Palmoplantar pustulosis and sternocostoclavicular arthro-osteitis

EVA EDLUND,¹ ULF JOHNSON,² LARS LIDGREN,⁴
HOLGER PETTERSSON,³ GUNNAR STURFELT,¹ BJÖRN SVENSSON,⁶
JAN THEANDER,⁷ AND HELENA WILLÉN⁵

From the ¹Department of Rheumatology, ²the Blood Bank, and Departments of ³Diagnostic Radiology, ⁴Orthopedic Surgery, and ⁵Pathology, University Hospital, Lund; the ⁶Department of Rheumatology, General Hospital, Helsingborg; and the ⁷Department of Rheumatology, Kristianstad General Hospital, Sweden

SUMMARY Seventeen patients with shoulder pain and radiographic involvement of the sternoclavicular or sternocostal joints, or both, are described. Eleven of these patients also had palmoplantar pustulosis. Histological examination of the joints showed chronic and subacute inflammation, increased osteoblastic activity, and cartilage degeneration. *Propionibacterium acnes* was cultured in tissue samples from seven of the 15 biopsied patients, a finding at variance with those of previous reports. The possibility that sternoclavicular arthro-osteitis is of infectious origin should be the subject of further investigation. Non-steroidal anti-inflammatory drugs (NSAIDs) may provide pain relief, possibly owing to their inhibitory action on osteoblasts. In cases of severely restricted movement or severe pain resection of the medial clavicle may be considered.

Key words: arthritis, osteoblasts, *Propionibacterium acnes*.

Pustulosis palmoplantaris (PPP) is a chronic skin disease with intraepidermal, sterile pustules of the palms and soles. There is no effective treatment. The prevalence of PPP in Sweden is about 0.05%, and most patients are women, 40–60 years of age.^{1,2} Sternocostoclavicular arthro-osteitis has been described as a painful condition of unknown aetiology but with characteristic radiographic findings, usually affecting the anterior chest wall and the sternoclavicular, manubriosternal, and sternocostal joints. Although sternocostoclavicular arthro-osteitis, with or without PPP, is not infrequent in Japan,^{3–7} it has been considered rare among Caucasians, and, with few exceptions,^{8–10} published reports deal with individual cases.^{11–18} Recently a larger series of patients with the disease was reported.¹⁹ The aim of this study was to investigate clinical features in an extended series of 17 patients and to draw attention to the possibility that the condition may not be as rare in Caucasian populations as previously thought.

Patients and methods

From November 1982 all patients with anterior chest wall pain seen at the departments of rheumatology and orthopaedics in Lund, Helsingborg and Kristianstad were investigated and included in the present study, provided that they presented with the following criteria³: swelling and tenderness of the costoclavicular or manubriosternal joints, or both, in conjunction with abnormal x ray or scintigraphy findings, or both.

Patients with previously diagnosed ankylosing spondylitis, Reiter's syndrome, or psoriatic arthropathy were excluded. Seventeen consecutive patients had been included by March 1986.

The following investigations were performed: (a) Conventional radiographic examination with a lateral view of the sternum, and conventional tomography in the posteroanterior view, including the upper sternum, the medial ends of the clavicles, the sternoclavicular joints, and proximal sternocostal synchondroses. (b) Bone scintigraphy with ^{99m}Tc methylene diphosphonate. In all the patients the chest, including the sternum, sternoclavicular joints,

and sternocostal synchondroses, was examined. (c) Open biopsy of affected joints and para-articular bone was carried out in 15 patients. From each biopsy specimen five tissue samples were taken for aerobic and anaerobic bacterial and fungal cultures.²⁰ (d) Laboratory tests (erythrocyte sedi-

mentation rate (ESR), haemoglobin, white blood cell counts, C reactive protein, antinuclear antibodies and rheumatoid factor (Waller-Rose), serum calcium and serum alkaline phosphatase) all by routine procedures.

