Periarticular calcifications and arthropathy as the first manifestation of polymyositis

Sirs,

Polymyositis (PM) is a systemic inflammatory disorder affecting skeletal muscles and other organs (1). Antisynthetase antibodies, including anti-J01 antibody, have been shown to be associated in PM patients with interstitial lung disease, Raynaud's phenomenon, a specific dermatitis known as "mechanic's hand" and arthritis (2-4). While joint manifestations are common in anti-J01 positive PM patients (2-4), only a few cases of deforming arthropathy have been reported, i.e. subluxation of interphalangal joints of the thumbs, periarticular hydroxyapatite calcifications and erosions of metacarpophalangeal and interphalangeal joints,and wrists (5-9). We describe a new case, which is of particular interest as the patient developed arthropathy and multiple periarticular calcifications associated with anti-J01 antibody revealing PM.

In March 1997, a 46-year-old woman was admitted for polyarthritis of one month duration. She also had a 6-month history of Raynaud's phenomenon. Physical examination revealed arthritis involving the hands and wrists. Laboratory findings were positive for antinuclear antibodies (1:600), whereas rheumatoid factors, lupus-like anticoagulant, anti-native-DNA and anti-centromere antibodies and cryoglobulin were negative; antibodies to extractable nuclear antigens (ENAs) were also negative: anti-Ro/SSA, anti La/SSB, anti-RNP, anti-Sm, anti-PM-Scl and anti-Scl 70 antibodies. Nailfold capillaroscopy was normal. Bone radiographs revealed juxta-articular demineralization at distal and proximal interphalangeal and metacarpophalangeal joints. A diagnosis of inflammatory polyarthritis of unknown origin was made. The patient was treated with nonsteroidal anti-inflammatory drugs; due to poor response of the joint symptoms, she was given prednisone at a dose of 25 mg/day; hydroxychloroquine therapy, which had been initiated concomitantly, was discontinued after one month due to adverse digestive effects.

In June 2000, the patient remained free of joint features, while receiving a 10 mg/day prednisone regimen, although she complained of shortness of breath on exertion. Physical examination revealed crackles over the basal lung fields. Laboratory data were normal, notably creatine kinase (CK) levels 61 IU/L. Autoantibody screen was positive for antinuclear antibodies (1:1000) with a speckled pattern; among anti-ENA autoantibodies, anti-J01 antibody was detected. Pulmonary function tests showed decreased diffusing capacity of carbon monoxide (66% of predicted values). Lung CT-scan revealed bibasilar pulmonary shadowing. Bone radiographs demonstrated periarticular calcifications and erosions of proximal and distal interphalangeal joints involving both hands. The steroid regimen was increased to 30 mg/day, resulting in disappearance of the pulmonary symptoms. In May 2002, the patient was still receiving prednisone (7 mg/day). Polymyositis diagnosis was performed by Bohan and Peter criteria (10): (i) symmetric muscle weakness (muscle power, gauged for 8 proximal muscles by a modification of the British Medical Research Council grading system (1), was 68 points); (ii) increased serum muscle enzymes (CK 2029 IU/L); (iii) myopathic changes on electromyography; and (iv) characteristic histological muscle damage. Other tests, including esophageal manometry and nailfold capillaroscopy, were normal. Autoantibody screening was positive for antinuclear antibodies (1:1000) with anti-J01 antibody. The patient also exhibited recurrence of polyarthritis in the hands and wrists and bone radiographs showed: peri-/intraarticular calcifications, and erosions of proximal and distal interphalangeal joints involving hands and wrists (Fig. 1). The patient was treated with prednisone at a dose of 1 mg/kg daily, which resulted in improvement of clinical manifestations. At 9 month follow-up, she remains free of clinical symptoms taking prednisone 15 mg/day.

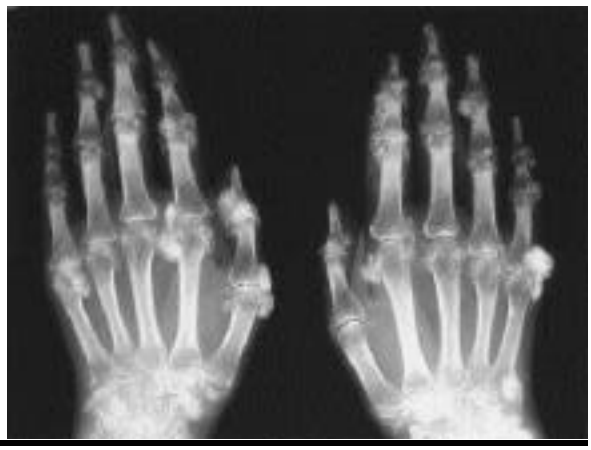

Fig. 1. Bone radiographs: Numerous periarticular and intra-articular calcifications at several levels in both hands and wrists, and erosions of proximal and distal interphalangeal joints involving both hands.

