

## Case Report

# Unusual cause of endobronchial mass lesion in immunocompetent host

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### Abstract:

We report case of a 39 year old immunocompetent gentleman, who presented with complaints of cough and hemoptysis for 2 months. Chest X ray and CT chest revealed left lower lobe collapse. Bronchoscopic examination showed an endobronchial mass lesion, occluding the left lower lobe bronchus. A diagnosis of "Obstructive tracheobronchial aspergillosis with invasion" was made as a result of bronchoscopic findings, histopathological and microbiological examination. The patient was treated with oral Voriconazole. Repeat bronchoscopy performed after 3 weeks of treatment showed complete resolution of the previously noted endobronchial lesion. We describe this case to consider role of antifungal treatment in an immunocompetent patient and discuss Tracheobronchial aspergillosis (TBA) which responded dramatically to treatment.

**Key-words :** Aspergillosis, Immunocompetent, Bronchial disease, Voriconazole

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### Introduction :

Tracheobronchial aspergillosis (TBA) is an uncommon clinical form of invasive aspergillus infection, which is predominantly confined to the tracheobronchial tree. TBA is commonly reported in immuno compromised hosts. The importance of early diagnosis and initiation of antifungal therapy in the immunocompromised host is well known. As TBA is rare in immunocompetent hosts, the role of antifungal therapy has not been defined in these hosts. We report a rare case of "Obstructive TBA with invasion" in an immunocompetent host, which responded very well to antifungal treatment.

### Case History :

A 39 year old gentleman, resident of West Bengal, nondiabetic and normotensive, complained of dry

cough for two months, associated with recurrent streaky hemoptysis. He had no fever, chest pain, weight loss or loss of appetite. He was treated by local physician with cough syrups and antibiotics but his symptoms did not subside. Later he had moderate hemoptysis, for which he was evaluated at a local hospital and was referred to our hospital for further management. There was no significant past history. The patient had no chronic disease and was not on any immunosuppressant therapy. He denied any addictions including smoking.

On examination, he was comfortable at rest and was hemodynamically stable. He was afebrile and had no lymphadenopathy. Respiratory system examination revealed dull percussion note and decreased air entry at left lung base. Physical examinations of other systems were within normal limits. Basic hemogram showed

haemoglobin of 13.7 gm%, WBC count of 7,600/cumm, eosinophils 3%, platelet count of 2.5 lakhs/cumm, ESR of 46mm/hr and normal coagulation profile. Liver and renal function tests were within normal limits. Enzyme Linked Immunosorbent assay of human immunodeficiency virus antibody was negative. Chest X ray showed 'Sail sign' suggestive of left lower lobe collapse (Figure 1).

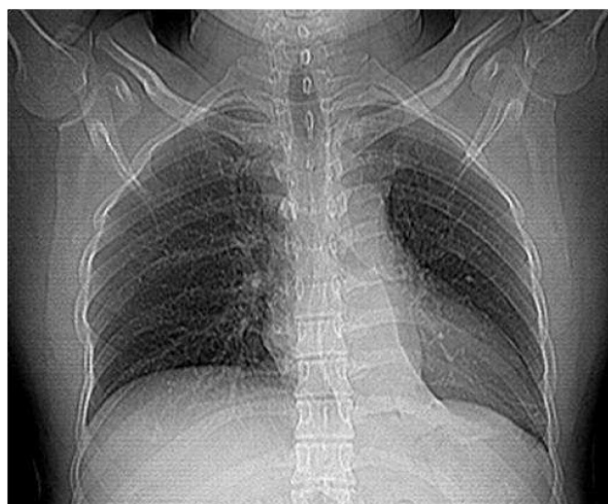


Fig. 1

CT scan thorax revealed left lower lobe collapse with possibility of endobronchial lesion. We subjected the patient to bronchoscopy and found an endobronchial whitish mass occluding left lower lobe bronchus (Figure 2).



