Another rare presentation of allergic bronchopulmonary aspergillosis in a high burden tuberculosis country

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Abstract

Mediastinal lymphadenopathy is often associated with tuberculosis, especially in a high burden country like India. We present a case of an asymptomatic female, who had mediastinal lymphadenopathy and middle lobe collapse, both of which pointed towards a diagnosis of tuberculosis. Patient was later diagnosed as a case of allergic bronchopulmonary aspergillosis (ABPA) on basis of clinical, radiological and serological findings. Hence, even in a high burden country and in background of bronchial asthma, ABPA should be kept as a differential diagnosis in patients presenting with mediastinal lymphadenopathy.

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Introduction

Mediastinal lymphadenopathy is often associated with tuberculosis, especially in a high burden country like India [1]. A concomitant lobar collapse increases the suspicion further. However, mediastinal lymphadenopathy is a rare finding in allergic bronchopulmonary aspergillosis and only 4 cases have been reported. Here, we present a case of mediastinal lymphadenopathy and middle lobe collapse due to allergic bronchopulmonary aspergillosis (ABPA).

Case Report

An asymptomatic, 29-year-old female, got chest X-ray for medical examination which showed right-sided hilar prominence with right middle lobe collapse (Figure 1A). CECT chest was suggestive of enlarged mediastinal lymph nodes in prevascular, paratracheal, aortopulmonary, and subcarinal locations, largest measuring 25 x 16 mm, and right middle lobe bronchiectasis with hyperdense mucus impaction and collapse (Figure 2). Fibreoptic bronchoscopy showed thick secretions in the right middle lobe bronchus. Bronchoalveolar lavage (BAL) was taken and transbronchial needle aspiration (TBNA) from subcarinal lymph node was done and sent for investigations. BAL fluid was negative for Ziehl-Neelson and Gram's staining, Gene Xpert, and pyogenic culture. TBNA revealed numerous lymphoid cells with small lymphocytic predominance and few macrophages along with single branched fungal hyphae. Serum levels of angiotensin converting enzyme were within normal range (61 U/L). There was no history of fever and loss of appetite or weight, and there was no family history of tuberculosis. She had a history of intermittent cough with expectoration, wheezing, and expectoration of thick mucoid sputum for past 4 years with worsening of symptoms in the winter season. Also, the patient was on inhalation therapy for past 2 years on an as-needed basis. Tuberculin skin test did not show any induration. Spirometry was performed which showed an obstructive pattern with bronchodilator reversibility. So, a working diagnosis of bronchial asthma was made and allergic bronchopulmonary aspergillosis (ABPA) was kept as a differential. Total IgE, *Aspergillus fumigatus* specific IgE, and IgG were found to be raised (4444 kU/L, 11.30 kUA/L, and 130 mgA/L, respectively). Patient was diagnosed as ABPA according to diagnostic criteria proposed by Agarwal *et al.* [2], and was started on oral itraconazole 200 mg BD and prednisolone 0.75mg/kg OD for 6 weeks and later tapered off.

BAL fluid on fungal culture also showed growth of *Aspergillus flavus*, but unfortunately *A. flavus* specific IgE could not be done due to unavailability of testing facility. On follow-up, chest radiograph did not show evidence of mediastinal lym-

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phadenopathy or collapse (Figure 1B) and total IgE were reduced significantly (2554 kU/L) after 6 weeks of therapy.

Discussion

Allergic bronchopulmonary aspergillosis (ABPA) is a complex immunological pulmonary disorder caused by hypersensitivity to *Aspergillus* species with resultant systemic immune activation, chronic asthma, recurrent pulmonary infiltrates, and bronchiectasis, and it often complicates bronchial asthma and cystic fibrosis. *Aspergillus fumigatus* is the major causative fungus but *A.flavus, A. niger,* and *A. oryzae* can also cause ABPA. Patients of ABPA usually present with poorly controlled asthma, wheezing, hemoptysis, and productive cough. Common CT findings include bronchiectasis, mucoid impaction, mosaic attenuation, centrilobular nodules, and tree-in-bud opacities [3]. ABPA does present with varied clinical and radiological features and several different presentations of ABPA have been reported [4]. Our case presented

