Case Report

Secondary spontaneous pneumothorax in a patient of allergic bronchopulmonary aspergillosis: an unusual presentation

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ABSTRACT

Allergic bronchopulmonary aspergillosis (ABPA) is a complex immunologic pulmonary disorder caused by hypersensitivity to fungus, Aspergillus fumigates. It clinically manifests with non-specific respiratory and systemic symptoms. ABPA is typically seen in patients with long-standing asthma or cystic fibrosis. Pleural involvement in ABPA is uncommon and secondary spontaneous pneumothorax is very rare. Herein, we report a case of 33 years old male patient presented with dyspnoea, low grade fever and productive cough. High Resolution Computed tomography (HRCT) scan of thorax was suggestive of ABPA with secondary pneumothorax.

Keywords: Allergic bronchopulmonary aspergillosis, Aspergillus fumigates, High resolution computed tomography, Secondary spontaneous pneumothorax

INTRODUCTION

Allergic bronchopulmonary aspergillosis (ABPA) was first described in 1952 from the United Kingdom by Hinson et al.¹ It is a hypersensitivity reaction to Aspergillus antigens and is usually caused by Aspergillus fumigatus. It is typically seen in patients with long-standing asthma or cystic fibrosis. It is believed that the Aspergillus-specific IgE-mediated type I hypersensitivity reaction and the specific IgG-mediated type III hypersensitivity reactions play an important role in the pathogenesis of ABPA.²

The estimated global burden of ABPA is about 4.8 million cases with about 1.38 million cases in India alone. The diagnosis of ABPA is based on a combination of clinical, radiological and immunological findings.³

It clinically manifests with non-specific symptoms such as low grade fever, wheezing, productive cough and dyspnoea. In ABPA, exacerbations are characterized by episodes of increasing dyspnea, cough with sputum production, pleuritic pain and fever. The sputum of the patients may consist of brownish mucous plugs and hemoptysis is rare. Examination reveals rhonchi and wheezing on auscultation of the chest during acute exacerbation. Laboratory examination in ABPA may reveal peripheral blood eosinophilia. Total serum IgE is often >1000 ng/ml.⁴

Chest radiographs that demonstrate fleeting parenchymal opacities or bronchiectasis should trigger a consideration of ABPA. Opacities may also reflect bronchoceles, mucus plugging, atelectasis, or lobar collapse. On high resolution computed tomography (HRCT) of thorax, the presence of central bronchiectasis in multiple lobes is highly suggestive of ABPA. Mucus plugging can manifest a “finger in glove” appearance from impacted mucus occluding an airway and its branches. Perilesional infiltrates and nodules are often seen in active phase. Fibrosis and cavitation of dilated airways are end-stage findings in ABPA.⁵ Radiologically, ABPA is classified
depending on the absence or presence of bronchiectasis and high attenuation mucus.⁶

Diagnostic criteria of ABPA are: obligatory criteria: asthma, elevated serum level of total IgE (>1000 IU/mL), elevated serum level of IgE against A. fumigatus. Other criteria: positive Aspergillus skin test (type I hypersensitivity reaction), presence of serum precipitins (IgG) against A. fumigatus, fleeting pulmonary infiltrates on chest X-ray, total eosinophil count in peripheral blood >1000 cells/mL, central bronchiectasis on HRCT thorax. Presence of all obligatory criteria and three of five other criteria confirms the diagnosis.⁷

Pleural involvement is less common but can occur, including effusions, pleural thickening, and calcifications.⁸ Pleural involvement generally occur due to parenchymal lesions extending up to the pleural surface The other reported pleural manifestation is secondary spontaneous pneumothorax which is very rare.⁶

Pneumothorax is defined as the presence of air in the pleural space. A secondary spontaneous pneumothorax (SSP) is defined as a pneumothorax that occurs as a complication of underlying lung disease. In contrast, primary spontaneous pneumothorax occurs without a precipitating event in the absence of clinical lung disease.⁹,¹⁰

CASE REPORT

A 33 years male patient presented with history of cough with expectoration and low grade fever for one week and shortness of breath for last two days.

Figure 1: HRCT thorax, axial image showing central bronchiectasis with “finger in glove” pattern due to high attenuation mucus within dilated bronchi in left hemithorax (white arrows). Right sided Pneumothorax with dilated bronchioles seen in direct communication with pleural air suggestive of bronchopulmonary communication (red arrow). Associated minimal pleural fluid is seen in right pleural space in dependent part.

History of atopy and recurrent chest infection was present for last 2 years. He was non-smoker and non-alcoholic. He was farmer by occupation. No past history and family history of tuberculosis was there.

Figure 2: HRCT thorax, coronal reformatted image showing bilateral central bronchiectasis with “finger in glove” pattern due to high attenuation mucus within dilated bronchi in left hemithorax (white arrow). Right sided Pneumothorax with dilated bronchioles seen in direct communication with pleural air suggestive of bronchopulmonary communication (red arrows). Associated right lung collapse is seen.

On examination the patient was febrile (99.3°F), heart rate was 104 beats/min, blood pressure was 122/78 mmHg, and respiratory rate was 24 breaths/min. On auscultation, diffuse wheeze in left hemithorax was present with absent breath sounds in right hemithorax. On percussion, hyper-resonant note was revealed in right hemithorax.

