Case Report

Acute Pleuritis in Sarcoidosis

Aditya Jindal[1] and Alok Gupta[2]

Jindal Clinics[1], Altus Laboratories[2], Chandigarh, India

Abstract

We report a case of a 48-year-old housewife diagnosed to have sarcoidosis based upon the characteristic clinical features and presence of non-caseating granulomas on endobronchial ultrasound guided fine needle aspiration biopsy. After an year, she developed severe chest pain due to acute pleuritis and dramatically responded to corticosteroids. To the best of our knowledge; acute pleuritis is a very rare presentation of sarcoidosis and has not been reported in the literature in recent times. **[Indian J Chest Dis Allied Sci 2017;59:91-93]**

Key words: Sarcoidosis, Acute pleuritis, Endobronchial.

Introduction

Although uncommon, pleural effusion in sarcoidosis is described in several studies from time to time, with a reported incidence in 3% to 10% patients.[1-8]

Pleural effusion reported in sarcoidosis is generally exudative; though transudative effusions have also been described,[2,9] and these are occasionally bilateral as well as massive. Rarely, haemorrhagic, chylous and eosionphilic effusions have been reported.[2,6,10-14] Pleural thickening has also been reported in about 33% of patients on chest computed tomography (CT), mostly in patients with pulmonary fibrosis.[15] *To the best of our knowledge*, presence of acute pleuritis in sarcoidosis as seen in our patient has not been described earlier.

Case Report

A 48-year-old housewife presented with complaints of occasional chest pain for few weeks around 15 months back. There was no other pulmonary or systemic symptom. General physical and systemic examinations were within normal limits. Chest radiograph and computed tomography (CT) showed bilateral hilar and mediastinal lymph node enlargement. There was no effusion or pleural thickening either on chest radiograph or on CT (Figure 1 A, B and C). Routine biochemical investigations were normal; but serum angiotensin converting enzyme (SACE) value was elevated at 82 U/L (normal range 8-65U/L).

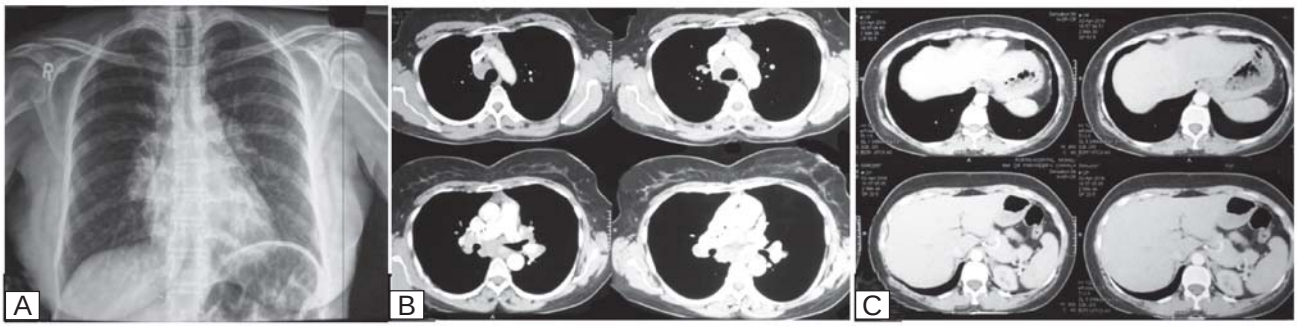

Figure 1. (A) Chest radiograph (postero-anterior view) showing bilateral hilar and right paratracheal lymphadenopathy with normal bilateral lung fields and clear costophrenic angles, (B and C) contrast-enhanced computed tomography of chest (axial view) showing enlarged hilar and mediastinal lymph nodes without necrosis. There is no evidence of pleural effusion or thickening.

Endobronchial ultrasound (EBUS) guided transbronchial needle aspiration revealed non-necrotising granulomatous inflammation consistent with the diagnosis of sarcoidosis (Figure 2A and B). The patient was already receiving some indigenous medications. In view of the asymptomatic Stage I disease, corticosteroid therapy was stopped. Patient was prescribed hydroxychloroquine for a few weeks; but this drug was also discontinued by the patient on her own.

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Correspondence and reprint requests: Dr Aditya Jindal, Jindal Clinics, SCO 21, Sector 20 D, Chandigarh-160 020, India; E-mail: adijindal@gmail.com

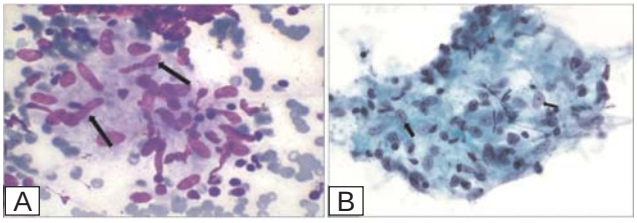

Figure 2. Histopathological photograph of endobronchial ultrasound guided fine needle aspiration showing (A) non-caseating granulomas with epitheloid cells (arrows) background showing lymphoid cells (Haematoxylin and Eosin × 100); (B) showing epitheliod cells but no evidence of acid-fast bacilli (Ziehl-Nelsen stain × 40).