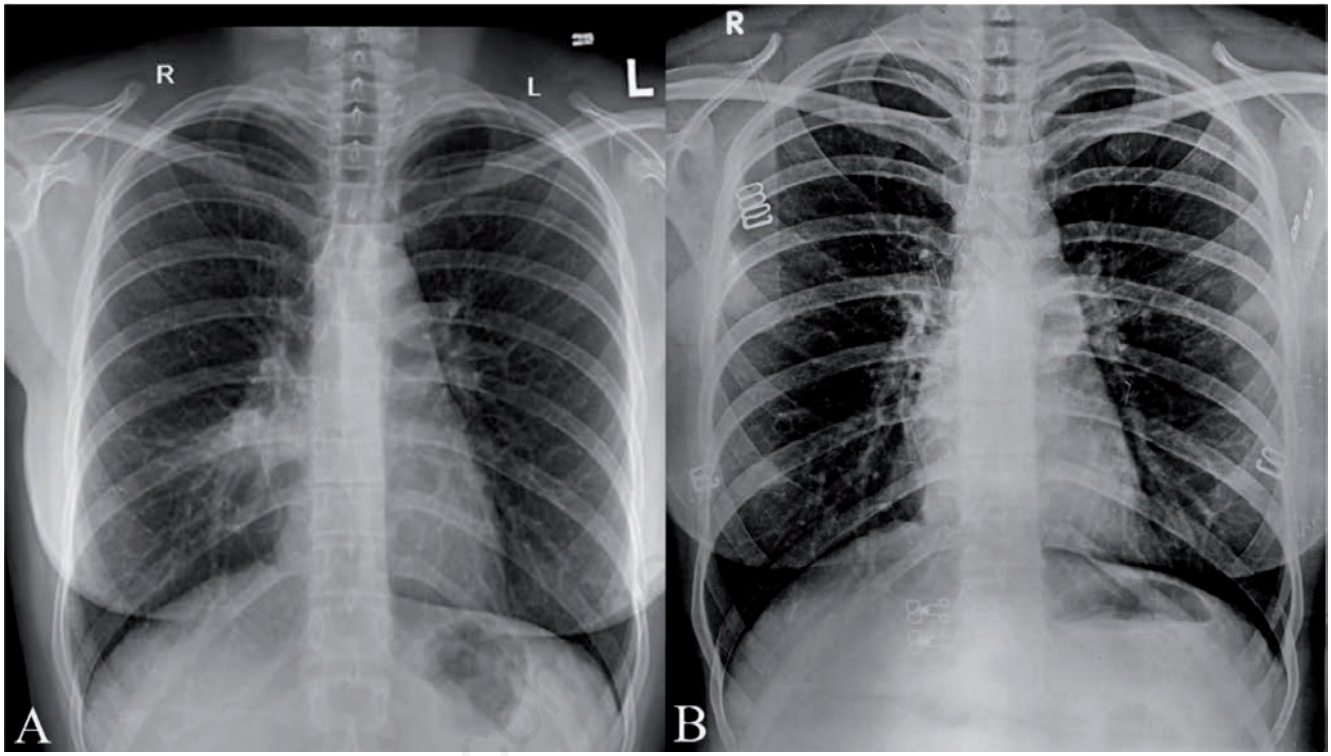

Figure 1. Patient's chest radiograph at the time of presentation (A), and after 6 weeks of treatment with 0.75 mg/kg prednisolone and itraconazole 200 mg BD (B).

