Juvenile Arthritis and Development of Symptoms of Temporomandibular Disorders: A 15-year Prospective Cohort Study

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Aims: *To compare the development of symptoms of temporomandibular disorders (TMD) in a sample of patients with juvenile arthritis (JA) and a matched control sample.* ***Methods:*** *In 1986, 40 patients with JA (28 girls and 12 boys; mean age ± SD, 18 ± 4.5 years) and an age- and sex-matched control sample were examined for signs and symptoms of TMD. Fifteen years later in 2001, a questionnaire concerning symptoms of TMD was sent to these subjects. Twenty-eight individuals (68%) in the JA sample (20 women and 8 men; mean age ± SD, 35 ± 5.2 years) and 26 controls (19 women and 7 men; 34 ± 4.0 years) were available for the follow-up.* ***Results:*** *The overall prevalence of symptoms of TMD increased between the 2 examinations in both groups. The prevalence of reported TMD symptoms, such as jaw pain, fatigue in the jaws, and difficulty opening the jaws wide, as well as awareness of tooth clenching, headaches, neck and shoulder pains, was significantly greater among the JA sample than among the controls at the follow-up.* ***Conclusion:*** *The study indicates that prevalence of pain and dysfunction in the craniofacial or cervical regions of JA patients is increased more than 20 years after the onset of JA compared to healthy individuals.* J OROFAC PAIN 2007;21:120–126

Key words: epidemiology, juvenile arthritis, longitudinal study, orofacial pain, temporomandibular joint

Juvenile arthritis (JA) is an inflammatory joint disease of unknown etiology. The inflammation causes stiffness and pain and may damage components of the joint.[1] JA comprises several entities which differ in immunologic response, clinical manifestations, and prognosis. The individual course of JA is difficult to predict but in the long term, about one third of those affected can achieve remission.[2] Patients with oligoarthritis are more likely to have longer periods of inactive disease compared to patients with polyarthritis.[3]

According to Manners and Bower,[4] prevalence of JA ranges from 0.07 to 4.01 in 1,000, while incidence has been reported as 0.008 to 0.226 in 1,000 in different studies. The variations found may be explained by differences in the definitions used and by the use of small study populations. The prevalence and incidence of JA in Scandinavia are reported to be about 0.1% and 0.01%, respectively. Incidence usually peaks between birth and 4 years, and girls are more frequently affected than boys.[5] In a Scandinavian study, the incidence was 15 per 100,000 children; the number of cases per country ranged from 7 in Iceland to 23 in Norway.[6]

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Presence of symptoms of functional disturbances of the jaws in patients with JA varies between 26% and 74%.[7–9] Pain, stiffness, and temporomandibular joint (TMJ) crepitation have been the most frequently reported symptoms, while restricted maximal mouth opening capacity, pain during function, TMJ clicking sounds, and tenderness to palpation are the most common signs registered.[8] Children with JA are at risk of developing mandibular micrognathia due to destruction of the TMJ, decreased forward growth of the mandible, and posterior rotation of the mandible resulting in distal occlusion and a frontal open bite.[9,10] Other craniofacial and dentoalveolar abnormalities found in individuals with severe JA have been low posterior facial height, micrognathia, overerupted mandibular incisors, and a small interincisal angle.[7]

Few studies have been published on the long-term effects of JA on jaw function. Flato et al[11] conducted a prospective study of 316 patients with JA; after a median of 14.9 years of disease duration, they found that the disease was in remission for half of the patients; 24% had developed joint erosion, and 36% had impaired physical functioning. Generally, patients with JA had more disability, more bodily pains, and poorer general health than the controls. In another study, an 11-year follow-up study of 26 patients, those with JA of pauciarticular type had the best prognosis, while growth abnormalities and radiographic changes were more commonly found in those with polyarticular JA and systemic onset of the disease.[12]

The long-term effects of JA on the jaw, including the potential functional disturbances, are largely unknown. It is important to evaluate JA patients prospectively in order to evaluate the risk for contracting symptoms and disability in the craniofacial and cervical regions. The aim of this study was to examine a cohort of cases with JA and their matched controls 15 years after they were included in a study that examined the presence of mandibular dysfunction.[13] The specific aims were to compare the development of symptoms of temporomandibular disorders (TMD) in a sample of patients with JA and a matched control sample.