Hematological investigations revealed hemoglobin 12 gm%, total leukocyte count 7100/mm3 with 25% eosinophils on differential count, ESR=40mm in 1st hour, fasting blood sugar 78 mg/dl with normal liver and renal functions. Immunological survey reveals total serum IgE=2750 IU/ml. Levels of serum IgG and IgE against A. fumigates were increased.

Sputum smear microscopy for acid fast bacilli was negative in three samples. Gram staining of sputum smear examination did not revealed any organisms.

High resolution CT scan of thorax revealed predominant central bronchiectasis along with “finger-in-glove” opacities due to mucoid impaction. Associated right sided pneumothorax was present due to communication between dilated bronchioles and pleura (broncho-pleural communication or fistula), resulting into collapse of right lung. Minimal pleural fluid was also seen. (Figure 1 and 2)

Clinical, hematological, immunological and radiological features suggested diagnosis of ABPA complicating as secondary spontaneous pneumothorax.

Patient was treated with intercostal drainage under water seal for pneumothorax. Nebulisation, O₂ inhalaton, antibiotics, antifungal and oral steroids were given.
DISCUSSION

Pleural involvement in ABPA is uncommon, and involvement of the pleura has been described in the form of parenchymal lesions extending up to the pleural surface or pleural thickening on radiology in up to 43-82% patients with ABPA, in various small series.

Secondary spontaneous pneumothorax a rarely reported clinical presentation or complication of ABPA. Only few cases of secondary spontaneous pneumothorax are reported in literature.

Ricketti et al in 1984 reported a case spontaneous pneumothorax occurred in a patient with well-advanced (stage V) allergic bronchopulmonary aspergillosis (ABPA). The pneumothorax responded to chest tube evacuation.11

Judson et al in 1993 described a case report of secondary spontaneous pneumothorax with bronchopleural fistula in ABPA.12

Denning et al in 2003 reported Secondary pneumothorax (bullae) in 16.7% (21 out of 126) of chronic ABPA. Six of these patients had pneumothorax plus bullae, while the remainder had pneumothorax without bullae. Camuset et al in 2007 and Smith et al in 2011 also reported few cases of secondary pneumothorax in ABPA in small series.13-15

Gupta et al in 2007 reported a case of Pneumothorax due to rupture of a mycetoma into the pleural space in an immune-competent patient.16

Zhang et al in 2010 reported a case of recurrent pneumothorax in a patient suffering from ABPA. They believed that pneumothorax in their case was caused by rupture of a subpleural bleb. The three occurrences of spontaneous pneumothorax in less than half a year indicated that some underlying cause resulted in progressive hyperinflation and damaged these blebs.17

Das et al in 2014 described two cases of right-sided pneumothorax and right upper lobe collapse in patients of allergic bronchopulmonary aspergillosis.18

Five clinical stages of ABPA has been described by Patterson et al i.e. acute stage, stage of remission, stage of exacerbation, stage of steroid dependent asthma and fibrotic stage. Pleural complications like pleural effusion, pneumothorax and pleural thickening are mainly seen in fibrotic stage of disease.19,20

Radiologically, ABPA is classified as seropositive ABPA (ABPA-S), ABPA with central bronchiectasis (ABPA-CB) and ABPA with central bronchiectasis and with high attenuation mucus (ABPA-CB-HAM), depending on the absence or presence of bronchiectasis and high attenuation mucus.21 Although radiographic findings may be normal, findings in early-stage disease typically include transient pulmonary opacities or homogeneous, tubular, gloved-finger areas of increased opacity in a bronchial distribution, usually either predominantly or exclusively involving the upper and central lungs. Fibrotic lung stage in ABPA may, at times, be difficult to differentiate from the fibrocavitary lesions of tuberculosis.19

It is found that HRCT had a sensitivity of 83% and a specificity of 92.5% when compared to bronchography.22 HRCT is modality of choice to diagnose ABPA and its complications. In case of secondary spontaneous pneumothorax HRCT examination is required to demonstrate the cause i.e., subpleural blebs, cavity communicating with pleura or broncho-pleural fistula etc. HRCT is also helpful to rule out other related lung disorders like tuberculosis.

High prevalence of ABPA reported in hospital studies from India, ABPA is still under-recognised and under-diagnosed. There is high prevalence of tuberculosis (TB) in India and due to ignorance and lack of suspicion, a large number of ABPA cases may be mis-diagnosed as TB and go on receiving prescribed anti-tuberculosis treatment. In some Indian studies, ABPA was misdiagnosed as TB in as high as 17% to 50% cases.23 Only Few cases of ABPA with pleural complications are reported from India till date.

CONCLUSION

In conclusion, secondary spontaneous pneumothorax is rare manifestations of allergic bronchopulmonary aspergillosis. It should be considered in the differential diagnosis of such cases, particularly for those with a chronic lung disease. ABPA is still under-diagnosed especially in India due to high prevalence of pulmonary tuberculosis. HRCT examination is useful in diagnosing ABPA and complications associated with it.

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