HLA-A, B, and C antigens were typed with the

Table 1 Clinical findings and response to treatment in 17 patients with sternocostoclavicular osteitis

Patient No	Age/sex	Duration (years)		Distribution of CO	Treatment and pain relief at follow up		
		CO	PPP		NSAIDs	Antibiotics	Other
1	44/F	5	5	stcl jj	I ±, Na ±	Pc, D, Cl -	res
2	36/F	3	3	stcl j	I ±	None	res
3	35/F	6	0	stcl jj, si j	Su ±	Pc -	Sul +
4	46/M	1.5	3	stcl j	Na ±	D -	
5	39/F	10	10	stcl jj	Na ±	D ±	
6	56/M	7		stcl jj	Su ±	None	
7	57/F	1.5		stcl jj	Su ±	None	
8	15/M	0.5		stcl j	I +	Fc +	
9	41/F	3	3	stcl j, ms j, si j	Na -	Pc -	
10	44/M	14		stcl j, s, si j	None	None	
11	44/F	10	15	stcl jj, si j	Na -	None	res
12	51/M	8	6	stcl jj, ms j	Ib +	None	
13	48/M	4	6	stcl jj	Di ±	D -	
14	63/F	6	4	ms j	Su -	None	
15	47/F	10	10	stcl jj, ms j	Na -	None	
16	37/F	1	1	stcl jj, s	ASA ±	None	
17	52/F	0.25		stcl j	I +	None	

ASA=acetylsalicylic acid; Cl=clindamycin; CO=costoclavicular osteitis; D=doxycycline; Di=diflunisal; Fc=flucloxacillin; I=indomethacin; Ib=ibuprofen; j, jj=joint, joints; ms=manubriosternal; Na=naproxen; NSAIDs=non-steroidal anti-inflammatory drugs; Pc=penicillin V; PPP=palmoplantar pustulosis; res=resection of medial clavicle; s=sternum; si=sacroiliac; stcl=sternoclavicular; Su=sulindac; Sul=sulphasalazine; +=no pain; ±=some pain relief; -=no pain relief.

Table 2 Radiographic changes and scintigraphic uptake

Patient No	Radiographic change					Scintigraphic uptake				
	Sterno-clavicular		Sterno-costal		Manubrio-sternal	Sterno-clavicular		Sterno-costal		Manubrio-sternal
	R	L	R	L		R	L	R	L	
1	S	S	S	S	-	+	+	+	+	-
2	0	0	0	0	-	-	+	-	-	+
3	E, S	E, S	E, S	E, S	S	+	+	+	+	+
4	0	0	0	S	-	-	-	-	-	-
5	S	0	0	0	-	+	-	-	-	-
6	S	S	S	S	E, S	+	+	+	+	+
7	S	S	0	0	-	+	+	-	-	-
8	E, S	E, S	0	0	-	+	-	-	-	-
9	0	E, S	0	0	E, S	+	+	-	-	+
10	S	S	0	0	-	-	+	-	-	-
11	E, S	E, S	E, S	E, S	E, S	+	+	+	+	+
12	E, S	E, S	0	0	E, S	-	-	-	-	+
13	E, S	E, S	S	S	S	+	+	-	-	-
14	S	S	0	0	E, S	-	-	-	-	+
15	S	S	0	0	E, S	+	+	-	-	+
16	0	0	0	0	-	+	+	-	-	-
17	0	0	0	0	-	+	+	-	-	-

E=erosion; 0=no radiographic change; S=sclerosis; +=increased scintigraphic uptake; -=no increase in scintigraphic uptake.

Fig. 1 Patient No 1. Bilateral sclerosis at the sternoclavicular and sternocostal joints, and a pronounced reaction, especially between the clavicles and first ribs.

Fig. 2 Patient No 13. Combined sclerosis and erosions, most pronounced in the medial end of the left clavicle.

Fig. 3 Patient No 8. ^{99m}Tc phosphate bone scan of a male patient with arthro-osteitis showing increased bone uptake of the radionuclide in the right sternoclavicular joint, indicating periosteal new bone formation.

lymphocytotoxicity test described by Kissmeyer-Neilsen and Kjerbye.²¹ HLA-DR antigens were typed using the standard NIH technique.²²

Table 1 shows the anti-inflammatory and antibiotic drugs used and the surgical treatment given.

Results

CLINICAL FINDINGS

In all but three patients the onset of symptoms was insidious with local pain, tenderness, and swelling over the affected joint. The symptoms were similar but more intense in cases with acute onset. The range of shoulder movement was reduced during intense pain and the ability to lift heavy objects was temporarily limited, mainly owing to pain in the sternoclavicular joint.