Materials and Methods

Study Population

The original cases were all referrals to the Departments of Pediatrics and Rheumatology at the University Hospital in Umeå, Sweden, where the diagnoses were also established. Inclusion in the study required the development of arthritic symptoms before 16 years of age and symptom duration of at least 3 months. Patients with psoriatic arthritis were excluded. Forty patients (28 girls and 12 boys) with JA fulfilled the criteria. Their mean age ± SD at examination was 18 ± 4.5 years. Age at JA onset ranged from 1 to 14 years, and the duration of the disease varied from 8 to 24 years.[13] Sixteen had a polyarticular form of JA; 24 had a pauciarticular form. A control was matched to each case for age and sex. The controls were chosen from the files of the Public Dental Health System in the city of Umeå, Sweden. None of the subjects in the control sample had been diagnosed with general joint disease or experienced any symptoms; their closest relatives were also free of joint disease. The study population completed a questionnaire on the presence of signs and symptoms of TMD; they were also examined clinically.[13]

Of the 80 individuals originally in the sample, 2 individuals had died by the time of the 15-year follow-up examination, and addresses could not be obtained for another 8 individuals. Therefore, the follow-up sample comprised 70 individuals, 36 with JA (24 women, 12 men) and 34 from the control group (22 women, 12 men). Fifty-four subjects (68% of the original sample) returned the questionnaire, 28 from the JA group (20 women, 8 men; mean age ± SD, 35 ± 5.2 years) and 26 from the control group (19 women, 7 men; mean age, 34 ± 4.0 years. Among the cases 9 individuals (32%) answered that they no longer had any joint disease, 12 did not know what type they had, 4 had polyarticular arthritis, and 2 had systemic arthritis (data are missing on 1 subject). Since the individuals were spread all over the country, it was not possible to conduct a follow-up clinical examination.

Questionnaire

In January 2001, questionnaires and stamped return envelopes were mailed to the 70 individuals. The questionnaire was essentially the same as the one used in 1986; most of the questions were yes-or-no questions. The first part concerned the subject's general health, medication, and chewing ability. The second part concerned the presence, frequency, and severity of jaw-related symptoms such as pain, difficulty opening the mouth wide, stiffness/fatigue, TMJ sounds, headaches, and neck pain. The third part concerned awareness of different parafunctions. The answers from the questionnaire were used to calculate the anamnestic dysfunction index (Ai) as devised by Helkimo.[14]

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For the 15-year follow-up, questions about employment, type of joint disease, quality of life, impairment of daily life, and utilization of health care owing to TMD were added to the questionnaire. The subject was asked to state the presence and frequency of each of the following symptoms: jaw or facial pain, pain on jaw movement, difficulty in opening the jaw wide, tiredness in the jaw, TMJ clicking, TMJ crepitation, TMJ locking, headaches, neck pain, and shoulder pain. For each symptom the following 5 alternatives were given: never, once or twice a month, once a week, several times a week, and daily. The answers that related to TMD symptoms were dichotomized before the statistical analysis as "present" or "not present." Presence of pain in the head, neck, or shoulders was dichotomized into "symptoms reported less than once a week" and "symptoms reported at least once a week." The subject's general state of health was self-reported as good, neither good nor bad, or bad. The subjects also reported all ongoing medicines prescribed by physicians. Questions on the ability to do household work unhindered, to move or walk freely, and whether their quality of sleep was good were answered with a yes or no. These questions were not specifically directed to symptoms in the craniofacial region. Utilization of health care because of TMD or pain in the head or upper spine region was ascertained by asking patients to state yes or no when asked whether they had consulted a dentist, a physician, a physiotherapist, a psychologist, or a chiropractor. Those who answered "yes" to 1 or more of the items were asked which of the following treatments they had received: surgery, steroid therapy, acupuncture, heat, analgesics, occlusal adjustment, splint, transcutaneous electrical nerve stimulation, massage. The subjects were also asked to state any other consultations they had had and any other treatment modalities they had utilized.

Dropout Analysis

In an analysis of subjects who dropped out, 4 cases and 5 controls were interviewed by telephone. The reported reason for not sending back the questionnaire was that they had no symptoms in the jaw-head region.

Statistical Methods

SPSS for Windows was used for the statistical analysis. Since the original database was not available, statistical analysis of individual changes between 1986 and 2001 could not be performed. The analysis was instead based on the cross-sectional data obtained in 2001 and compared on a group basis to data presented previously.[13] To test for differences in the distribution of symptoms, general health, and disability, the χ^2 test was used. If the expected count on 1 or more cells was less than 5, the Fisher exact test was used after transferring data to 2×2 tables. The level for statistically significant differences was based on 2-tailed calculations and a P value less than .05.

