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CASE REPORT

A Rare Cause of Pleural Effusion: Ankylosing Spondylitis

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ABSTRACT

Ankylosing spondylitis (AS) is a seronegative spondyloarthropathy involving the axial skeleton and major joints. Pleuropulmonary involvement is rare manifestation of AS mostly presented as apical fibrobullous disease and pleural thickening. Herein, we report a rare case with right sided pleural effusion in a male patient that was determined on a chest x-ray performed routinely before initiation of anti-TNF therapy. Pleural effusion was a lymphocytic exudate with a normal glucose level. The histopathological examination of closed pleural biopsy revealed nonspecific chronic inflammatory cell infiltration. The patient was followed up with etanercept treatment. One year after the initiation of therapy, due to persistent pleural effusion, video assisted thoracoscopic surgery was performed. Pleural biopsies were also consistent with chronic inflammation.

Key words: Ankylosing spondylitis, connective tissue disease, pulmonary involvement, pleural effusion

INTRODUCTION

Ankylosing spondylitis (AS) is a seronegative spondyloarthropathy involving axial skeleton and major joints. The disease may be accompanied by extraarticular findings such as acute anterior uveitis, aortic insufficiency, amyloidosis, osteoporosis, pleuropulmonary disease and cauda equine syndrome [1]. Pleuropulmonary involvement is rare and mostly manifested as apical fibrobullous disease and pleural thickening. Pleural effusion is extremely rare [2]. Herein we aimed to present a patient with AS and associated pleural effusion.

CASE PRESENTATION

A 36-year-old male patient was referred to our clinic for the evaluation of right sided pleural effusion etiology which was determined on a chest x-ray performed routinely before initiation of anti-TNF therapy (Figure 1a).

In his past medical history, he had a diagnosis of AS for the last 2 years and received indomethacin and sulfasalazine. He had a smoking history of 21 pack-years and environmental asbestos exposure. He declared that he neglected his symptoms mild cough and dyspnea on exertion which were present for the last 3 months.

On physical examination, there was kyphosis, absence of respiratory sounds on the right lung base and dullness on percussion. Computed tomography of thorax revealed pleural effusion on the

right hemithorax and vertebral lesions related to AS. Complete blood count and biochemical analysis were within normal limits. C-reactive protein was mildly elevated 3.56 mg/L (0-0.8 mg/L). Quantiferon test was negative. Diagnostic thoracentesis revealed that the effusion was an exudate (P/S LDH: 272/174 U/L, P/S protein: 5/7.5 g/dL, P/S Alb: 3.19/4.3 g/dL, P/S Glu: 95/97 mg/dL, ADA: 19.94 U/L). Direct microscopic examination and cultures of pleural effusion were unremarkable. Acid-fast staining, tuberculosis polymerase chain reaction (PCR) and culture for tuberculosis were all negative. Cytological examination revealed numerous lymphocytes. A closed pleural biopsy was performed and histopathologic examination was reported as "Nonspecific chronic inflammatory cell infiltration." Based on these findings pleural effusion was suggested to be due to AS. Besides the patient refused any further investigation that he was followed up under etanercept therapy. One year after the initiation of therapy, the pleural effusion was still present and slightly increased with an increasing pleural thickness (Figure 1b).

In regard to the presence of environmental asbestos exposure, a pleural biopsy with video assisted thoracoscopic surgery (VATS) was performed. Macroscopically there wasn't any tumoral infiltration. Multiple pleural biopsies obtained from different sites were compatible with chronic inflammation.

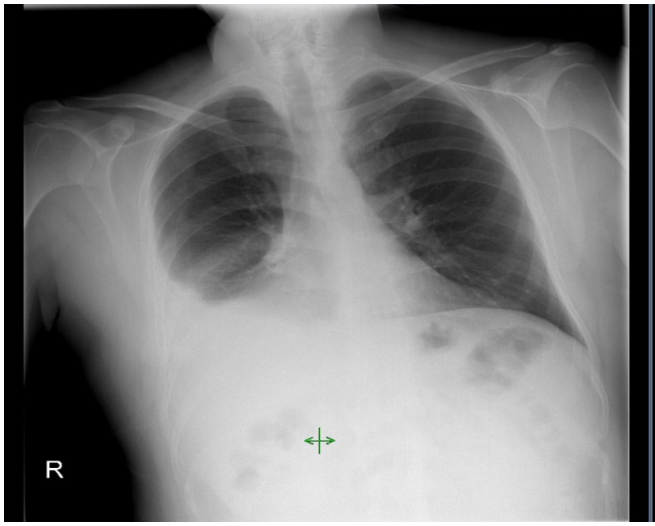

Figure 1a: Right sided pleural effusion seen on the chest x-ray which was performed before the initiation of anti-TNF therapy.

