Multiple Bronchoceles in a Non-Asthmatic Patient with Allergic Bronchopulmonary Aspergillosis

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Abstact

Allergic bronchopulmonary aspergillosis (ABPA) is a hypersensitivity reaction due to a fungus, Aspergillus fumigatus. It is typically seen in patients with long-standing asthma. Our patient was a non-asthmatic 18 years old male who presented with chronic cough for 2 years. Peripheral blood eosinophilia and elevated serum IgE were observed. His x-ray chest revealed v- shaped opacity in the left upper lobe close to the hilum. High resolution computed tomographic scan of the chest revealed multiple dilated bronchi filled with mucous (bronchoceles) and central bronchiectasis (CB) involving main segmental bronchi. Central bronchiectasis (CB) was typical of ABPA but bronchocele formation was a rare manifestation of the disease. The patient was managed with oral prednisolone and was relieved of his symptoms. Occurrence of ABPA in non-asthmatics is very rare and deserves reporting.

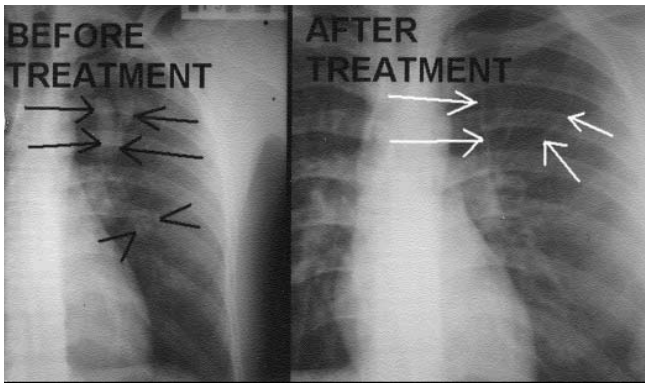
Introduction

Allergic Bronchopulmonary Aspergillosis (ABPA) is a hypersensitivity disorder induced by Aspergillus species colonizing the bronchial tree. Aspergillus is a ubiquitous soil dwelling fungus. It is commonly isolated as an upper respiratory tract saprophyte and a frequent containment in laboratory specimens. From over 200 species that belong to the Aspergillus group, Aspergillus fumigatus and Aspergillus Niger produce disease in humans with significant frequency[1]. ABPA occurs in asthmatic patients and belongs to the hypersensitivity disorders induced by a fungus Aspergillus fumigatus. This results in elevated IgE titres.[2,3] Radiological techniques are important to diagnose ABPA. Imaging techniques help in establishing the diagnosis and monitoring the progress of the disease. Although the disease has received international attention, it is still not detected as frequently and as early as it should be. This results in patients receiving inappropriate therapy leading to lung damage which could have been prevented with early diagnosis. Demonstration of central bronchiectasis (CB) is considered a sine qua non for the diagnosis of the disease and when present, can be categorized as ABPA-CB[4].

Case Report

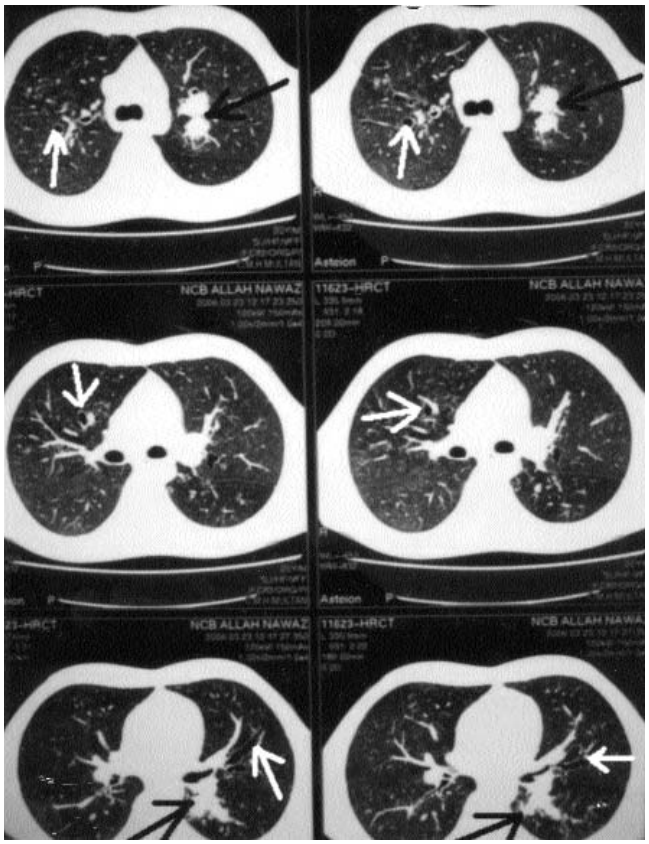
An 18 years old, non-smoker male patient presented with chronic productive cough of two years duration. There was no history of asthma, fever, night sweats or weight loss. There was no family history of tuberculosis. His laboratory investigations showed Blood total leukocyte count (TLC) of 12.5x10^9/L. The differential leukocyte count (DLC) revealed neutrophils 58%, lymphocytes 31%, monocytes 3% and eosinophils14%. His haemoglobin was 12.6gm/dl and ESR 8mm at the end of one hour. His breath sounds were normal and there were no crepitations or wheeze. Sputum microscopy revealed eosinophils and fungal hyphae. The serum IgE antibodies were increased (2275 ng/mL). The X-ray chest showed v-shaped well defined soft opacity in the