Results

The prevalence of reported symptoms of TMD among cases and controls at baseline and at the 15-year follow-up examination in 2001 is presented in Table 1, and the distribution according to Helkimo's anamnestic dysfunction index is shown in Fig 1. The prevalence of symptoms in the craniofacial region was higher at the follow-up examination compared to baseline data among both cases and controls. TMJ clicking sounds was the most common reported symptom among both cases and controls. At the 15-year follow-up, JA cases reported statistically significant higher prevalence of jaw and facial pain, impaired maximal jaw-opening capacity, and feelings of tiredness in the jaws compared to controls. None of the controls and few of the cases reported impairment in chewing (Table 1). Only 1 JA case reported difficulties in ability to bite off with the front teeth.

Reported habits of parafunctions and symptoms related to the general health at the examinations in 1986 and in 2001 are presented in Table 2. At the baseline examination, cases and controls did not differ significantly with respect to symptoms; however, in 2001 several symptoms were found significantly more often in the JA group. The JA cases were more aware of tooth clenching and grinding habits at both examinations, but the difference only reached a significant level for tooth clenching at the follow-up examination. The prevalences of headaches and neck pain in the JA group increased 3 and 5 times, respectively, from 1986 to 2001. Only 50% of the individuals with JA reported that they had good general health in 2001, compared to 77% of the controls. Among the JA patients who reported difficulties in opening the jaw wide, 90% also reported frequent pain in the neck and/or shoulder region (P = .02). All cases with frequent pain in the jaws and/or pain on jaw movements reported frequent pain in the neck/shoulder region.

Thirty-two percent of the JA group (9 individuals) reported that they had no ongoing joint dis-

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Table 1 Number and Percentage Distribution of Affirmative Answers Regarding Presence of TMD Symptoms Among Patients with JA (Cases) and Matched Controls

	1986					2001				
	Cases n = 40		Controls n = 40			Cases n = 28		Controls n = 26		
Symptom	n	%	n	%	*P*	n	%	n	%	*P*
Tiredness of the jaws	3	8	3	8	NS	13	46	5	20	.04
TMJ clicking sounds	9	23	6	15	NS	15	54	10	39	NS
TMJ crepitation	5	13	0	0	< .05	7	25	2	8	NS
Pain in face or jaws	3	8	2	5	NS	11	39	3	12	.02
Pain on jaw function	4	10	1	3	NS	8	29	3	12	NS
Difficulty in opening mouth wide	9	23	2	5	< .05	10	36	2	8	.02
Difficulty in chewing	0	0	0	0	NS	2	7	0	0	NS

NS = not significant (*P* > .05).

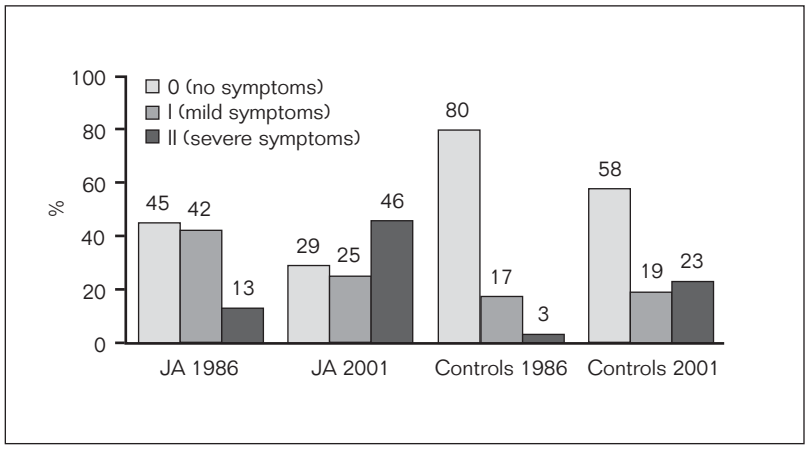

Fig 1 Percentage distribution of TMD symptoms in patients with JA and matched controls according to Helkimo's anamnestic dysfunction index (Ai) at the baseline examination in 1986 and the 15-year follow-up in 2001.

