



CASE REPORT

Isolated pleural and pericardial effusion in a patient with ankylosing spondylitis

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KEYWORDS

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Summary Ankylosing spondylitis (AS) is a chronic seronegative spondyloarthritis with the major histocompatibility antigen HLA B27. Pulmonary involvement in AS is rare and is usually in the form of upper lobe fibrocavitary disease. Herein, we present a case with recurrent pleural and pericardial effusion without apical fibrobullous disease who responded to prednisolone treatment well. It is believed that this is the first case report complicating AS without parenchymal involvement in the literature.

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Introduction

Upper lobe fibrocavitary disease is most common pulmonary manifestation of ankylosing spondylitis (AS).¹ Other forms of pleuropulmonary manifestation is rare. We present a case of AS with pleural and pericardial effusion who responded well to systemic prednisolone treatment.

Case report

A 57-year-old teacher presented with dyspnea, pleuritic chest-pain on his left hemithorax and fever which had began 20 days ago and had

gradually improved. He was an ex-smoker of 15 p/y who stopped smoking 15 years ago. He denied any hemoptysis, cough and weight loss. His past-medical history revealed that he was diagnosed as AS 6 months ago when he had a back-pain and also peripheral arthritis. At that time, an ophthalmologist revealed an uveitis. He has been on sulfasalazine and local steroid eye-drop treatment since then. On admission, he was anxious and mildly dyspneic with a pulse rate of 95/min, respiratory rate of 24/min and a blood pressure of 130/80 mmHg. His breath sounds were diminished in the left lower-zone and flatness was remarkable on percussion on that side. Spinal movements were mildly restricted in all directions. Rest of his examination was normal. During his stay in hospital, his temperature increased to 38–39 °C several times a day. His chest X-ray was compatible with a pleural effusion on the left side showing an

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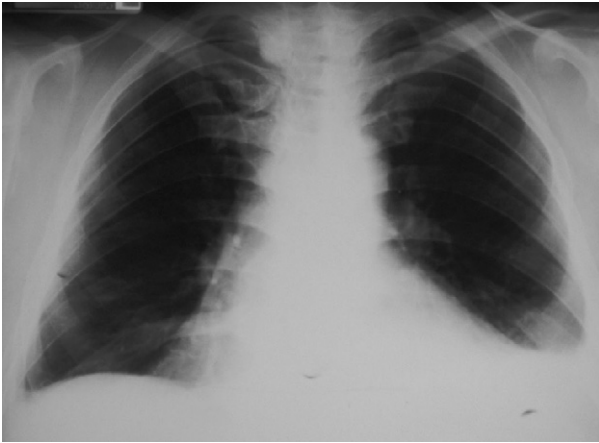


Figure 1 Chest X-ray of the patient compatible with left pleural effusion.

homogenous opacity obscuring left costophrenic angle (Fig. 1). Obliteration of his sacroiliac joints as well as calcification of the anterior longitudinal ligament was remarkable on his pelvic and spine X-rays (Fig. 2). Laboratory investigations showed: haemoglobin 12 g/dl, white blood cell-count 12,000/mm³, with 75% neutrophils, 19% lymphocytes, 7% monocytes and 3% eosinophils. Erythrocytes sedimentation rate was 86 mm/h. Electrolyte levels, urine analysis renal and liver function tests were all within normal limits. No bacterial growth observed on urine and blood cultures. Antinuclear antibody was positive in low titers, and rheumatoid factor and anti-double-strand DNA were negative. Pleural fluid aspiration revealed an exudative effusion with a LDH of 500 IU/dl, glucose level of 50 mg/dl and total protein of 4 g/dl. Tubercle bacilli were not seen in the fluid. Then, a closed pleural biopsy was performed which showed chronic pleuritis with a predominantly lymphocytic inflammation. Meanwhile, sefuoksime IV 750 mg tid and clarithromycin 500 mg BID were begun with a probable diagnosis of parapneumonic effusion. His symptoms improved gradually, sedimentation rate dropped to 60 mm/Hg, fever disappeared and his chest X-ray was cleared. He is discharged from the hospital 1 week later with a recommendation of clarithromycin 500 mg BID one more week.

Twenty days later from his discharge, he returned with the same complaints as left-side chest pain, fever and malaise. This time, pericardial rub was also remarkable on cardiac examination which echocardiography revealed 10 mm-thickness pericardial effusion, later. His pleural effusion had also recurred on his chest X-ray. However, no parenchymal involvement was remarkable on thorax CT (Fig. 3a and b). No tuberculous bacilli growth was observed on culture since the previous investiga-

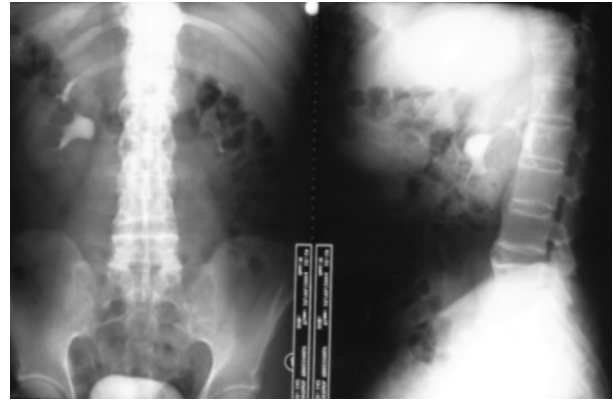


Figure 2 Pelvic and spine X-ray of the patient showing obliteration of the sacroiliac joint and calcification of the anterior longitudinal ligament.