The patient reported to us after a period of about 15 months, with a history of severe, unbearable chest pain in the left lower chest which increases with respiratory movements and coughing for the last few days. The patient did not get any relief in chest pain after taking several analgesic drugs. On respiratory examination, she was found to have tenderness on deep palpation and an audible pleural rub in the left infra-axillary and infra-scapular regions. Chest radiograph and CT showed almost the same size of hilar and mediastinal lymph nodes as earlier; without any parenchymal lesion. In addition, there was minimal blunting of left cardiophrenic angle on chest radiograph (Figure 3A) and mild pleural thickening on CT of chest (Figure 3B). These findings were attributed to acute pleural inflammation.

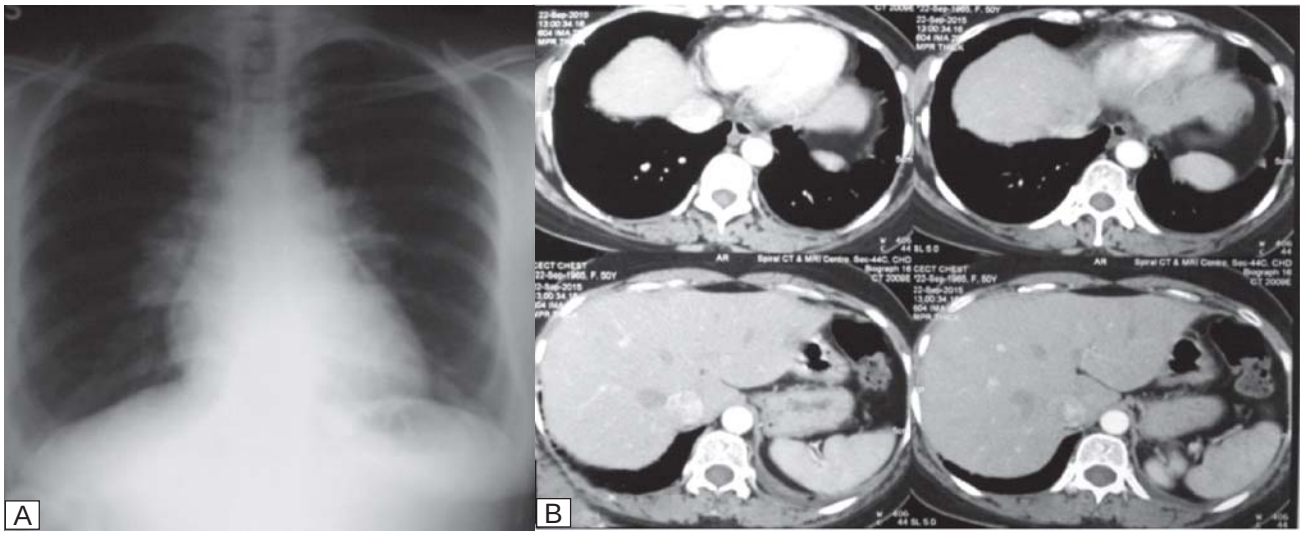

Figure 3. (A) Chest radiograph showing minimal blunting of left costophrenic angle and (B) contrast-enhanced CT chest (axial view) showing mild left pleural thickening.

In view of the severe intractable pain with an established diagnosis of sarcoidosis earlier, the patient was administered intramuscular injection of hydrocortisone (100 mg) and started on oral prednisolone (30 mg per day). Chest pain and tenderness as well as pleural rub decreased significantly on auscultation within 24 hours and almost completely disappeared within a week. She was continued on the same medication for six weeks followed by gradual tapering to a maintenance dose of 7.5 mg per day of oral prednisolone over three months.

On follow-up after six months, she was doing well with a normal repeat chest radiograph. Ultrasound examination of chest did not show any pleural effusion or thickening.

Discussion

Our patient had initially presented with non-specific chest pain off and on without any obvious evidence of pleural involvement for over a year. She was diagnosed to have sarcoidosis on radiological features, high angiotensin converting enzyme level and presence of non-caseating granulomas on EBUS-guided lymph node aspirate. There was no obvious pleural involvement. Therefore, chest pain was not initially attributed to pleural disease. Apparently, the patient developed acute pleuritis after over a year of her initial diagnosis of sarcoidosis. Acute pleuritis was diagnosed on the basis of clinical features of acute onset of chest pain, tenderness and pleural rub substantiated by CT evidence of pleural thickening; which was not observed in the previous CT. Moreover, the clinical features as well as the radiological findings completely resolved after treatment.

Pleural involvement in sarcoidosis manifesting as pleural effusion has been reported from time to time.[7-11] Pleural effusion is known to lead to pleural fibrosis and thickening. Pleural thickening can also happen in the presence of parenchymal fibrosis. It was reported in 20 of 61 patients (33%) on thoracic CT in a study from Mount Sinai Hospital.[15] Most of these patients had advanced disease with parenchymal

fibrosis which was the apparent cause of restrictive physiology in these patients.

In one study,[16] chest pain in sarcoidosis was reported in 14 of 22 patients; only 4 of these patients had chest pain as the primary symptom. However, no significant correlation of pain with presence or location of pleural disease or other thoracic structures was observed in these patients on chest CT. None of these patients had any clinical features suggestive of acute pleural inflammation.

In our patient, corticosteroid therapy was not initially started at first presentation in view of Stage I disease with minimal symptoms. Later on, she developed acute pleuritis with severe chest pain and corticosteroid therapy was administered. The patient responded extremely well with complete resolution of acute pleuritis. In view of the temporal course of the patient, it is postulated that pleural involvement in sarcoidosis can also manifest with acute, dry pleurisy. Moreover, acute pleuritis in the presence of Stage I disease should be considered as an indication for corticosteroid therapy.

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