

Case Report

An interesting case of Contarini's syndrome in a young patient

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ABSTRACT

Pleural effusion is a common disease of the pleura. The pleural effusion is usually unilateral and bilateral pleural effusion is less commonly seen. In bilateral pleural effusions usually the same etiology is present on both the sides but bilateral pleural effusions due to the two different etiologies on the two sides is rare. Here we are describing an interesting case of Contarini's syndrome where exudative pleural effusion was present on each side but due to the two different etiologies on the two sides.

Keywords: Bilateral pleural effusion, Contarini's syndrome, Bilateral thoracentesis

INTRODUCTION

Bilateral pleural effusion is not very common and constitutes about (20-25) % of all pleural effusion cases. Bilateral exudative pleural effusion is usually more commonly seen than the bilateral transudative pleural effusion. Malignancy is usually the most common cause of bilateral exudative pleural effusion followed by other systemic diseases but infection is usually a less common cause of bilateral pleural effusion.¹ Here we are reporting an interesting case of bilateral pleural effusion due to the two different infective etiologies in the two pleural effusions also known as Contarini's syndrome.

CASE REPORT

A 30-year-old male patient presented to our department with complaints of cough with expectoration for two months, high grade fever for two months along with loss of appetite and bilateral chest pain for one month. Patient also had streaky hemoptysis for the last fifteen days. The patient had no previous history of any anti-tubercular treatment. There was no history of diabetes mellitus in the patient. No other comorbid disease was present in the patient. The patient was a bidi smoker with tobacco

chewer. Patient was an occasional alcoholic. He was a crane worker with exposure to particulate dust throughout the day. The general examination in the patient revealed pallor with a temperature of 102.8°F. The blood pressure in the patient was 100/66 mm Hg with a pulse rate of 98/min and a respiratory rate of 24/min. The respiratory system examination revealed bronchial breath sound in the right suprascapular and infraclavicular area with vesicular breath on the left side and the breath sounds were diminished in the infrascapular area on both the sides. Bilateral fine crepts were present in the suprascapular and the infraclavicular area. The systemic examination was normal.

Investigations

The blood examination revealed a Hb%- 9.2 gm/dl, TLC- 12,600/mm³, Platelets- 2.1 lakh/mm³. Liver function test and kidney function test were normal in the patient. Blood sugar was normal in the patient. The viral markers for HIV, HCV and HBsAg were negative in the patient. Chest X-ray showed heterogenous opacities in right upper and mid lung zones with similar opacities in left upper zone (Figure 1). Chest X-ray also showed loculated pleural effusion on both the sides. HRCT Thorax showed bilateral lung

parenchymal opacities in both the upper lobes suggestive of bilateral pulmonary tuberculosis (Figure 2).

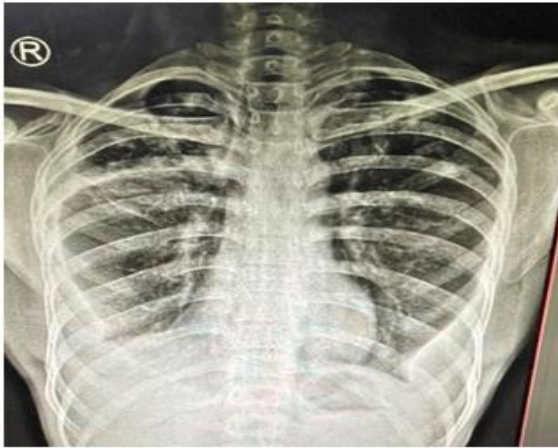


Figure 1: Chest x-ray PA view at the start of antitubercular treatment in the patient showing bilateral loculated pleural effusion with pulmonary tuberculosis.

Cy-TB test was positive with an induration of 8 mm after 48 hours. Ultrasound thorax revealed anechoic pleural fluid with a sharp margin due to the loculation on right side while there was a cloudy and speckled pleural fluid with multiple web-like white strands suggestive of fibrin septa and with a sharp margin on the left side. Pleural fluid examination from right side showed a TLC- 2500 cells/mm³, DLC- Neutrophils 20% and Lymphocytes 80%, Protein- 3.8 mg/dl, Sugar- 78 mg/dl and ADA- 52 U/L.

The pleural fluid examination from the left side showed a TLC- 9500 cells/mm³, DLC- Neutrophils 70% and Lymphocytes 30%, Protein- 3.4 mg/dl, Sugar- 35 mg/dl and ADA- 185 U/L. The pleural fluid from right side was straw coloured while that from the left side was slightly turbid. Pleural fluid triglycerides and cholesterol level were normal on both the sides. The bacterial culture of the pleural fluid from the left side was negative. Sputum for CBNAAT (Cartridge based nucleic acid amplification test) for MTB (*Mycobacterium tuberculosis*) was positive and MTB was sensitive to Rifampicin.

