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The Journal of Rheumatology

The Journal of Rheumatology

Volume 38, no. 10

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J Rheumatol 2011;38;2272-2273

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The Journal of Rheumatology is a monthly international serial edited by Duncan A. Gordon featuring research articles on clinical subjects from scientists working in rheumatology and related fields.

Temporomandibular Joint Arthritis in Pediatric Sjögren Disease and Sarcoidosis

To the Editor:

Arthritis of the temporomandibular joint (TMJ) is a well recognized entity in patients with juvenile idiopathic arthritis (JIA). It is usually painless and even minor radiographic changes of the TMJ have been associated with disruption in mandibular growth and a number of other craniofacial changes¹. Recently, magnetic resonance imaging (MRI) evaluations have proven to have much greater sensitivity to detect arthritis compared to ultrasound, and placed the prevalence of TMJ arthritis in this population between 63% and 75%^{2,3}. Due to this high prevalence, patients with JIA frequently receive contrast-enhanced MRI scans of the TMJ as a standard evaluation for arthritis in several clinical centers.

There are other rheumatologic diseases that can also present with TMJ inflammation [e.g., sarcoidosis, Sjögren disease, mixed connective tissue disease (MCTD)]^{4,5,6}, but the prevalence is less known. During the course of clinical care at the Pediatric Rheumatology Clinic at Children's Hospital of Alabama (Birmingham, AL, USA), several non-JIA arthritis patients have presented with symptoms and/or signs consistent with TMJ disease and were subsequently evaluated by MRI for TMJ arthritis. We describe the MRI findings of TMJ arthritis in these patients.

The protocol for this study was approved by the University of Alabama at Birmingham Institutional Review Board. Between September 2007 and December 2010, 75 patients with various non-JIA arthritis-associated rheumatic diseases (34 with Sjögren disease, 19 sarcoidosis, 22 MCTD) were identified by electronic medical records and studied. Diagnoses were based on the expert opinion of one of the 3 pediatric rheumatologist authors, in some cases based on strong clinical suspicion when lacking biopsy evidence. Acute TMJ arthritis was defined as the presence of effusion or synovial enhancement, and chronic disease as synovial thickening or evidence of bony changes (e.g., condylar flattening or erosions, both typical of TMJ arthritis). Electronic medical records, including clinical progress notes, laboratory results, and TMJ imaging reports of the 75 patients, were thoroughly examined and results documented.

A total of 18 (25%) non-JIA patients received MRI of the TMJ during the study period, and 89% of those had documented arthritis elsewhere. Of the 18 MRI evaluations, 7 (39%) showed evidence of TMJ arthritis. Ten of 34 (29%) patients with Sjögren disease were screened for TMJ arthritis by MRI; 4/10 had positive MRI evidence of TMJ arthritis. Five of 19 (26%) sarcoidosis patients were screened by MRI; 3/5 had positive MRI evidence of TMJ arthritis. Three of 22 (13.6%) MCTD patients were screened by MRI; 0/3 had positive MRI evidence of TMJ arthritis.

The characteristics of the patients are shown in Table 1. Subjective symptoms were present in 6/18 (33%), and 13/18 (72%) had clinical signs of TMJ dysfunction. The precise indication for MRI for 2 patients could not be deduced from clinical records. Common subjective symptoms noted

were pain and locking of the jaw. Common clinical findings included jaw deviation during opening and reduced maximal interincisor distance. Clinical characteristics for the 7 patients with radiographic evidence of TMJ arthritis (4 with Sjögren, 3 with sarcoidosis) are shown in Table 2. Only 2/7 (29%) of the patients found to have TMJ arthritis had subjective symptoms. The remaining 5/7 (71%) had a positive clinical finding (jaw deviation). None of the patients with abnormal MRI had both subjective symptoms and positive clinical findings. Concurrent peripheral arthritis was present on examination in 6/7 (86%).