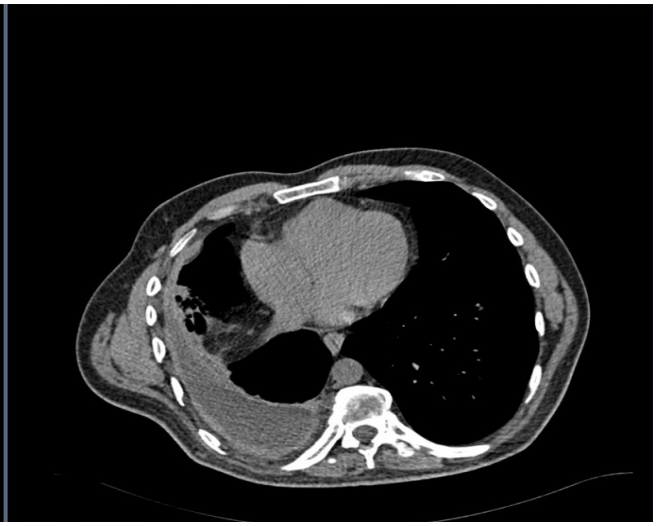

Figure 1b: Computed tomography of thorax showing persistent right sided pleural effusion and increased pleural thickness one year after the initiation of etanercept therapy.

DISCUSSION

Pleural effusion due to connective tissue disease (CTD) is a well known clinical entity especially in systemic lupus erythematosus and rheumatoid arthritis. It develops secondary to increased capillary permeability due to immune or non-immune inflammation [3]. Pleural effusion due to AS is extremely rare. Rosenow et al. reviewed 2080 patients with AS and disclosed that 28 patients (1.3%) had pleuropulmonary manifestations. While the most common involvement was upper lobe fibrobullous disease, only 3 patients (1.4/1000) had transient exudative pleural effusion. Two of these patients had non-specific pleuritis on pleural biopsy [2].

Pleural effusion developed in AS patients is usually exudative with a normal pH and glucose values. Inflammatory cells including eosinophils have been identified in cytological examination [3, 4]. Other than involvement of the pleura, pleural effusion can be secondary to the drugs (sulfasalazine therapy) used for treatment of AS [5] or bilateral transudative effusion due to involvement of heart [6, 7].

In the present case, the pleural effusion was also an exudate with normal glucose level. The predominant inflammatory cells were lymphocytes. Histopathologic examination of two pleural biopsies (closed and VATS pleural biopsy) performed one year apart were compatible with chronic inflammation. Therefore we suggested that this pleural effusion was associated with involvement of pleura due to AS. We excluded sulfasalazine drug reaction since the pleural fluid persisted after the drug was discontinued.

In the literature, there are different treatment approaches for pleural involvement in patients with

AS. In some patients pleural effusion can resolve spontaneously. Systemic prednisolone (30-60 mg/day), intrapleural steroids (20 mg prednisolone after pleural effusion aspiration) and phenylbutazone (200 mg/day) had been successfully used in the treatment of pleural effusion [8-10]. Kinnear WJ et al. reported a case with bilateral exudative pleural effusion which was resolved after 32 mg systemic steroid therapy [8]. In the case reported by of Tanaka et al. pleural effusion was decreased after intrapleural steroid therapy [9]. In another case reported by Erkan L et al., pleural and pericardial effusion was developed 6 months after the diagnosis of AS. The pleural effusion was again an exudate and histopathologic examination of pleural biopsy denied any malignant infiltration or granulomatous inflammation. Both the pleural and pericardial effusion disappeared after the treatment of 60 mg of prednisolone and did not recur in the following 2 years [10].

It is well known that anti-TNF therapy improve clinical signs and symptoms, patient's physical function and quality of life and normalize the increased acute phase response [11]. In the present patient anti-TNF therapy was initiated. Despite anti-TNF therapy pleural effusion has continued to be stable after 1 year. Since the patient was asymptomatic at that time, he did not receive systemic steroids.

In conclusion; pleural effusion is a rare manifestation of AS. It develops secondary to increased capillary permeability due to immune or non-immune inflammation. It is mainly exudative and can be diagnosed via exclusion of other etiologies such as parapneumonic effusion, tuberculosis and malignancies. Pleural biopsies can be performed. They mostly revealed chronic inflammation. The treatment of AS associated pleural effusion is not well documented, but systemic and intrapleural steroids can be used in selected symptomatic cases.

CONFLICT of INTEREST STATEMENT

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

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