Muscular tension was sometimes present in the trapezius and levator scapulae muscles.

Eleven patients had PPP. One patient (No 12) developed psoriatic changes in the finger nails during the study.

RADIOGRAPHIC FINDINGS

Radiographic changes were seen in the sternoclavicular joints in 13 patients, in the sternocostal synchondroses in six, and in the manubriosternal synchondrosis in eight patients (Table 2).

The most common finding was sclerosis involving the subchondral bone, combined with periosteal reaction (Fig. 1). Erosion was less common and only present in combination with sclerosis (Fig. 2).

Changes in the sternocostal and manubriosternal

synchondroses were combined with changes in the sternoclavicular joints in all but one patient and were bilateral in all cases but three.

SCINTIGRAPHIC FINDINGS

Table 2 shows that increased scintigraphic activity (Fig. 3) was almost always accompanied by a radiological change. In one patient (No 2), however, there was increased uptake at scintigraphy, but the radiological findings were normal.

HISTOLOGICAL FINDINGS (Table 3)

The most common findings were chronic inflammatory changes in the bone marrow with a predominance of plasma cells and Russell's bodies (Fig. 4), bone marrow fibrosis, increased osteoblastic activity (Fig. 5), and cartilage degeneration (six cases). Subacute inflammation with a mixed cellular infiltration was present in four cases. Soft tissue involvement with chronic inflammation and a large number of plasma cells and some Russell's bodies was observed in two cases (Fig. 6). In three cases the bone tissue was normal. Three surgical specimens did not include bone, cartilage, or joint tissue and were considered to be non-representative.

BACTERIOLOGICAL FINDINGS (Table 3)

From each of 15 patients, in addition to fungal cultures, five aerobic and anaerobic cultures were performed with tissue biopsy specimens. In eight patients all the cultures were negative. In seven patients, however, growth of *Propionibacterium acnes* was obtained in at least two out of five cultures.

LABORATORY TESTS

Biochemical tests were essentially normal, with the

Table 3 Histological and bacteriological findings in 15 open biopsy specimens

	Number of patients	Patient Nos
Chronic inflammation	6	1,3,4,5,11,12
Subacute inflammation	4	2,6,13,15
Soft tissue involvement	2	8,17
No representative findings	3	9,10,14
Growth of		
<i>Propionibacterium acnes</i>	7	1,2,4,5,8,10,13
No growth of bacteria	8	3,6,9,11,12,14,15,17

Fig. 4 Patient No 8. Bone marrow with numerous plasma cells. (Haematoxylin and eosin.)

Fig. 5 Patient No 15. Increased osteoblastic activity. (Haematoxylin and eosin.)

Fig. 6 Patient No 8. Plasma cell infiltration of periosteal tissue. (Haematoxylin and eosin.)

exception of an increased sedimentation rate seen at the onset of disease in seven patients. Anaemia was not noted. Serum calcium and alkaline phosphatase were normal. The antinuclear antibody test was negative and the Waaler-Rose test was positive (1/64) in one patient (No 14).

Table 4 shows the distribution of HLA antigens. No significant deviations from normal were observed.

TREATMENT

Table 1 shows the treatment used. The follow up was of short duration in most cases (6–47 months). Pain relief was achieved in 12 of 17 patients (Table 1).

Resection of the medial clavicle was performed in three cases and gave total pain relief in one case (No 2). Recurrent swelling of the operated sternoclavicular region was seen in one case (No 1). The effect of antibiotic treatment started in five of the cases with positive cultures is difficult to evaluate, and so far (i.e., after four to six months of treatment) definite improvement has been noted in only one of these cases.

