics discrepancies, and malocclusions connected to reduced dental function, compared to traits in category C. Treatment of the minor discrepancies in category D are mainly performed as a subjective wish from the patients or parents. Twelve of the 22 patients with an improved category placement from childhood to adulthood did not receive orthodontic treatment. Four of these had crowding, treated with extractions only. Three patients with a deep bite showed biteraising during growth. The remaining five patients had various occlusal traits that improved over time, probably due to growth changes.

Six patients (15%) were registered with a more severe category in adulthood than in childhood. Two developed a severe deep bite, from a moderate deep bite (one without TMJ involvement throughout the study and one that developed bilateral TMJ involvement between 11 and 13 years of age). Three subjects developed an anterior open bite. Among these three, two had bilateral TMJ involvement from before 6 and 10 years of age, and the third developed unilateral TMJ involvement between 13 and 37 years of age. The last one was registered with crowding of teeth as adult.

One could expect a greater orthodontic treatment need among JIA patients than in the average population. However, in the present study the number of JIA patients receiving orthodontic treatment was 34%, corresponding with healthy individuals in Norway (35%) [20]. This fact may be explained by periods of high disease activity and complex medical and surgical treatment that may have kept some patients from undergoing lengthy orthodontic treatment. Another factor may be limited knowledge of JIA and its possible effect on craniofacial growth and dento-alveolar development among orthodontists.

When one or both TMJs are involved during childhood, mandibular growth may be affected, and craniofacial growth disturbances are commonly seen [11]. Reduced mandibular growth will have an impact on choice of treatment. Orthopedic functional appliance, distraction splint type, has been suggested by Pedersen et al. [26] and Stoustrup et al. [27] with the aim to unload the TMJs and normalize mandibular growth. In the present study, the percentage of

JIA patients going through orthognathic surgery in adulthood (7.3%), was much higher than in the normal population (< 0.5%), illustrating the severe effect that JIA may have on facial growth and development. The correlation between bilateral TMJ involvement and micrognathia has previously been described [11]. This underlines the importance of carefully monitoring the patients during growth due to the risk of developing malocclusion. There is no consensus on development of particular occlusal traits in JIA patients, except for increased overjet as a result of micrognathia and/or posterior rotation of the mandible. In the present study, an anterior open bite was found in a few JIA patients, both with and without TMJ involvement. However, Barriga et al. [28] did not find any relationship between anterior open bite and JIA. This might be due to compensatory alveolar growth mechanisms such as increased vertical growth of the lower anterior alveolar ridge [7,21] or overeruption of mandibular or maxillary incisors.

Conclusion

The present study showed that children and adolescents with JIA and TMJ involvement more frequently had a large overjet and thus “great orthodontic treatment need” compared to healthy controls. JIA children with normal TMJs did not differ significantly from the control subjects regarding occlusal traits or orthodontic treatment need. As adults the occlusion of the patients was improved because of earlier orthodontic/ orthognathic surgery treatment and probably because of craniofacial growth and development; two thirds were diagnosed with little or no treatment need. However, the percentage of patients that had received orthognathic surgery was higher than in the normal population. To monitor the occlusal development in growing JIA patients is important.

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