

CASE HISTORY

Acute bilateral pleural effusions in ankylosing spondylitis

Aravinthan Narani¹, Selladurai Pirasath²¹Consultant Rheumatologist, Teaching Hospital, Jaffna, Sri Lanka.²Registrar in Medicine, Professorial Medical Unit, Teaching Hospital, Jaffna, Sri Lanka**Article information**Total number of
Words 918*Key words:* Ankylosing spondylitis, Pleural
effusion*Date of submission:* 12.12.2018*Date of acceptance:* 09.06.2019**Author responsible for correspondence:**Dr. Selladurai Pirasath,
Registrar, Professorial Medical Unit,
Teaching Hospital, Jaffna, Sri Lanka.
Email: selladuraipirasath81@gmail.com<https://orcid.org/0000-0002-4274-4919>DOI: <http://doi.org/10.4038/cjms.v56i1.4950>**Abstract**

Ankylosing spondylitis (AS) is a chronic seronegative spondyloarthritis associated with the major histocompatibility antigen HLA B27. Apical pleural thickening in ankylosing spondylitis is common in association with apical fibrobullous disease. Pleural disease is otherwise rare. Here, we present a case of bilateral pleural effusion in 38 years old man with ankylosing spondylitis.

Introduction

Ankylosing spondylitis is a relatively common chronic inflammatory disease that affects the spine, sacroiliac joints, peripheral joints and affects multiple other tissues, which can progress to bony fusion of the spine. Pleural and pulmonary manifestations are rare, though upper lobe fibro cavitory disease is recognized in Ankylosing spondylitis. We present a 38-year old man with bilateral pleural effusion with accompanying fever, who responded to steroid therapy.

Case report

A 38 -year-old man presented with dyspnea, left sided pleuritic chest-pain and fever. He had no history of hemoptysis, cough and weight loss. He had been on sulfasalazine for 8 months before his presentation and specific physiotherapy for his

back pain and shoulder pain. On examination, he was febrile, tachypnoeic, with a pulse rate of 90 /min, and a blood pressure of 140/80 mmHg. He had features suggestive of bilateral pleural effusions. Spinal movements and movements of both shoulders were mildly restricted in all directions. He had no evidence of active arthritis. Rest of his examination was normal.

His chest X-ray showed a homogenous opacity obscuring lingual lobe and lower lobe of left lung and lower lobe of right lung compatible with a pleural effusion (Figure 1A). Obliteration of his sacroiliac joints as well as calcification of the anterior longitudinal ligament, squaring of vertebrae and fusion of syndesmophytes were remarkable on his pelvic and spine X-rays (Figure 2).

His complete blood count showed normal cell count with differential count (Haemoglobin- 12.8g/dL, White cell count- 5800/mm³ with 75% neutrophils



This is an open-access article distributed under the terms of the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original author and source are credited.

and 15% lymphocytes). His inflammatory markers were high (Erythrocyte Sedimentation Rate-108 mm/h, C-Reactive protein 211 mg/dL). His serum electrolyte, urine analysis, renal and liver function tests were all within normal limits. No bacterial growth was observed on urine and blood cultures. Both retroviral and syphilitic screenings were negative. Sputum for Acid fast bacilli was negative on 3 occasions and his mantoux was negative. Antinuclear antibody and rheumatoid factor were negative.

Pleural fluid aspiration revealed an exudative effusion with a lactate dehydrogenase of 2208 IU/dL, glucose level of 50 mg/dL and total protein of 4 g/dL. Culture, including for tubercle bacilli, was negative. Pleural fluid for tuberculosis polymerase chain reaction was negative. Pleural fluid cytology showed lymphocytic rich effusion with reactive mesothelial cells and no malignant cells. Serum angiotensin converting enzyme level was normal. Mycoplasma IgM was negative, IgG was positive, however there was no four-fold increase between the initial and convalescent samples. However pleural fluid adenosine deaminase level was slightly elevated at 52IU/L (45-160 U/L with a mean level of 100U/L suggestive of tuberculosis). Contrast enhanced computed tomography of chest showed bilateral pleural effusion due to ankylosing spondylitis or infection.

Figure 1 A. Chest X-ray showing a homogenous opacity obscuring lingual lobe and lower lobe of left lung and lower lobe of right lung compatible with a pleural effusion. **Figure 1 B.** Residual pleural effusion following complete treatment

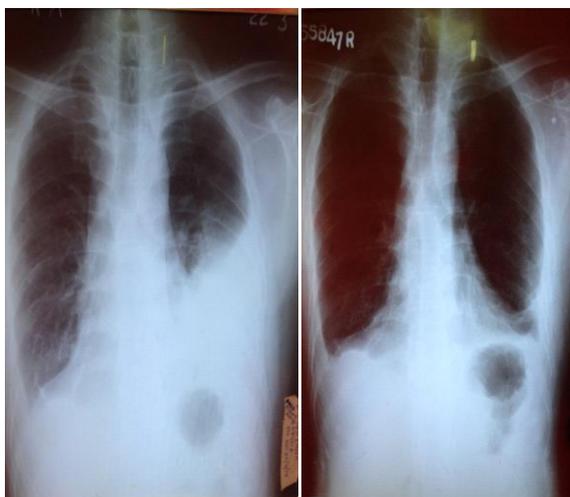


Figure 1 A.

Figure 1 B.

There was no evidence of active tuberculosis (Figure 3). A pleural biopsy was not done. Echocardiogram was normal with no evidence of a pericardial effusion.

