Pleural effusion in ankylosing spondylitis: successful treatment with intra-pleural steroid administration


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Introduction

Apical pleural thickening with fibrobullous disease is common in pleuropulmonary manifestation of ankylosing spondylitis (AS). Other forms of pleural disease are rare (1) and there are few reports of treatment of pleural effusions associated with AS (2-4). We report a case of persistent exudative pleural effusion in AS and the therapeutic effects of intra-pleural administration of prednisolone.

Case Report

A 56-year-old farmer with a 2-yr history of documented AS was admitted in May 1993 because of a 6-month history of persistent pleural effusion and increase of dyspnoea. He had a history of mediastinal lymphoma 9 yr previously and a skin squamous cell carcinoma of the upper lip 4 months previously. Both diseases were in complete remission after irradiation therapy. His joint disease had required oral indomethacin but 1 month before his admission, he suffered from multiple gastric ulcers.

On admission, he was 145 cm tall and weighed 43 kg. Spinal movements were severely restricted in all directions and the thoracic spine was kyphotic. No synovitis was apparent in peripheral joints. A chest radiograph showed a little left pleural effusion and right apical fibrobullous lesions (Plate 1). CT scan revealed dilated bronchi and a cavity with thickened walls in the right upper lobe, but aspergilloma was not detected within the cavity. Radiographs of his spine confirmed severe spondylitis and obliteration of the sacroiliac joints. Laboratory investigation revealed: haemoglobin 9.8 g dl\(^{-1}\), white blood cell count 4800 mm\(^{-3}\), with 70% neutrophils, 14% lymphocytes, 13% monocytes, and 3% eosinophils. Erythrocyte sedimentation rate was 66 mm h\(^{-1}\), antinuclear antibody and rheumatoid factor were negative, and serum precipitins against *Aspergillus fumigatus* was positive. The quantification of serum immunoglobulins were as follows: IgG, 2371 mg dl\(^{-1}\) (range 1048-1646); IgA, 300 mg dl\(^{-1}\) (range: 146-340); IgM, 139 mg dl\(^{-1}\) (range: 70-170). Arterial blood gases [\(\text{PO}_2\) concentration of inhaled air (\(\text{FIO}_2=0.21\) indicated \(\text{PO}_2\) of 68.7 Torr, \(\text{PCO}_2\) of 45.6 Torr, pH of 7.45, and \(\text{HCO}_3\) of 31 mmol l\(^{-1}\)]. Sputum culture for tuberculosis and aspergillosis remained negative. A tuberculin skin test was negative. The HLA type was A3, A11, B27, B44, CW5, and CW6. Pleural fluid was straw-coloured and had total protein of 4.8 g dl\(^{-1}\), pI 7.6, glucose 93 mg dl\(^{-1}\), lactate dehydrogenase 147 IU l\(^{-1}\), and
Plate 2 Pleural biopsy specimen reveals fibrous thickening with an accumulation of lymphocytes and inflammatory cells.

adenosine deaminase 12·8U1 l−1; tubercle bacilli were not seen in the fluid nor grown on culture. Cytology was negative for malignant cells. The lymphocytes in the effusion were analysed for surface marker using monoclonal antibody UCHL-1 (pan T) or L26 (pan B), however there was no monoclonality.

Despite several pleural punctures over the next 4 months, the effusion reaccumulated rapidly and the characteristics did not change. Percutaneous pleural biopsy showed fibrous thickening with an accumulation of lymphocytes (Plate 2).

Oral prednisolene therapy was contraindicated because of the patient's gastric ulcers and the possibility of A. fumigatus infection of the right upper lobe. In October 1993, 20 mg of prednisolone was instilled into the left pleural cavity after complete drainage of the effusion. Subsequently, the effusion reaccumulated a little but did not increase over the following 13 months, and his forced vital capacity improved from 630 ml to 910 ml.

Discussion

Pleural effusion in AS is rare. Rosenow et al. (1) reported three out of 2080 patients (0·15%), and Spencer et al. (5) found none in 200 patients (0%). In contrast, a much higher prevalence was found by Nagyhegyi et al. (6), with four out of 86 patients (5%). It is not clear whether these differences represent true variations in different populations, in thresholds of reporting, or in patient selection. A case of haemothorax due to spinal fracture in AS has also been reported (7). In our case, the fluid was an exudate that had a normal glucose content and pH. These results are similar to reported cases in AS, in contrast with pleural effusions of rheumatoid arthritis; glucose and pH are low. In the five previously reported cases (1,2,4), three had bilateral effusion and the other two were on the left side; two cases were recurrent.

The pathogenesis of subpleural inflammation in pleuritis with AS remains unknown. Transbronchial biopsy specimens of the lung in AS showed interstitial fibrosis of a variable degree in five cases out of 12 (41·5%) (8). The fibrosing inflammatory process of pleura might have been caused by contiguous extension of the vertebral inflammatory fibrosis into the pulmonary parenchyma and subpleural space.

There are no reports of a temporal relationship between activity in the spinal column and pleural disease, or of the response to the treatment of pleural effusion in AS. The effusion was applied for some weeks and no treatment was required in three patients (1), although systemic administration of 30 mg day−1 of prednisolone (2), or indomethacin (3), or 200 mg day−1 of phenylbutazone (4) in another three patients did clear the effusions. As our case had been treated with various non-steroidal anti-inflammatory drugs for years, these drugs were not instilled into the pleural effusion. After intrapleural administration of steroid, the effusion built up slightly and did not increase over the following 9 months. Kinnear et al. (2) reported a case with bilateral pleural effusion complicating AS, and the effusion resolved completely after systemic administration of 30 mg of prednisolone. This case has some similarities to our case, but we are not able to explain the mechanism of the therapeutic effect of prednisolone. On the other hand, pericarditis is a rare association of AS which responds well to steroids (9), and uveitis associated with AS also responds to local steroid therapy. These were the reasons for choosing the successful steroid therapy.

References