Table 2 Number and Percentage Distribution of Reported Parafunctions, Upper Spinal Pains, General State of Health, and Ongoing Medication Among Patients and Matched Controls

	1986					2001				
	Cases		Controls			Cases		Controls		
Symptom	n	%	n	%	*P*	n	%	n	%	*P*
Parafunctions										
Tooth clenching	14	35	8	20	NS	16	57	7	27	.03
Tooth grinding	10	25	5	13	NS	13	46	6	23	NS
Nail biting	9	22	8	20	NS	6	21	6	23	NS
Cheek and lip biting	12	30	16	40	NS	7	25	6	23	NS
General health										
Headache	5	13	5	13	NS	10	36	3	12	.04
Neck pain	4	10	4	10	NS	14	50	5	19	.02
Shoulder pain	-	-	-	-	-	15	54	4	15	.003
Impaired general health	3	8	0	0	NS	14	50	6	23	.04
Any prescribed medication	6	15	0	0	< .05	12	43	6	23	NS

NS = not significant (*P* > .05).

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Table 3a Number and Percentage Distribution of Previous Dental, Medical, and Paramedical Counseling Due to Craniofacial Symptoms Among Patients and Matched Controls in 2001

	Cases		Controls		
	n	%	n	%	*P*
All Consulting	16	57	12	46	NS
Dentist	9	32	2	8	.03
Physician	2	7	1	4	NS
Physiotherapist	7	25	3	12	NS
Psychologist	1	4	0	0	NS
Chiropractor	3	11	6	23	NS
Other	7	25	2	8	NS

Table 3b Number and Percentage Distribution of Reported Treatments for TMD Symptoms Among Patients and Matched Controls in 2001

	Cases		Controls		
Treatment	n	%	n	%	*P*
Surgery	1	4	0	0	NS
Steroid therapy	4	14	0	0	NS
Acupuncture	4	14	4	15	NS
Heat	5	18	2	8	NS
Analgesic	8	29	1	4	.03
Occlusal adjustment	4	14	0	0	NS
Splint	7	25	3	12	NS
Transcutaneous electrical nerve stimulation (TENS)	0	0	3	12	NS
Massage	8	29	9	35	NS

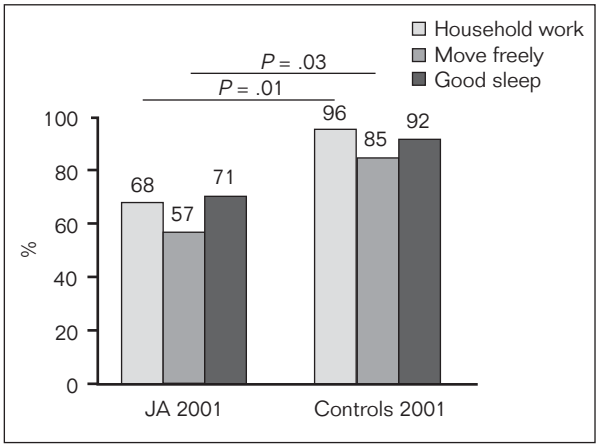

Fig 2 Percentage distribution of reported ability to perform normal household work unhindered, ability to move or walk freely, and good sleep quality in patients with JA and matched controls. *P* denotes the level of significance of the difference between cases and controls.

ease at the follow-up. Cases who stated that they had a joint disease (n = 19) had significantly more often frequent pain on jaw movements ($P = .03$) and difficulty in opening the jaw wide ($P = .01$) compared to those with no ongoing arthritis (n = 9). More than half of the sample had consulted dental or other health-care centers due to symptoms in the craniofacial region (Tables 3a and 3b). Almost one third of the JA sample had consulted a dentist because of TMD symptoms compared to 8% of the controls ($P = .03$).

Half of the cases reported difficulty in at least 1 area of daily living in 2001. They had significantly more difficulty in performing household work ($P = .01$) and impaired ability to move freely ($P = .03$). The JA group reported impairment in doing household work 8 times more often and difficulties in their locomotion 3 times more often than the controls (Fig 2). Those in the JA sample who reported an impaired ability to do household work, impaired movement and affected sleep were considered the most severe cases in this sample. This subsample comprised 5 subjects (18%), all of whom were female. All 5 of them reported pain in the jaws, difficulty opening the jaw wide, and neck/shoulder pain. For all statistically significant differences, the statistical power was found to be acceptable (> 0.8).

Discussion

The present long-term study could not at an individual level match and analyze changes in the status between the first examination and the follow-up examination. Consequently, the study does not have the power to show whether JA increases the risk of having pain and dysfunction in the craniofacial or cervical regions. The study, however, indicates that the prevalence of pain and dysfunction in these regions in JA patients was increased more than 20 years after the disease onset compared to healthy individuals, which in turn suggests a higher risk for pain and dysfunction in these patients.