Figure 2 A and B. X-ray of lumbosacral spine Anterior-Posterior and lateral view showed obliteration of his sacroiliac joints as well as calcification of the anterior longitudinal ligament, squaring of vertebrae and fusion of syndesmophytes



Figure 2 A.

Figure 2 B.

Figure 3. The CECT showed bilateral pleural effusion due to Ankylosing spondylitis



Meanwhile, an intercostal tube was inserted on his left side to drain pleural fluid and he was started with Co-amoxiclav and Meropenam and with antituberculosis treatment as tuberculosis is endemic in Sri Lanka. Despite therapy, his inflammatory markers continued to be markedly elevated and he continued to have bilateral pleural effusions. A course of prednisolone 50 mg daily was begun with a diagnosis as spondylitic pleural effusion after excluding possible aetiology. He responded quickly to treatment and all his

symptoms completely resolved within a few days. Bilateral pleural effusions completely resolved on chest X-ray after few days of starting steroids (Figure 1 B). Steroid-dose was gradually tapered to 10 mg/daily and was continued for 9 months.

Discussion

Ankylosing spondylitis (AS) is a chronic seronegative spondyloarthritis associated with HLA B27[1]. Pulmonary manifestations are rare causing upper lobe fibro cavitory disease[1]. These have a significant variation in the literature[2]. Dudley-Hart *et al*[3] reported two patients with history of pleurisy and one of them had a pleural effusion in 1950. Zorab has reported one tuberculous and one non-tuberculous pleural effusion among 53 cases of ankylosing spondylitis in 1962[4]. Crompton *et al* noted bilateral pleural calcification in out of 255 patients with ankylosing spondylitis unrelated to tuberculosis or asbestos exposure in 1974[5]. Kinnear and Shneerson had reported only pleural effusion without apical involvement[6]. These effusions were bilateral[1] or unilateral[2,6] and recurrent[6,7]. There were pleural effusions coexistent with pericardial effusion in some cases[8]. Spencer *et al* described bilateral pleural thickening in one out of 200 patients with ankylosing spondylitis[9]. The temporal relationship between activity in the spinal and pleural disease or of the response to treatment were not observed. Pleural fluid analysis had no specific features and is usually exudative with normal cell count and glucose[4]. In our case, the fluid was an exudate with a normal glucose content and pH. These results are similar to reported cases in AS. Pleural effusion resolves spontaneously in some cases. However, systemic or local steroids or phenylbutazone had been effective in the pleural effusion[10]. Our patient resolved completely after systemic administration of steroids. Pleural effusion occurs in autoimmune connective tissue diseases as a part of the inflammatory component. Here, it could also be due to serositis related to ankylosing spondylitis[11].

In summary, we present a case of AS who had pleural effusion and responded well to prednisolone. The clinician should be aware of this presentation of AS without apicobullous disease.

Conflict of interest

The authors declare that there is no conflict of interest regarding the publication of this paper.

References

1. Rosenow EC, Strimlan CV, Muhm JR, Ferguson RH. Pleuropulmonary manifestations of ankylosing spondylitis. *Mayo Clinics* 1977; 52: 641-649.
2. Ghimire A, Ramos F, Knauert M, Puchalski J T, Acute Pleuropericardial Effusion In a Patient With Ankylosing Spondylitis. *American Journal of Respiratory and Critical Care Medicine* 2010;181:A6668. Dudley-Hart F, Bogdanovitch A, Nichol WD. The thorax in ankylosing spondylitis. *Ann Rheum D* is 1950;9:116-31. doi.org/10.1136/ard.9.2.116.
3. Dudley-Hart F, Bogdanovitch A, Nichol WD. The thorax in ankylosing spondylitis. *Ann Rheum Dis* 1950;9: 116-31. doi.org/10.1136/ard.9.2.116.
4. Zorab PA. The lungs in ankylosing spondylitis. *Q J Med* 1962; 31:267-80.
5. Crompton GK, Cameron SJ, Langlands AO. Pulmonary fibrosis, pulmonary tuberculosis and ankylosing spondylitis. *Br J Dis Chest* 1974;68: 51-6.
6. W.J.M. Kinnear, J.M. Shneerson Acute pleural effusion in inactive ankylosing-spondylitis *Thorax* 1985;40:150-1. doi.org/10.1136/thx.40.2.150.
7. Blanch J, Guitierrez-Cobellada J, Lopez-Colomes J L, Faus S, Martinez-Pordo S. Pleural effusion in ankylosing-spondylitis: a new pulmonary disease or a casual association? *Revision Espan Rheumatology* 1989;16.: 91-2.
8. Erkan L, Uzun O, Findik S, Atici A G, Ozkaya S. Isolated pleural and pericardial effusion in a patient with ankylosing spondylitis. *Respiratory Medicine* 2007;101(2):356-8. doi.org/10.1016/j.rmed.2006.04.028.
9. Spencer DG, Park WM, Dick HM, Papazoglou SN, Buchanan WW. Radiological manifestations in 200 patients with ankylosing spondylitis: correlation with clinical features and HLA B27. *J Rheumatol* 1979; 6: 305-15.
10. Tanaka H, Itoh E, Shibusa T, Chiba H, Hirasawa M, Pleural effusion in ankylosing spondylitis: successful treatment with intra-pleural steroid administration. *Respiratory Medicine* 1995; 89: 509-11.
11. Bouros D, Pneumatikos I, Tzouvelekis A. Pleural involvement in systemic autoimmune disorders.2008; 75(4):361-7