Joint involvement is common in anti-J01 positive PM patients (2-4). It is usually characterized by non-erosive and distal polyarthritis (2-4), and few cases of deforming arthropathy and periarticular calcifications of the hands have been reported in these patients (5-9). Our observation suggests that periarticular calcifications and arthropathy involving the hands may be included in the spectrum of joint complications in anti-J01 positive PM patients. It is also original in that arthropathy and periarticular calcifications revealed antisynthetase syndrome. We suggest therefore that antisynthetase syndrome may be suspected in patients with periarticular calcifications and arthropathy, even if they have no previous history of muscle features. Finally, because periarticular calcifications and arthropathy may precede other systemic events, when this type of complication is noted, an evaluation for antisynthetase syndrome with a search for anti-J01 antibody should be performed.

I. MARIE, *MD, PhD*
P. FOURNET, *MD*
A. JANVRESSE[1], *MD*
H. LEVESQUE, *MD, PhD*
H. COURTOIS, *MD, PhD*

Department of Internal Medicine and [1]Department of Radiology, Centre Hospitalier Universitaire de Rouen-Boisguillaume, 76031 Rouen Cedex, France.

Address correspondence to: Dr. Isabelle Marie, Department of Internal Medicine, Centre Hospitalier Universitaire de Rouen-Boisguillaume, 76031 Rouen Cedex, France.

References

1. MARIE I,LEVESQUE H,CAILLEUX N, COURTOIS H, GUEDON C: Intravenous immunoglobulin for treatment of gastrointestinal haemorrhage in dermatomyositis. *Ann Rheum Dis* 2001; 60: 723-4.
2. MARGUERIE C, BUNN CC, BEYNON HLC *et al.*: Polymyositis, pulmonary fibrosis and autoantibodies to aminoacyl tRNA synthetases. *Q J Med* 1990; 77: 1019-338.
3. TARGOFF IN: Immune manifestations of inflammatory muscle disease. *Rheum Dis Clin North Am* 1994; 20: 857-80.
4. VENABLES PJW: Polymyositis-associated overlap syndromes. *Br J Rheumatol* 1996; 35: 305-8.
5. BUNCH TW, O'DUFFY D, MCLEOD RA: Deforming arthritis of the hands in polymyositis. *Arthritis Rheum* 1976; 19: 243-8.
6. CITERA G, LAZARO MA, MALDONADO COCCO JA, DE BENYACAR MA, SCHEINES EJ, CIAFFI M: Apatite deposition in polymyositis subluxing arthropathy. *J Rheumatol* 1996; 23: 551-3.
7. COHEN MG, HO KK, WEBB J: Finger joint calcinosis followed by osteolysis in a patient

with multisystem connective tissue disease and anti-Jo-1 antibody. *J Rheumatol* 1987; 14: 605-8.
8. ODDIS CV, MEDSGER TA JR., COOPERSTEIN LA: A subluxing arthropathy associated with the anti-Jo-1 antibody in polymyositis/dermatomyositis. *Arthritis Rheum* 1990; 33: 1640-5.
9. QUEIRO-SILVA R, BANEGIL I, DE DIOS-JIMENEZ DE ABERASTURI JR,BELZUNEGUI-OTANO J, GONZALEZ-BENEITEZ C, FIGUEROA-PEDROSA M: Periarticular calcinosis associated with anti-Jo-1 antibodies sine myositis. Expanding the clinical spectrum of the antisynthetase syndrome. *J Rheumatol* 2001; 28: 1401-4.
10. BOHAN A, PETER JB: Polymyositis and dermatomyositis. *N Engl J Med* 1975; 292: 344-7 and 403-7.

Seasonal variation in dry mouth symptoms of Sjögren's syndrome patients: A clinical follow-up study

Sirs,

The prevalence of xerostomia is estimated to vary from 6.2% to 46% of the population (1-3). Generally subjective symptoms of dry mouth correlate with decreased salivary flow, even though this is not necessarily true on the individual level (4, 5). Because there are no longitudinal clinical studies on the seasonal variation in symptoms of oral dryness, our aim was to evaluate xerostomic symptoms in a clinical follow-up study monitored with subjective visual assessment (VAS) scoring.

Twenty-seven volunteers, 25 women and 2 men with a mean age of 52.9 ± 13.1 years, participated in the study group; 26 completed the study. They all suffered from subjective symptoms of dry mouth; however, they had macroscopically normal oral mucosa. All xerostomic subjects had one or more systemic diseases; the most common diagnosis being Sjögren's syndrome (SS) (15 subjects). The unstimulated saliva flow (UWS) was normal in 5 subjects, low in 14 subjects (0.1 - 0.25 ml/min) and very low in 8 subjects (< 0.1 ml/min).

We also recruited 25 controls (24 women and one man) without any symptoms of dry mouth. The mean age of the control subjects was 52.4 ± 17.2 years; 18 completed the study. All subjects signed an informed consent form. The Ethical Committee of the Faculty of Medicine, University of Turku, approved the study protocol.