Fig. 2

Narrow band imaging(NBI) showed abnormal vascularity of the lesion. Multiple biopsies were taken from endobronchial mass lesion. Bronchial wash was negative

for AFB and gram staining but fungal staining showed many septate hyaline hyphae. Bronchial wash cytology showed inflammation and fungal hyphae consistent with *Aspergillus*. Histopathology of the endobronchial biopsy showed necrotic tissue with hyphal fungal organism consistent with *aspergillus* species (Figure 3).

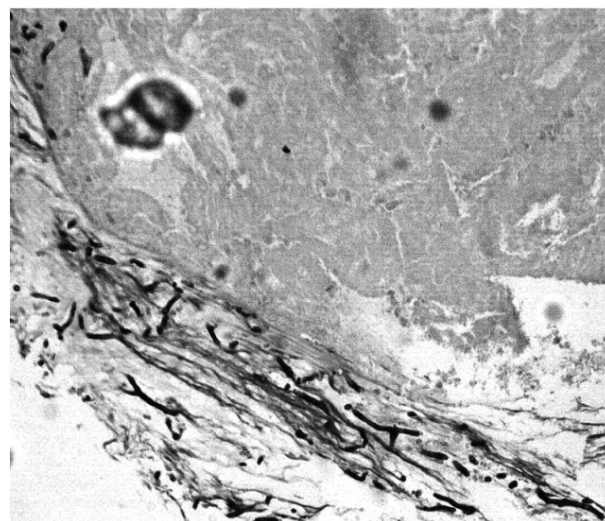


Fig. 3

Bronchial wash grew *aspergillus flavus*. Bronchial wash AFB culture was negative. Serum galactomannan was positive (Optical Density Index -2). In view of endobronchial growth causing left lower lobe collapse, biopsy showing fungal hyphae with evidence of tissue damage, growth of *aspergillus flavus* and positive serum galactomannan; diagnosis of "Obstructive TBA with invasion" was made. The patient was started on oral

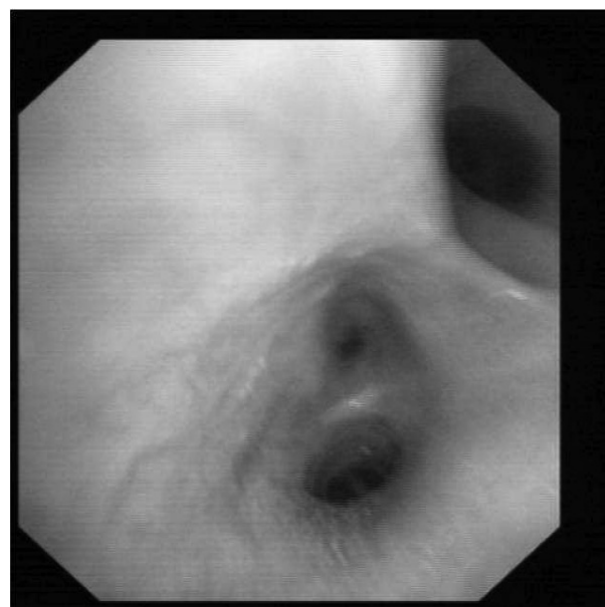


Fig. 4

Voriconazole 400mg twice a day on day one followed by 200mg twice daily. The patient had significant symptomatic improvement and a repeat Chest X ray showed expansion of left lower lobe. The patient underwent bronchoscopy after 3 weeks of treatment, which showed clear left lower lobe bronchus with complete resolution of the endobronchial lesion (Figure 4).