Recent studies have concluded that MRI is the most sensitive^{2,3} and specific⁷ method for detecting TMJ arthritis in children with chronic peripheral arthritis. All patients in our study with non-JIA childhood arthritis and suspected TMJ disease were evaluated by this imaging method. Signs or symptoms of TMJ arthritis were found in all 7 patients with MRI-confirmed TMJ arthritis. This highlights why it is appropriate to look for masticatory muscle tenderness, restricted maximal interincisor distance, and deviation in mouth opening⁴. For children with JIA, the most common subjective symptoms noted in patients with TMJ arthritis are pain at rest, crepitation, swelling, and clicking⁸. While the sensitivity of symptoms and clinical findings may be low, the risk of overlooking this diagnosis in a child can be devastating, and it is therefore important to be cautious about any deviation from normal in a maxillofacial examination. Perhaps more concerning is the fact that in some studies the great majority of JIA patients with TMJ arthritis were asymptomatic³. This suggests that perhaps all children with chronic arthritis should be evaluated for TMJ arthritis.

Our data show that TMJ arthritis can be present in children with non-JIA inflammatory rheumatic diseases with chronic joint involvement, possibly with lower prevalence. Moreover, TMJ arthritis can be similarly destructive. Thus, a maxillofacial screening examination should be completed at each visit of a patient with non-JIA childhood arthritis, and subjective symptoms or clinical findings should prompt physicians to consider a TMJ MRI for further evaluation. We are currently evaluating the potential benefit of intraarticular triamcinolone hexacetonide injections of the TMJ in this cohort of patients with non-JIA chronic arthritis.

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Table 1. Characteristics of patient groups requiring MRI of temporomandibular joint.

Characteristic	Sjögren, n = 10	Sarcoidosis, n = 5	MCTD, n = 3
Sex, M/F	1/9	4/1	1/2
Age, mean yrs (range)	13.5 (4–19)	11 (9–16)	13 (13)
Race, White/African American	5/5	3/2	3/0
Peripheral arthritis present %	100	80	100
Symptoms, no. patients (%)	4 (40)	1 (20)	1 (33)
Locking	1 (10)	—	—
Pain	3 (30)	1 (20)	1 (33)
Clinical findings, no. of patients (%)	7 (70)	4 (80)	2 (67)
Reduced MIO	2 (20)	—	2 (67)
Asymmetric opening	7 (70)	4 (80)	2 (67)

MCTD: mixed connective tissue disease; MIO: maximal interincisor opening.

Table 2. Characteristics of patients with MRI-confirmed TMJ arthritis.

Patient	Age, yrs, sex	Race	Diagnosis	Serology	Peripheral Arthritis, (Y/N)	Indication for MRI	Original MRI Results
1	17 F	AA	Sjögren	ANA 1:640, speckled, SSA+, RF+	Y	Asymmetric opening	Mild Bi TMJ arthritis
2	13 F	AA	Sjögren	ANA 1:80, speckled, SSA+, RF-, HLA-B27-	Y	TMJ pain	Flattening of mandibular condyle, Bi synovial enhancement
3	14 F	AA	Sjögren	ANA 1:640, speckled, SSA+, RF-	Y	Asymmetric opening	Bi TMJ synovial enhancement
4	11 F	W	Sjögren	ANA 1:1280, speckled, SSA+, RF-	Y	Asymmetric opening	Bi TMJ synovial enhancement
5	14 M	AA	Sarcoidosis	ANA-, ACE 104*, RF-, HLA-B27-	Y	Asymmetric opening	Mild cartilage thinning and unilateral synovial enhancement
6	9 M	W	Sarcoidosis	ANA-, ACE 80, RF-, HLA-B27-	Y	Asymmetric opening	Bi TMJ synovial enhancement
7	14 M	W	Sarcoidosis (by LN biopsy)	ANA-, ACE 49, lysozyme 7.5, HLA-B27-	N	TMJ pain	Bi TMJ fluid, increased enhancement R TMJ

* Reference ranges: ACE 13–100; lysozyme 4.0–10.3. AA: African American; ACE: angiotensin-converting enzyme; ANA: antinuclear antibody; Bi: bilateral; LN: lymph node; R: right; RF: rheumatoid factor; SSA: Sjögren syndrome A antibody; TMJ: temporomandibular joint; W: white.

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