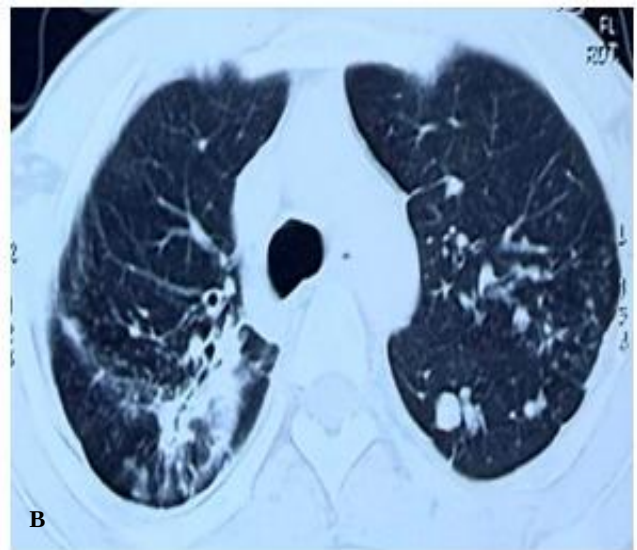


Figure 2 (A and B): HRCT Thorax showing bilateral lung parenchymal opacities suggestive of bilateral pulmonary tuberculosis.

Treatment and follow up

The patient was initiated on the four drug antitubercular treatment with Rifampicin (600 mg), Isoniazid (300 mg), Ethambutol (1100 mg) and Pyrazinamide (1600 mg) as per the weight of the patient. He was also given Ceftriaxone-Sulbactam injection intravenously in a dose of 1.5 gram twice daily for seven days. After two weeks of antitubercular treatment, there was significant relief in the high-grade fever, cough and right sided chest pain. Cough and right sided chest pain gradually subsided but there was persistence of left sided chest pain and intermittent low-grade fever even after six months of antitubercular

treatment. The chest x-ray after six months of antitubercular treatment showed persistence of left sided loculated pleural effusion (Figure 3) and hence antitubercular treatment was extended for one month. But there was persistence of left sided loculated pleural effusion on the chest x-ray even after seven months of antitubercular treatment (Figure 4) and hence the left sided pleural effusion was then aspirated. This time it was more seropurulent in appearance. The CBNAAT MTB and MTB liquid culture were negative in the pleural fluid from the left side. The left sided pleural fluid was again sent for gram stain and bacterial culture which showed gram negative bacilli with ESBL (extended spectrum beta-lactamase) producing *Escherichia coli*. A pigtail catheter

was placed in the left pleural space (Figure 5 and Figure 6) and around 600 ml of turbid fluid was drained. The patient was given Meropenem 1 gm iv thrice daily for ten days as per the culture and sensitivity report of the pleural fluid from the left side. After ten days the pigtail catheter was removed and the patient was discharged on Faropenem 300 mg thrice daily for the next ten days. A repeat chest x-ray after ten days showed almost complete resolution of left sided loculated pleural effusion (Figure 7) and the patient was relieved of his symptoms.

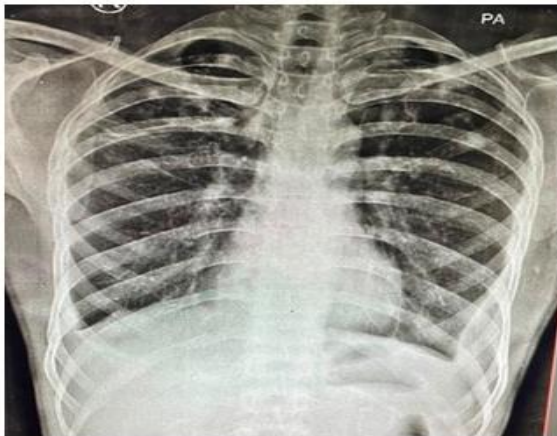


Figure 3: Chest x-ray PA view at the end of six months of antitubercular treatment showing persistent left sided loculated pleural effusion in the patient.



Figure 4: Chest x-ray PA view at the end of seven months of antitubercular treatment in the patient showing persistent left sided loculated pleural effusion.

Differential diagnoses included malignant pleural effusion, pleural mesothelioma, chylothorax, benign asbestos-related pleural effusion, pleural effusion associated with connective tissue diseases such as systemic lupus erythematosus (SLE) and rheumatoid

arthritis (RA), pancreatitis-associated pleural effusion, and pulmonary embolism-related pleural effusion.