## Discussion :

*Aspergillus* species are responsible for a broad spectrum of lung diseases, depending upon host fungus relationship. The spectrum of pulmonary involvement of aspergillosis ranges from aspergilloma, allergic bronchopulmonary aspergillosis, chronic necrotizing pulmonary aspergillosis and invasive aspergillosis<sup>1-4</sup>. Invasive pulmonary aspergillosis (IPA) is a severe disease that is usually reported in immunocompromised patients like those with hematological malignancy. TBA is an uncommon form of aspergillus infection which is predominantly confined to the tracheobronchial tree. It occurs in less than 7% cases of pulmonary aspergillosis<sup>5</sup>. Invasive forms of TBA had not been fully described until 1991 by Kramer et al, who described it in patients after lung transplantation<sup>6</sup>. Obstructive TBA was first described by Denning et al, in three patients with AIDS<sup>7</sup>. After that cases of TBA in immunocompetent hosts have also been reported<sup>8-14</sup>. Several classifications of TBA were proposed since 1991. In 2010, Wu et al<sup>12</sup> performed a retrospective study of 19 patients and classified isolated invasive TBA into four different types based on the morphological features of intraluminal lesions. Type 1 was with plaques of pseudomembrane formation (superficial infiltration type), type 2 was with deep ulceration of bronchial wall (ulcerative type), type 3 was with significant (>50%) airway occlusion (obstructive type) and type 4 was pertaining to patients with two or more characteristic lesions (mixed type). Our patient has endobronchial growth with airway occlusion and can be obstructive type.

Patients with TBA can be asymptomatic. Most common presenting complaints are cough, fever, dyspnea, chest pain and hemoptysis<sup>5,15</sup>. Our patient presented with cough and hemoptysis. The diagnosis of TBA is typically delayed because of its insidious onset, non specific symptoms and lack of specific radiological abnormalities. Chest X ray may show nonspecific findings such as tree in bud appearance, bilateral consolidation, collapse or it may even be normal. Obstructing bronchial aspergillosis was reported to present with middle lobe

collapse<sup>16</sup>. Our patient presented with left lower lobe collapse.

TBA is classified as invasive aspergillosis, which is diagnosed according to the European Organization for Research and Treatment of Cancer/Mycosis Study Group definition, based on culture of *aspergillus* species with evidence of tissue invasion on histopathology<sup>17</sup>. Our patient's bronchial wash grew *aspergillus flavus* and histopathology of bronchial biopsy showed *aspergillus* species with evidence of tissue damage (necrosis). In our patient serum galactomannan was also positive. Prognosis of TBA in immunocompetent hosts is not well known due to its rarity but prognosis in immunocompromised hosts is poor with mortality rate upto 70%<sup>18</sup>. Spontaneous regression of TBA without antifungal agents in immunocompetent host is reported in literature, but this patient was having superficial bronchial involvement (Type 1)<sup>19</sup>. Although obstructive pulmonary aspergillosis is regarded as a saprophytic form, it can progress to invasive pulmonary aspergillosis and can have a fatal outcome<sup>7, 16, 18, 20</sup>.

Infectious Disease Society of America has recommended voriconazole as initial therapy in management guidelines for invasive pulmonary aspergillosis including TBA in 2008<sup>21</sup>. For seriously ill patients, the recommended dosing regimen of voriconazole is 6 mg/kg IV twice a day on day one followed by 4 mg/kg IV twice daily. Oral voriconazole is started once the patient is stable and can tolerate oral medications. Our patient was hemodynamically stable, so we started him on oral voriconazole 400 mg twice a day on day one followed by 200 mg twice daily. The duration of antifungal therapy for invasive pulmonary aspergillosis including TBA in immunocompetent patient is not well defined. Duration of therapy depends upon the location of infection, the patient's underlying disease and response to therapy. It is recommended that treatment of invasive pulmonary aspergillosis including TBA in immunocompetent patient should be continued for a minimum of 6-12 weeks<sup>21</sup>. Even though, our patient showed dramatic improvement in 3 weeks, after endobronchial debulking by multiple biopsies and antifungal therapy, his antifungal treatment was continued for total period of 6 weeks.

In conclusion, we suggest that all endobronchial masses are not malignant. Obstructive TBA should be considered as differential diagnosis in a patient presenting with obstructive collapse, even in the immuno

competent patient. Key factors for a favorable outcome in immunocompetent patients are early diagnosis and adequate antifungal therapy.

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