The study was based on a case-control study with 40 individuals in each group. In long-term follow-up studies, dropouts frequently occur. Considering the long time-gap between the 2 examinations, a response rate of 68% was consid-

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ered acceptable. In addition, information about 9 of the dropouts was obtained and added to the results, which made the response rate 79%. The dropouts were relatively evenly distributed between cases and controls; thus, the study population could still be considered matched. The small sample size increased the risk of β error; ie, the risk of accepting a false null hypothesis. For all statistically significant differences, the statistical power was found to be acceptable (> 0.8). Therefore, despite the small sample size, the authors are confident in the conclusion that patients who develop arthritis at a young age run a higher risk of experiencing pain and dysfunction in the craniofacial or cervical regions later in life. The use of the questionnaire as well as the description of symptoms in accordance with Helkimo's anamnestic dysfunction index were based on the original examination and presentation of the material.

The cases included in this study seem to have followed the same path as those previously reported,[2,3,11,12] with regression of the disease for about 25% to 50% of the patients. The prevalence of symptoms of TMD increased in both cases and controls from late adolescence up to the early middle ages, which also is in line with results from longitudinal studies on TMD.[15–17] The controls reported, at both examinations, an overall prevalence of symptoms of TMD close to that expected from population-based studies.[15,16] The prevalence of severe symptoms (Ai II) among the controls was, however, somewhat higher than expected. The effect on prevalence of dysfunctional symptoms when defined criteria of frequency and intensity are used has recently been reported.[18] It is probably more clinically significant to consider the frequency with which symptoms occur than just their presence or absence. Because the present study was designed as a follow-up to the first study, the same criteria for prevalence were used. However, if only frequent symptoms were considered, only 4% of the controls and 36% of the cases would be classified as having severe symptoms (P = .015) at follow-up. The patients diagnosed with JA during their childhood reported significantly more TMD symptoms compared to their controls as well as more than were expected based on previous epidemiologic studies.[15,16,18,19] About twice as many (46%) of the cases reported severe symptoms of TMD compared to their controls at the follow-up.

The reported prevalence of bruxism among both cases and controls at baseline was in line with a population-based sample of approximately the same age, same study period, and the same region[19] and did not indicate any difference between the 2 samples. At the follow-up, awareness of bruxism had increased more in the JA group compared to controls and was twice as commonly reported among cases than among controls. The prevalence of bruxism among cases at follow-up exceeded the expected figure based on results from the general population.[17] Despite the high prevalence of jaw pain and functional disturbances in the masticatory system, very few reported difficulties in chewing food such as bread or meat, and few experienced any difficulties in ability to bite off with the front teeth. In this respect, the results were in line with previous studies[8,13] and may indicate a high level of adaptation to the changed masticatory system. Interestingly, few JA patients reported an impaired general state of health in the first study. In a recent study, children with JA reported that their health-related quality of life was better than their parents' reports had suggested.[20] This may reflect coping strategies among young people with arthritis.

The relationship between impaired jaw-opening ability and neck/shoulder symptoms is interesting. It has been shown previously that subjects with JA can develop severe damage to the TMJ.[21–23] Inflammation, pain, and internal derangements of the structures of the TMJ can restrict jaw mobility. The integrated function involving jaw and neck movements during jaw opening has been convincingly shown.[24] In a recent study of patients with JA, 62% were found to have inflammatory changes of the cervical spine, with apophyseal joint ankylosis at multiple levels.[25] Clinically these changes tend to limit neck movements. Involvement of cervical spine function among JA subjects may therefore affect jaw function.

Surprisingly, many individuals from both groups had consulted medical or dental practitioners for their symptoms in the craniofacial or cervical regions. The finding that about one third of the JA patients visited a dentist due to their symptoms indicates that a considerable part of their symptoms are located in the craniofacial region. A recent study indicated that well being in children with JA was related to 3 clusters of variables: pain "as normally is," number of pain-free days, and attendance at physical education classes.[26] It was reported that treatment of these children was primarily aimed at reducing chronic joint pain. However, dental management aimed at reducing the functional and/or parafunctional load to the masticatory system can be advocated for affected individuals.

In conclusion, the majority of JA subjects reported pain and dysfunction in the craniofacial

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or cervical regions as adults. Compared to that in a control sample, the prevalence of pain and dysfunction in these regions among JA patients was increased more than 20 years after the onset of the disease and indicates a higher risk for pain and dysfunction in the masticatory system among these patients.

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