Table 4 Distribution of HLA types in 17 patients with sternocostoclavicular arthro-osteitis

Patient No	Sex	A	B	C	DR
1	F	3, 9	7, 40	w3	2, w6
2	F	1, 3	8, 27	w2	
3	F	1, 11	8, 35	w4	
4	M	1	8	w7	
5	F	2, w19	12, 27		
6	M	2, 11	15, 18	w3, w5	3, 4
7	F	2, 11	8, 35	w4	1, 3
8	M	2, 9	5	w3	
9	F	2, 3	7, 15	w3	4, w6
10	M	2, 3	7, 12		
11	F	2	5, 40	w3	
12	M	3, 9	7, 27	w2	2, w6
13	M	1	8	w7	3, 4
14	F	3, 28	7, 8	w7	3, w6
15	F	2, w19	21, 27		4
16	F	2, 11	5, 18	w3	
17	F	3, w19	12, 21	w5, w6	

Discussion

The present series represents sternocostoclavicular arthro-osteitis in Caucasians, obtained from southern Sweden with a population of one million people during 40 months, suggesting that the disease is more prevalent than previously believed. Two thirds of the patients also had palmoplantar pustulosis, a finding in accordance with those of earlier reports. The course of the disease was slowly progressive in 15 of the patients, with a duration of one year or more.^{3–7 11–18} Although some had acute clinical symptoms, the histology showed chronic or subacute inflammatory changes.

The relation between psoriatic arthritis and arthro-osteitis is not clearly defined. Obviously, psoriatic joint disease may occasionally be associated with a clinical picture similar to that of arthro-osteitis.²³ PPP, however, occurs in Caucasians with psoriasis sufficiently often to rule out chance.^{24–26} This is of particular interest as one of our patients, in addition to PPP, also developed psoriasis vulgaris of the nails (patient No 12). Prolonged prospective studies of a sufficiently large series of unselected patients are needed to decide the possible interrelationships between arthro-osteitis, PPP, and psoriasis vulgaris.

The radiographic changes are consistent with those reported by Junk *et al*,²⁷ with a good correlation between radiographic change and scintigraphic uptake. In practice there is no need to use both methods. Scintigraphy is more sensitive and exposes the patient to lower radiation than tomography.

Although the pathogenesis of the sternocostoclavicular arthro-osteitis remains unknown, various

possibilities have been proposed.^{4-6 10 28} One is that bacterial infection of low virulence triggers the bone reaction. Attempts to show the presence of bacteria in aerobic or anaerobic cultures of tissue specimens have seldom been successful. Kawakami *et al*, however, found growth of *Propionibacterium acnes* and *Propionibacterium granulosum* in the sternoclavicular lesions of two patients.²⁸ Le Goff *et al* have described one case, in which *Corynebacterium* was cultured from a sternocostal joint.^{14 16} Our finding of *Propionibacterium acnes* in cultures from seven of 15 patients would seem to suggest an infectious mechanism. The culture techniques with multiple tissue biopsy specimens should exclude the risk of false positive cultures. Palmoplantar pustulosis at a given site is more likely to be a reflection of a focal reaction elsewhere than an entry for an infectious agent,⁶ as is borne out by the reported failure to isolate bacteria in the pustules of PPP.²⁹

Our investigations of HLA types do not suggest that sternocostoclavicular arthro-osteitis is associated with a specific HLA haplotype, though the series was too small to detect minor deviations.

An increase in osteoblastic activity was noticed in six cases. Inhibition of this by NSAIDs may explain the successful pain relief as suggested by others.⁶ We found no evidence that the treatment used affected the course of disease, and this is in agreement with other authors.⁶ One patient treated with sulphasalazine regained full working capacity, relief from pain, and a normal ESR. Corticosteroid treatment was found to be ineffective in a study by Patterson and Bentley-Corbett,¹³ though Björkstén *et al* reported beneficial effects in three of their nine cases studied.¹²

Four of the seven patients providing positive bacterial cultures are currently being treated with antibiotics. So far (i.e., after four to six months of treatment) there has been no obvious effect (Table 1) except in one case (No 8) of complete recovery. It should be noted that, in contrast with the other cases, this patient was given antibiotic treatment early in the course of the disease. In an uncontrolled study Chigira *et al* found that administration of antibiotics to 11 patients brought some relief of pain that was resistant to NSAIDs.⁶

We believe that sternocostoclavicular arthro-osteitis and concomitant palmoplantar pustulosis are related conditions. Although the pathogenesis of the disease remains unclear, the finding of positive bacterial cultures in the biopsy specimens from almost one half of the group of 15 patients suggests the possibility of an infectious mechanism.

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