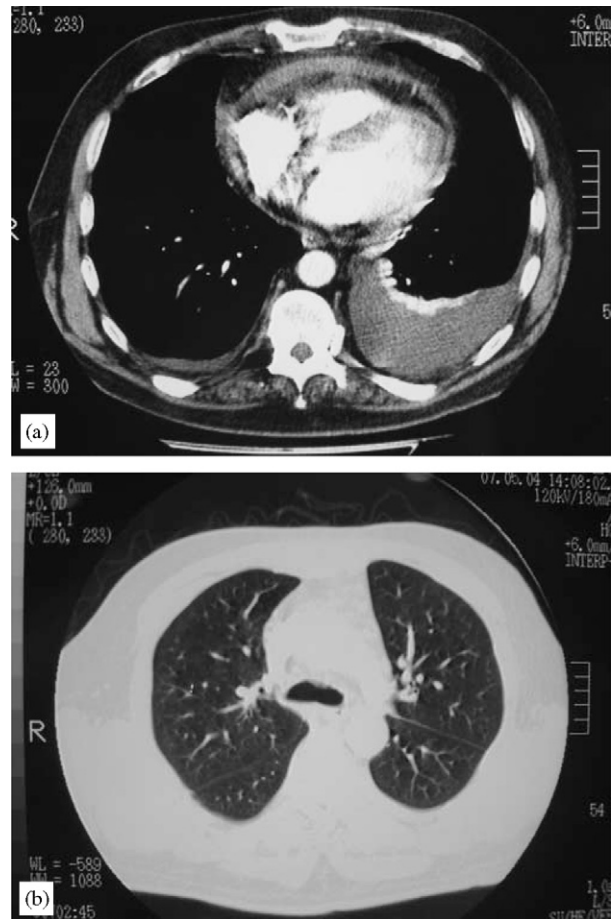


Figure 3 Thorax CT of the patient showing pleural and pericardial effusion (a) without any parenchymal involvement (b).

tion. IgG, IgA, IgE and IgM and complement 3 and 4 levels were normal. He was positive for HLA-B27 antigen. Neither malignant cell infiltration nor granulomatous infiltration of the pleura was

observed on the second pleural biopsy and cytology. This time, prednisolone 60 mg daily was begun with a diagnosis as spondylitic pleural and pericardial effusion. He responded quickly to treatment and all his symptoms completely resolved within a few days. Pleural and pericardial effusion completely resolved on chest X-ray and echocardiography 1 week later. Steroid-dose gradually tapered to 10 mg. Two years after he presented, the patient remains well off all medications.

Discussion

AS is chronic inflammatory disease of the axial skeleton manifested by back pain and progressive stiffness of the spine. AS is one of the spondyloarthropathies which show inflammation around the entheses (the site of ligament insertion into bone). It characteristically affects young adults with a peak age of onset between 20 and 30 years. Although classically thought of as a spinal disease, transient acute arthritis of peripheral joints occurs in up to 50 percent of patients. In addition, systemic involvement of other organs such as eyes, lungs and heart can be affected.

Pulmonary involvement in AS is a well-known feature of the disease. Apical pleural thickening with underlying apical fibrobullous disease is the common manifestation.¹ The incidence of pleuropulmonary involvement shows a big variation in the literature, probably as a result of selection biases. While Spencer et al.² reported the pleuropulmonary incidence as 0%, Lauritzen et al.³ found it to be 30%. Reviewing the radiographs of 2080 AS patients, Rosenow et al.¹ found 26 cases of apical fibrobullous disease. In five of the patients, aspergillosis had been complicated fibrobullous disease some time during the disease course.

Although apicobullous disease can frequently be seen in AS patients, pleural effusion is a very rare manifestation of the disease. Rosenow et al.¹ reported three cases after reviewing the chest X-rays and reports of 2080 AS cases (0.15%). Two of the Rosenow's cases had apicobullous disease as well as pleural effusion. One of the cases of Rosenow et al.¹ and another case from Kinnear and Shneerson⁴ had only pleural effusion without apical involvement as in our case. Pleural effusion can be left-side^{1,5} or bilateral.^{1,4,6}

Although pericardial effusion has been rarely reported in AS patients,⁷ to our knowledge, there is no previous report showing pleural and pericardial effusion association in AS as manifested in our case. Pleural fluid analysis of patients with effusion had no discriminative feature, and is usually exudative and non-hemorrhagic with normal glucose and pH levels.

Pleural effusion can clear spontaneously in some AS patients.¹ However, there is usually a need of systemic or local antiinflammatory treatment as recurrence may be prominent feature of the disease as in our case. Systemic prednisolone (30 mg daily),⁴ local administration of steroids (20 mg prednisolone locally to pleural cavity after complete drainage of the effusion)⁵ or phenylbutazone (200 mg daily)⁶ had been successfully used in the treatment of pleural effusion. We used 60 mg prednisolone daily successfully as our patient had prominent systemic symptoms.

In summary, we present a case of AS patient who had polyserositis, namely pleural and pericardial effusion, and responded well to prednisolone. This association had not been reported in the literature and clinician should be aware of this presentation of AS without apicobullous disease.

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