Allergic broncho-pulmonary aspergillosis with aspergilloma developing in a cane sugar mill worker

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Citation

Abstract
Coexistence of allergic bronchopulmonary aspergillosis (ABPA), a hypersensitivity disorder to Aspergillus antigen and aspergilloma, a saprophytic colonization of Aspergillus into tracheobronchial tree in same individual is of rare occurrence. Here we report a case of a 37 year male who is a sugar mill worker by occupation. He had taken two courses of adequate anti tubercular treatment without any improvement and was found to have ABPA with aspergilloma.

INTRODUCTION
Pulmonary aspergillosis is a clinical spectrum of lung disease caused by the fungus Aspergillus species. The spectrum includes saprophytic aspergillosis in the form of pulmonary aspergilloma, immune disease in the form of ABPA and hypersensitivity pneumonitis, and infectious disease in the form of invasive and semi-invasive necrotizing aspergillosis. Cavitation is known to occur in ABPA, but coexistent aspergilloma is rather uncommon. Reported hereunder is a case of ABPA with aspergilloma, in a cane sugar mill worker.

CASE REPORT
A 37 year old non smoker, male patient, sugar mill worker by occupation presented to us with chief complaints of recurrent cough, breathlessness and haemoptysis for 13 years. He also gave history of recurrent low-grade febrile episodes associated with malaise without any weight loss, and passage of mucus plugs with sputum. He had never smoked, consumed alcohol, or chewed tobacco and had no history of diabetes mellitus or hypertension. There was no family history of hypertension, diabetes mellitus, immunodeficiency disease or neoplasm. Patient was having no history of contact to a tuberculosis patient. He had received two courses of adequate anti tubercular treatment without any bacteriological evidence of Mycobacterium tuberculosis. On physical examination, the patient was afebrile with a pulse rate of 96/min, respiratory rate of 26/min and a right arm supine blood pressure of 120/68 mmHg. There was no pallor, cyanosis or clubbing. Chest examination was unremarkable on inspection, palpation and percussion. On auscultation bilateral rhonchi were audible. Examination of other systems was unremarkable.

Routine investigation showed; hemoglobin: 11 gm%, total leukocyte count: 14800/mm³, differential count: neutrophils 52%, lymphocyte 30%, eosinophil 18% (absolute eosinophils count was 2664), platelets count: 2.9 lacs/mm³ and Erythrocyte sedimentation rate: 18 mm/hr. ELISA for human immunodeficiency virus was negative. Sputum smears for acid-fast bacilli were repeatedly negative and the culture by BACTEC did not show any mycobacteria. But the sputum on fungal culture grew aspergillus fumigatus. Mantoux test (10 tuberculin units) showed an induration of 4mm at 72 hours. On further evaluation, his detailed history revealed that he had episodes of running nose, sneezing, breathlessness and wheezes, since childhood, especially with change of season in months of March and November for which he received symptomatic medications. Family history of bronchial asthma was also present in his mother. Review of his serial chest radiographs revealed evidence of cavity with ‘air crescent sign’ in right middle zone, gloved finger appearance in left upper zone and fleeting pulmonary infiltrates.

High resolution computed tomography (HRCT) of thorax revealed bilateral central bronchiectasis with aspergilloma in anterior segment of right upper lobe (fig.1).
Thus a possibility of Allergic broncho-pulmonary aspergillosis with aspergilloma was suspected. Patient was investigated further for this. Skin prick test with Aspergillus fumigatus antigen showed a positive reaction for type I and also late type III (Arthus) hypersensitivity in comparison to positive control (Histamine Phosphate 1mg/ml). Total IgE was 2504 IU/ml (reference range 0 to 100 IU/ml). Specific IgG and IgE against A. fumigatus by ELISA were 124.2 IU/lit and 24.24 KU/lit. Thus a diagnosis of ABPA with concomitant aspergilloma was established.

Oral corticosteroid being the mainstay of treatment, the patient was initiated on oral prednisolone with a dose of 40 mg once daily along with inhaled budesonide 400 g with formetrol in two divided doses and inhaled salbutamol as and when required. After one month of therapy, the patient had improved clinically and radiographic pulmonary infiltrates got cleared, along with fall in serum titres of IgE antibody.

DISCUSSION

Regarding coexistence of ABPA and aspergilloma, it is postulated that, in genetically predisposed individuals, aspergilloma may act as nidus for antigenic exposure leading to development of ABPA. On the other hand, it is also said that chronic lung damage induced by ABPA appears to provide a favorable conditions for the formation of aspergilloma which might be accelerated by therapy with corticosteroids. Although cavitations is uncommon in a case of ABPA, but it may occur in 3% of cases, which can be colonized by Aspergillus to form aspergilloma.

Occurrence of ABPA in our patient who is a sugar mill worker is significant in the light of previous literature on occurrence of this disease entity in sugar mill workers. Process of lung destruction proceeds silently in ABPA, as about one third of these cases are completely asymptomatic. In present case it appears that patient had already developed central bronchiectasis and aspergilloma.

It was this indolent course of disease which prevented our patient from seeking timely medical advice on one hand and delayed the diagnosis on the other hand. Frequently, symptoms like haemoptysis, cough, fever etc. caused by ABPA/aspergilloma are attributed to active tuberculosis and managed incorrectly, as happened in our case. Since it is not uncommon to detect evidence of hypersensitivity to Aspergillus, in presence of aspergilloma, and aspergilloma can develop as a complication of ABPA, it may almost be impossible to establish whether ABPA followed or preceded the development of aspergilloma.

A wide variety of chest radiographic changes are known to be associated with ABPA. These changes include normal chest radiography, hyperinflation, various infiltrate patterns, consolidation, parallel lines and ring shadows, nodules, avascular areas, “honeycombing”, “tooth paste” shadows, “gloved finger”, band shadows and tramline shadows, changes like fibrosing alveolitis, lobar shrinkage and atelectasis as well as pseudohilar adenopathy and pleural thickening. Although the chest radiographic appearance is commonly abnormal in patients with ABPA, it is not invariably so, nor does a normal appearance exclude the diagnosis of ABPA. CT of the thorax can provide a sensitive method for the assessment of bronchial, parenchymal and pleural abnormalities in patients with ABPA and should constitute a part of the diagnostic work-up of the disease along with plain chest radiographs.

Oral corticosteroids, to date, remain the cornerstone for the treatment of ABPA. The exact role of antifungal agents is yet to be determined for both ABPA as well as aspergilloma. Specific therapy is not required for patients with asymptomatic aspergilloma. Surgical treatment carries risk with mortality ranging from 7% to 23%, due to which it should be considered in patients with massive haemoptysis and adequate pulmonary reserve.

References

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