This study was a clinical follow-up of subjective symptoms of oral mucosal dryness. The appointments were set at 12-week intervals. During the first appointment the subjects received information about the study, and underwent the baseline examination. They were given a mildly flavoured, detergent-free toothpaste to be used during the study. The examination included an interview about general health and medications, evaluation of subjective dry mouth symptoms and visual inspection of oral mucosa. During the following appointments, the evaluation of subjective dry mouth symptoms (VAS; scale 0-10) was made using ten questions (Table I) and the changes in general health and medication were recorded. The statistical evaluations of the variables recorded at each appointment in VAS-scored symptoms of dry mouth were carried out using parametric repeated measurements analysis of variance (RANOVA; SPSS 10.0; level of statistical significance $p < 0.05$).

For all four seasons, the normal, low and very low UWS groups reported the severity of most VAS-scored symptoms to be of similar magnitude. The VAS scores were constantly highest for lip dryness, dryness during daytime, need to drink during daytime and swallowing difficulties. All control subjects reported VAS values lower than 2 on the 10 questions in the interviews. The xerostomic group (VAS range of means 1.2 - 6.0) and control group (VAS range of means 0.1-1.8) differed significantly from each other with regard to all ten VAS-scored symptoms ($p \leq 0.015$).

No statistically significant seasonal variation for any of the dry mouth symptoms was detected in the normal and low UWS groups. However, there was statistically significant, season-related variation in 4 symptoms in the very low UWS group (Table I). For the need to drink during daytime the VAS values were higher in winter than in summer and fall ($p < 0.05$). The VAS scores were found to be higher for swallowing difficulties in summer than in spring ($p < 0.02$), and for talking difficulties, higher in summer than in fall ($p < 0.04$). Oral pain was evaluated higher in summer than in spring and fall ($p < 0.05$), though rather low VAS scores were detected for oral pain throughout the study. Thus, the lowest VAS scores were registered either in spring and/or in fall. The differences were not related to the use of medication affecting the salivary flow.

Table I. The VAS-scored symptoms of xerostomia in very low UWS group (mean ± SD, n = 8). P-values for seasonal variation are also listed (RANOVA). For statistical significances in individual symptoms between the seasons, see text.

Symptom	Winter	Spring	Summer	Fall	RANOVA for seasonal variation
Dry mouth during daytime	5.1 ± 2.2	4.5 ± 3.1	4.8 ± 3.6	4.4 ± 2.3	n.s.
Need to drink during daytime	5.5 ± 2.5	5.0 ± 3.1	4.3 ± 3.5	3.7 ± 3.1	p = 0.000
Dry mouth during nighttime	2.6 ± 1.8	3.1 ± 2.9	3.2 ± 3.3	2.1 ± 2.8	n.s.
Need to drink during nighttime	2.0 ± 3.0	3.1 ± 3.1	2.7 ± 3.4	2.1 ± 2.7	n.s.
Oral pain and discomfort	1.3 ± 1.7	1.4 ± 1.5	3.1 ± 2.4	1.7 ± 1.3	p = 0.001
Lip dryness	4.9 ± 3.4	5.7 ± 3.2	5.0 ± 4.1	4.9 ± 3.2	n.s.
Swallowing difficulties	4.7 ± 3.6	4.2 ± 3.3	5.2 ± 3.7	3.7 ± 3.2	p = 0.008
Talking difficulties	4.2 ± 2.9	3.3 ± 2.7	4.1 ± 3.3	3.0 ± 2.6	p = 0.03
Easting difficulties	5.7 ± 3.6	4.2 ± 3.3	5.3 ± 3.3	3.4 ± 3.1	n.s.
Taste of food	1.9 ± 2.2	1.5 ± 1.8	1.7 ± 2.0	1.6 ± 2.2	n.s.

I.I. RANTANEN, *DDS**
J.O. TENOVUO, *DDS, PhD*
K. PIENIHÄKKINEN, *DDS, PhD*
E.M. SÖDERLING, *MSc, PhD*
Institute of Dentistry, University of Turku, Turku, Finland.
Address correspondence to: Irma Rantanen, Institute of Dentistry, University of Turku, Lemminkäisenkatu 2, FIN-20520 Turku, Finland. E-mail: irma.rantanen@utu.fi

References

1. NEDERFORS T: Xerostomia and hyposalivation. *Adv Dent Res* 2000; 14: 48-56.
2. NEDERFORS T, ISAKSSON R, MÖRNSTAD H, DAHLÖF C: Prevalence of perceived symptoms of dry mouth in an adult Swedish population-relation to age, sex and pharmacotherapy. *Community Dent Oral Epidemiol* 1997; 25: 211-6.
3. HOCHBERG MC,TIELSCH J, MUNOZ B, BANDEEN-ROCHE K, WEST SK, SCHEIN OD: Prevalence of symptoms of dry mouth and their relationship to saliva production in community dwelling elderly: the SEE project. Salisbury Eye Evaluation. *J Rheumatol* 1998; 25: 486-91.
4. LUNDSTRÖM IM, LINDSTRÖM FD: Subjective and clinical oral symptoms in patients with primary Sjögren's syndrome. *Clin Exp Rheumatol* 1995; 13: 725-31.
5. NEDERFORS T, HOLMSTRÖM G, PAULSSON G, SAHLBERG D: The relation between xerostomia and hyposalivation in subjects with rheumatoid arthritis or fibromyalgia. *Swed Dent J* 2002; 26: 1-7.