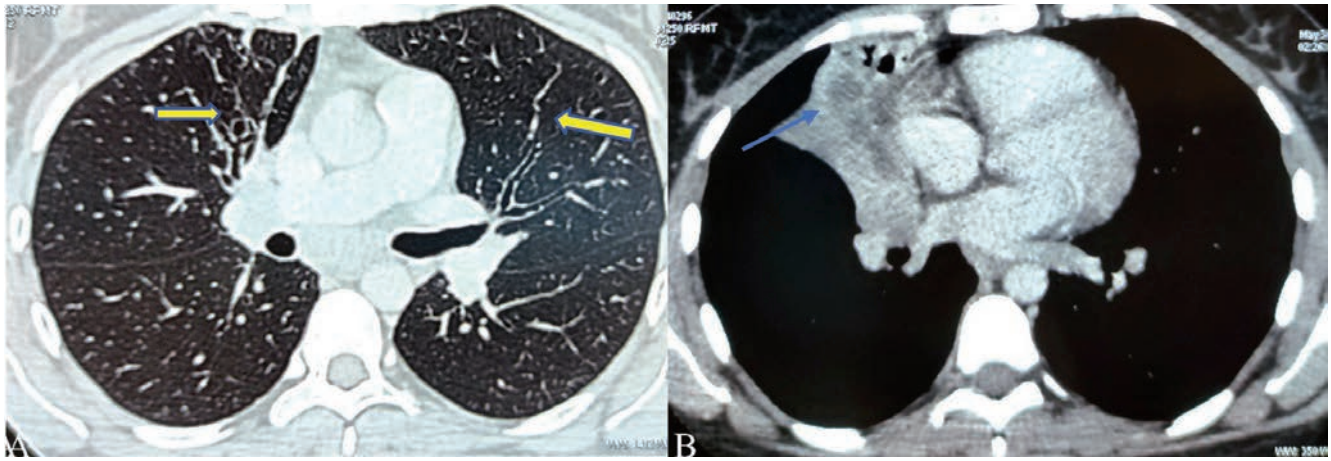

Figure 2. Patient's CT chest showing (A) central bronchiectasis (yellow arrows) and (B) middle lobe atelectasis (blue arrow).

with findings of middle lobe collapse and mediastinal lymphadenopathy, both of which are not typical radiological features of ABPA. Most probable differential diagnosis of mediastinal lymphadenopathy are pulmonary tuberculosis and sarcoidosis. Lobar collapse which is rare in sarcoidosis and common in tuberculosis, has been reported in several cases of ABPA [5]. There can be "pseudo" hilar adenopathy in ABPA which is due to the formation of bronchocele in central bronchi, but "true" mediastinal lymphadenopathy is a rare finding in ABPA [6]. Only 4 cases of mediastinal lymphadenopathy due to ABPA have been reported so far [7-10]. Hantsch et al described case of a 27-year-old male with history of asthma who presented with symmetrical hilar lymphadenopathy on X-ray. The patient underwent mediastinoscopy, bronchoscopy and open lung biopsy for diagnosis of lymphadenopathy. Open lung biopsy and BAL showed growth of *Aspergillus fumigatus*, and serological profile confirmed the diagnosis of ABPA [7]. A case of 21-year-old male with bronchial asthma was reported by Hachiya *et al.* in 1998. Patient's chest radiograph was suggestive of right upper lobe atelectasis and left hilar nodular opacity. Contrast CT was suggestive of central bronchiectasis and paratracheal and hilar lymphadenopathy. Patient was diagnosed as ABPA according to Rosenberg's criteria [11]. Patient was treated with prednisolone which resulted in resolution of atelectasis, lymphadenopathy and infiltrates [8]. Shah *et al.* in 1999 reported case of a 20year old male who was diagnosed as pulmonary tuberculosis on radiological basis and was on antitubercular treatment (ATT) at the time of presentation. Serial chest radiographs demonstrated transient pulmonary infiltrates and right hilar prominence. CT Chest showed hilar lymphadenopathy along with central bronchiectasis and patchy infiltrates. Instead of invasive investigations, patient was diagnosed as case of ABPA on basis of clinicoradiological and immunological criteria, and was started on oral prednisolone and ATT was stopped [9]. A similar case of a 42-months-old child was reported by Shah *et al.* in 2007 who was on ATT for 6 months. The patient had history of wheezing, cough and dyspnoea for 1 year. CT showed pretracheal and right hilar lymphadenopathy with central bronchiectasis. Patient fulfilled immunological and clinicoradiological criteria for ABPA and was treated with prednisolone [10].

ABPA is often misdiagnosed as tuberculosis in India and such a scenario was seen in above two cases [12]. Also, in our case, the first differential diagnosis was tuberculosis but after correlating clinical, radiological, and serological findings, a final diagnosis of ABPA was made. According to newer diagnostic criteria proposed by Agarwal *et al.* [2]. patient had predisposing condition (i.e. bronchial asthma) and fulfilled obligatory criteria (elevated total IgE and elevated *Aspergilllus* specific IgE) as well as other criteria (i.e., raised IgG against *A. fumigatus*, increased total eosinophil count, bronchiectasis and mucus plugging on CT).

Conclusions

In a background of bronchial asthma with radiology showing mediastinal lymphadenopathy and collapse, even in high tuberculosis burden countries, ABPA should be kept as a differential diagnosis. ABPA should not be ruled out as a diagnosis solely on basis of the radiological picture as it can vary from patient to patient.

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