Pleural sarcoidosis: one case presenting with an eosinophilic effusion

D VITAL DURAND, A DELLINGER, C GUERIN, JC GUERIN, R LEVRAT

From the Centre Hospitalier Lyon Sud, France

Pleural manifestations are rare in sarcoidosis, occurring in about 1% of a cumulative series.¹ They include pleural effusions, pleural thickening, and spontaneous pneumothorax. We present a case of pleural sarcoidosis presenting as an eosinophilic effusion. Specific pleural lesions were established by thoracoscopy and biopsy, which showed appearances suggesting serous dissemination of sarcoid granulatum tissue.

Case report

A 20 year old man was first admitted to hospital on 8 February 1982, because of left scapular pain and a dry cough. He had noted weight loss of 4 kg and increasing malaise over the past month. He denied night sweats and fever. He had no previous medical history and a chest radiograph seven months earlier for routine purposes showed normal appearances.

On examination, the only abnormal findings were diminished breath sounds and dullness to percussion at the left base. A chest radiograph showed bilateral hilar and paratracheal lymph node enlargement, a diffuse interstitial pulmonary infiltrate, and a left pleural effusion (fig 1). The erythrocyte sedimentation rate (ESR) was 92 mm in one hour (Westergren); the haemoglobin (Hb) concentration was 13.2 g/dl; the white blood count (WBC) $13 \times 10^9/l$ (4% eosinophils); platelets $545 \times 10^9/l$; serum calcium concentration 2.26 mmol/l (9.04 mg/100 ml). Serum angiotensin converting enzyme activity was 34 nmol/ml/min (normal lower limit 40 nmol/ml/min). An intradermal tuberculin test (50 units IP 48, Institut Pasteur) gave a negative response. No tubercle bacilli were isolated from the many sputum samples examined. Serological studies for parasitic infection gave negative results.

Pulmonary function tests showed a forced vital capacity of 3.5 l (predicted 4.1 l), FEV₁ 71% (predicted 83%), and transfer coefficient (Kco) $5.1 \text{ min}^{-1} \text{ torr}^{-1}$ (predicted 5.9) ($1.7 \text{ mmol min}^{-1} \text{ kPa}^{-1} \text{ torr}^{-1}$ (predicted 2.0)). Pleural aspiration yielded 50 ml of clear yellow fluid with a protein content of 52 g/l. Microscopy showed the fluid to be profusely cellular; the white cell differential count was 2% neutrophils, 90% eosinophils (76% at subsequent thoracocentesis), 4% basophils, 3% lymphocytes, and 1% mesothelial cells; bacterial cultures were negative. Fibre-optic bronchoscopy and bronchial biopsy specimens were

Fig 1 Chest film on admission showing bilateral hilar nodes, diffuse interstitial infiltrate, and a left pleural effusion.

normal. Bronchoalveolar lavage fluid contained 56.75×10^6 cells/ml (normal $10 \pm 4 \times 10^6$ cells/ml), with 22% lymphocytes, 1% polymorphonuclear cells, and 77% macrophages; angiotensin converting enzyme bronchial activity was 0.22 nmol/ml/min (normal lower limit 0.30 nmol/ml/min). Mediastinoscopy showed greatly enlarged lymph nodes. Histological examination showed follicular epithelioid and giant cell granulomas. All cultures and special stains were negative. A provisional diagnosis of thoracic sarcoidosis was made and the patient was discharged without any treatment.

On 14 April 1982 a chest radiograph showed resolution of the left sided pleural effusion but the appearances were otherwise unchanged. On 3 May 1982 a chest radiograph showed an asymptomatic pleural effusion on the right. On examination no other abnormality was observed. The ESR was 55 mm in one hour; Hb 14.4 g/dl; WBC $9.6 \times 10^9/l$ (15% eosinophils). The results of further investigations for tuberculosis and parasitic infection were again negative. Serum angiotensin converting enzyme activity was 31 nmol/ml/min. Thoracoscopy showed that the pleural surface was very inflamed and there were multiple adhesions. Small whitish granulations were observed on the parietal pleura (fig 2). The pleural fluid was yellow with a protein concentration of 56 g/l; the white cell differential count

Address for reprint requests: Dr D Vital Durand, Centre Hospitalier Lyon Sud (Service Pr R Levrat), 69310 Pierre Benite, France.

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Fig 2 Thoracoscopic aspect showing multiple whitish granulations on the parietal serous membrane.

showed 1% neutrophils, 40% eosinophils, 3% basophils, 44% lymphocytes, and 2% mesothelial cells. Microscopic examination of pleural biopsy material showed multiple inflammatory granulomas with a few epithelioid and giant cell follicles. All cultures were negative. No treatment was prescribed and one month later a chest radiograph showed the resolution of the pleural effusion. In May 1983 the patient was doing well; all radiographic abnormalities had resolved.

Discussion

Pleural manifestations in the course of sarcoidosis have been recognised as an infrequent² and perhaps underdiagnosed³ manifestation of sarcoidosis. Three radiographic presentations are described: spontaneous pneumothorax,⁴ pleural thickening, and pleural effusions.^{1,2,5,6} The frequency of occurrence of these last two manifestations in sarcoidosis has been reported to be from 1%¹ to 10%.² Sarcoid pleural effusion may be occasionally induced by mediastinal venous compression⁷; more often specific pleural lesions may be established by the presence of non-caseating granulomas. Thoracoscopy might increase substantially the proved incidence of pleural lesions in sarcoidosis.

We found only 32 previous reports of histologically proved pleural sarcoidosis with effusion. Analysis of the principal data where they are recorded showed that the

average age was 35 years. There was no preference for side and in almost a third of reports effusions were bilateral. Pleural fluid was considered to be more often an exudate than a transudate. Cytological examination of pleural fluid showed scanty cells in half the cases; in the others lymphocytes were predominant (61–100%). Our patient was exceptional, having 80% eosinophils in the fluid from the left side and 40% in that from the right side on the first tap. Three other cases are reported with pleural eosinophilia: Berte⁸ found 29% eosinophils at the sixth thoracentesis; Beekman⁵ found 15% eosinophils in the first pleural aspirate. In a further case⁶ the percentage was not specified. Although eosinophilia in the blood is well recognised,⁹ the present case appears to be the first reporting the presence of eosinophilia in the pleural space and in the blood, although it was not present in the bronchoalveolar lavage fluid. Histological confirmation of sarcoid pleural granulomas was most often obtained by transparietal needle biopsy (18 patients) and an open biopsy was carried out in nine patients. Pleural effusion occurred at the time the patient first presented with sarcoidosis in five cases; in three of them, this was the sole radiological abnormality. In eight others pleural effusion developed during the first year of the disease. In five patients the interval between the initial diagnosis of sarcoidosis and the appearance of effusion varied from three to 17 years. Since most of the patients were given corticosteroids, the natural history of pleural effusion in sarcoidosis remains uncertain; complete resolution was usually reported, but progression to chronic pleural thickening also occurred.

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