Case Report

Sacroiliitis in Sarcoidosis:
Case Reports and Review of the Literature

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Summary
We report the occurrence of bilateral sacroiliitis in 2 cases of biopsy-proven sarcoidosis. Tuberculosis was excluded by tuberculin testing and bronchoalveolar lavage in both cases. In the literature, 5 cases of sacroiliitis and sarcoidosis have thus far been described, but in all tuberculosis was not excluded (tuberculin testing was not performed or revealed a positive test result). In 1 of these cases tuberculosis was even simultaneously suspected to be present. In all previous cases, antituberculous or other antimicrobial agents were given. Previously reported cases of sacroiliitis in sarcoidosis are briefly reviewed, and possible relations between seronegative spondylarthropathies, slow bacterial infections, sarcoidosis and other granulomatous diseases are discussed.

Key words
Sarcoidosis, Sacroiliitis, Granulomatous Diseases

INTRODUCTION

The incidence of articular involvement in sarcoidosis is about 20 % (1-4).

Different forms of arthritis in sarcoidosis are well described (1-6). The most common form is an acute, self-limiting one, the so-called Löfgren's syndrome. It consists of an acute, mainly symmetrical arthritis, bithal pulmonary lymphadenomas and erythema nodosum (5). Sacroiliitis as a manifestation of sarcoid arthritis has only occasionally been described (7-12) and up to now there have only been 2 descriptions of bilateral sacroiliitis in sarcoidosis (8,12), one of which finally proved to be tuberculous (12).

Within a period of one year (1991) in the outpatient clinic of our rheumatology unit, sarcoidosis was diagnosed in 18 patients with oligoarthritis. Two female patients had a bilateral, subacute sacroiliitis as established clinically and radiologically.

CASE REPORTS

Case 1

A 28-year-old woman presented with severe dyspnoea, bilateral arthritis of the ankles and erythema nodosum. She also reported severe low back pain starting with the occurrence of her other symptoms, 4 weeks before her presentation. Her medical history was unremarkable. Clinically, there was tenderness and pain upon palpation of the sacroiliac joints. Mennel sign was positive bilaterally. The chest radiograph revealed a bilateral polycyclic lymphadenopathy (Fig. 1). Radiography of the sacroiliac joints showed the classical picture of bilateral sacroiliitis (Fig. 2), and nuclear magnetic resonance tomography showed signs of acute inflammation in both sacroiliac joints (Fig. 3).

Laboratory investigations showed an erythrocyte sedimentation rate of 20/40, C-reactive protein level of < 0.23 mg/dl, an elevated ACE of 23 U/l (normal: < 21) (13). Values on haematological parameters, creatinine and liver function tests were normal. HLA B27 was negative. A tuberculin skin reaction was negative. In bronchoalveolar lavage, no mycobacteria or other pathogenic microorganisms could be found. Analysis of lymphocyte subpopulations showed 72% CD4 and only 4% CD8 cells, with a ratio of 18 (normal: 1-2,5)(14). In broncho-
Peripheral transbronchial biopsies revealed noncaseating epitheloid cell granulomas. The diagnosis of sarcoidosis with Löfgren's syndrome and sacroiliitis was made. A treatment with steroids (prednisolone 1mg/kg) was begun and the patient's symptoms rapidly improved. The steroids could be tapered, and the patient is now treated with 5 mg prednisolone. ACE-level and erythrocyte sedimentation rate have normalized, the chest radiograph has returned to normal. Clinically, there are no more signs of sacroiliitis.

Case 2

A 39-year-old woman presented in November 1990 with erythema nodosum, bilateral arthritis of the ankles and dyspnoea. There were no other symptoms present. Her medical history showed no previous diseases. The chest radiograph revealed bilar polycyclic lymphadenopathy. Laboratory investigations revealed a normal ACE-level with 14 U/l, an elevated erythrocyte sedimentation rate of 10/31 mm. Values on haematological parameters, creatinine and liver function tests were normal. A tuberculin skin test was negative after 48 hours. Bronchoalveolar lavage showed typical lymphocyte counts with an elevated CD4/CD8 ratio of 9. Histologically, noncaseating epitheloid cell granulomas were shown. Cultures for mycobacteria and other microorganisms were negative. The diagnosis of acute sarcoidosis with Löfgren's syndrome was made and a steroid treatment with 1mg/kg prednisolone begun. Her symptoms rapidly improved within 3 weeks and the steroids were then tapered.

In September 1991, the patient presented again, now with severe low back pain and exacerbation of bilateral ankle arthritis. There was no erythema nodosum and no dyspnoea. Prednisolone had been stopped four weeks earlier. Clinically, she had pain and tenderness of both sacroiliac joints with a positive Mennel sign. Chest radiograph was normal, radiography of the sacroiliac joints (Fig.4) revealed bilateral sacroiliitis. Sacroiliitis was also proven by NMR-tomography.

Laboratory investigations revealed an elevated CRP with 1.6 mg/dl and ESR with 10/23 mm, but ACE level was normal with 9 U/l. The values on haematology, renal and