left upper lobe close to the hilum. Both limbs of this opacity were well-visualized and extended upto 5cm distally. There was another well-defined rounded opacity inferolateral to the left hilum (figure 1). Both opacities were non-calcified. To characterize the opacities seen on X- Ray chest, High Resolution Computed Tomographic Scan (HRCT) of the chest was done. It revealed the V-shaped opacity seen on X-ray chest as a high attenuation branching structure that was characterized as a mucous filled bronchocele (mucocele). Both limbs of the bronchocele were well visualized. Additionally, there was another inverted V-shaped opacity close to left hilum which was characterized as another branching dilated bronchus filled with high attenuation mucous plug. There was also dilatation of central bronchi bilaterally without thickening of the bronchial walls. The typical tramline dilatation of the central bronchi was characterized as central bronchiectasis (CB). It was more marked on the left side (figure 2) and was not seen on X-ray chest. The combined features of central bronchiectasis and mucoceles on HRCT scan and laboratory findings of



CHEST X-RAYS BEFORE AND AFTER TREATMENT: Before treatment, V-shaped opacity (bronchocele) was seen in left upper lobe (arrows). Note, another bronchocele inferior to left hilum (arrowheads). After treatment, both bronchoceles have disappeared. Thin walls of the dilated mucous filled bronchocele remained with clearing of the mucous plug (white arrows).

peripheral blood eosinophilia and elevated serum IgE levels were diagnostic of ABPA Pulmonary function tests were carried out in this patient which were normal thus excluding bronchial asthma. He was managed as an outpatient with oral prednisolone tablets, 15mg thrice daily for 6 weeks. It was gradually tapered off over next three months. The patient was followed with regular X-ray chest every month. Repeat X-ray chest after one month of treatment revealed marked improvement in the appearance of bronchoceles. Only thin walls of upper lobe bronchocele remained with clearing of the mucous plug. Left sided infrahilar bronchocele disappeared completely (figure 1). A repeat DLC after five months revealed decrease of peripheral eosinophilia to 4%. His cough had also settled. His treatment was stopped with advice to avoid smoke and pollution.



HIGH RESOLUTION CT SCAN OF CHEST: HRCT demonstrates central bronchiectasis (white arrows). Dilated bronchi filled with mucous (bronchoceles) are also seen (black arrows).

Discussion

Diagnostic criteria for ABPA include the presence of asthma, a history of pulmonary infiltrates, peripheral blood eosinophilia, immediate-type skin reactivity, serum precipitating antibodies to Aspergillus-specific IgE and IgG and central bronchiectasis (CB). The disease is being recognized more frequently due to increased physician awareness and better diagnostic techniques. Bilateral, predominantly upper lobe bronchiectasis is seen most commonly in patients with cystic fibrosis and allergic bronchopulmonary aspergillosis.[5] Recognition of this disorder is important to avoid progression of bronchiectasis and lung parenchymal damage. Hoshino et al described a case of ABPA occurring in a non-asthmatic patient.[6] The mainstay of therapy for ABPA is oral corticosteroids to suppress the immunologic response to Aspergillus antigens and the secondary inflammatory reaction. Treatment with corticosteroids leads to the relief of bronchospasm, the clearing of pulmonary infiltrates, and a decrease in IgE level and peripheral eosinophilia. Soubani et al described mucoid impaction in dilated bronchi as a syndrome related to ABPA occurring in patients without asthma.[7]

In asthmatic patients, bronchiectasis affecting three or more lobes, centrilobular nodules, and mucoid impaction are

findings on high-resolution CT that are highly suggestive of allergic bronchopulmonary aspergillosis.[8] Allergic bronchopulmonary aspergillosis is seen most commonly in patients with long-standing bronchial asthma. At pathologic analysis, this form of aspergillosis is characterized by the presence of plugs of inspissated mucus containing Aspergillus organisms and eosinophils. This results in bronchial dilatation typically involving the segmental and subsegmental bronchi. In approximately 30% of patients, the impacted mucus has high attenuation or demonstrates frank calcification at CT. The radiologist plays a major role in the diagnosis of pulmonary Aspergillus infection. When radiographic findings are subtle or equivocal, CT frequently allows identification of the disease process as was seen in our case. Although imaging findings in various types of pulmonary aspergillosis may be nonspecific, in the appropriate clinical setting, familiarity with the thin-section CT findings may suggest and even help establish the specific diagnosis.[9]

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