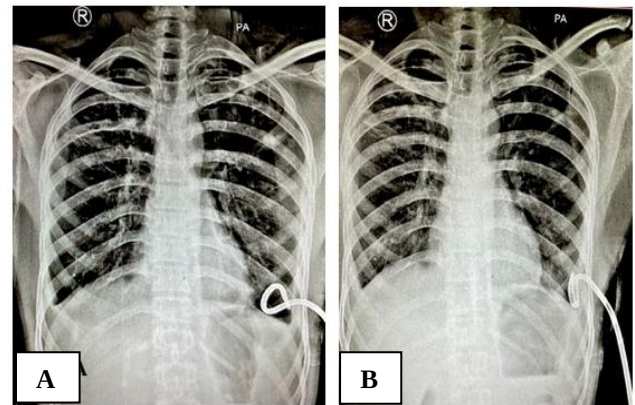


Figure 5: Chest x-rays showing Pigtail catheter in the left sided loculated pleural effusion- (A) on day 1 and (B) on day 8 showing complete drainage of left sided pleural effusion on day 8.

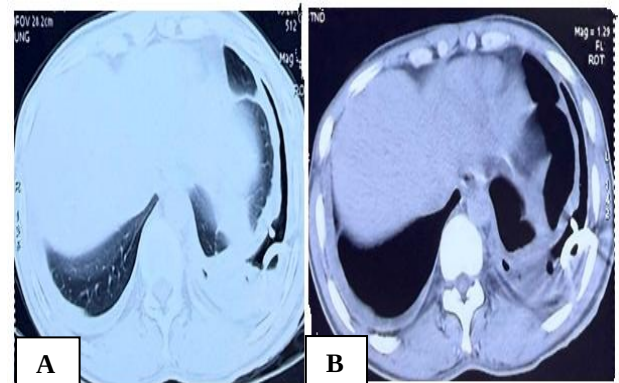


Figure 6 (A and B): HRCT Thorax showing left sided loculated pleural effusion with pigtail catheter in situ with pleural thickening.



Figure 7: Chest x-ray after ten days of removal of the Pigtail catheter from the left side and after completing his antibiotic course.

DISCUSSION

Pleural effusion is a common disease of the pleura. The unilateral pleural effusion is more common than the bilateral pleural effusion. The exudative pleural effusion is more common than the transudative pleural effusion in India.² The most common etiology of pleural effusion in India is tuberculosis in about 40% cases followed by parapneumonic effusion and malignancy.³ But our patient had bilateral pleural effusion which was exudative in nature. Bilateral pleural effusions usually have a single etiology but the term Contarini's syndrome refers to the occurrence of bilateral pleural effusion with different etiologies on the two sides.

The most common combination of etiology in Contarini's syndrome is a parapneumonic effusion triggering the heart failure in the patient leading to contralateral transudative pleural effusion.⁴ Vijayan et al reported bilateral pleural effusion with diagnosis of tuberculous pleural effusion on one side and the pleural effusion due to pancreatic disease on the other side.⁵ Shah et al also reported a case of bilateral pleural effusion with a transudative pleural effusion due to liver cirrhosis on one side and a parapneumonic exudative pleural effusion on the other side.⁶ Similarly Gupta et al reported a case of Contarini's syndrome with malignant pleural effusion on one side and a transudative pleural effusion due to hypoproteinemia on the other side.⁷ But in our case of bilateral pleural effusion, the patient had tuberculous pleural effusion on one side and a bacterial empyema on the other side.

Bilateral thoracentesis is usually not required in patients with bilateral pleural effusions due to the same etiology but Shizuku et al emphasized the importance of bilateral thoracentesis in patients with Contarini's syndrome.⁸ Bilateral thoracentesis is usually required when there is unilateral lung parenchymal involvement along with bilateral pleural effusions, persistence of pleural effusion despite adequate treatment of the pleural effusion, recurrence of pleural effusion despite optimal treatment of the pleural effusion or the thoracic imaging shows different morphology of the pleural effusions on the two sides. Patients with Contarini's syndrome having an empyema, hemothorax, chylothorax or malignant pleural effusion on one side may require placement of an intercostal drainage tube or pigtail catheter on that side.⁹ In our patient also a pigtail catheter was placed to adequately drain the empyema on the left side which was removed after ten days.

CONCLUSION

Contarini's syndrome is an uncommon pleural disease which requires bilateral thoracentesis to identify the two different etiologies on the two sides of the thorax. The treatment of this syndrome will be difficult if the two different etiologies for the bilateral pleural effusions are not properly identified.

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