Case Report

Pleural *Aspergillosis* in an otherwise healthy individual

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ABSTRACT

Pleural *Aspergillosis* is a rare entity, with most of the cases occurring on a background of lung disease or surgery. We report a case of a 16-year-old boy who developed pleural *Aspergillosis* in the absence of any obvious pre-disposing factors. Patient presented with fever, dry cough and left sided chest discomfort of 6 weeks duration. A chest radiograph revealed features of the left pyopneumothorax. Despite being started on presumptive antituberculous treatment and intercostal drainage his symptoms failed to resolve. The sputum and pus were then subjected to fungal smears and culture, which indicated growth of *Aspergillus fumigatus*. We report this case in view of the extreme rarity of pleural *Aspergillosis* occurring in a young healthy individual with no evidence of prior lung pathology. Furthermore, the source of infection was traced back to a very unusual possible focus – a decayed tooth infected with *A. fumigatus*, thus making the case even more interesting.

KEY WORDS: *Aspergillosis*, caries, hydropneumothorax, immunocompetent, pleural

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INTRODUCTION

Lung infection with *Aspergillus* manifests in a variety of patterns, the most important of which are allergic bronchopulmonary *Aspergillosis*, chronic necrotizing *Aspergillus* pneumonia, *Aspergilloma* and invasive *Aspergillosis*.[1,2] The most common isolated species are *Aspergillus fumigatus* and *Aspergillus flavus*.

Pleural *Aspergillosis* is a rarely reported disease. Overall, fungal infections accounts for less than 1% of all pleural effusions.[3] However, it is associated with a high risk of mortality, mostly due to late or missed diagnosis or lack of safe effective treatment.[4] Unlike the pulmonary forms of the disease, pleural *Aspergillosis* is not more common in immunocompromized individuals. Pulmonary *Aspergillosis* is also not seen to lead to pleural infection usually. Majority of the reported cases of pleural *Aspergillosis* have been in the presence of some underlying lung pathology or surgical procedure. The most common preceding conditions are pre-existing tuberculosis (87%), bronchopulmonary fistulas (74%), pleural drainage (56%) and lung resection (17%).[5] Here, we report pleural *Aspergillosis* occurring in a young healthy male, with no prior evidence of lung damage. An extensive PubMed search revealed no similar case report in the English literature.

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CASE REPORT

A 16-year-old Indian boy presented to the out-patient department of our hospital with complaints of low-grade fever, dry cough and left sided chest discomfort of 6 weeks duration. Cough was non-productive, non-spasmodic without postural or diurnal variation. The chest pain was worse on coughing. There was no history of similar complaints in the past. He had no history of hemoptysis, loss of appetite or weight loss. There was no history of bronchial asthma, diabetes mellitus, previous hospitalization or surgical intervention. On examination, patient was of fair nutritional status and his vitals were stable. General physical examination did not reveal any significant abnormality. On chest examination, left sided movements were decreased, percussion note was stony dull and air entry was absent. Succusion splash was also heard. On investigating, his routine hematological and biochemical parameters were normal (hemoglobin – 11 g/dl, total leucocyte count – 7600/cc, differential leucocyte count – P52 L40E8, blood sugar (fasting) – 86 mg/dl, blood

sugar (post-prandial) – 148 mg/dl, T. bilirubin-0.4 mg/dl, aspartate aminotransferase - 26 IU/ml, alanine aminotrans ferase 38 IU/ml, blood.urea-26 mg/dl, S. creatinine-1.0 mg/dl). Mantoux test and serology for human immunodeficiency virus were negative. Immunoglobulin E was significantly elevated (1602 IU/ml). A chest radiograph was done, which had features of left pyopneumothorax. Air fluid level was present on the radiograph [Figure 1]. Diagnostic intercostal drainage revealed frank brownish pus and tube thoracostomy was performed draining 1000 ml pus on the 1st day itself. The pus and induced sputum stained negative for pyogenic organisms and acid fast bacilli (AFB). Considering the fact that the patient belonged to a high incidence area for tuberculosis, a therapeutic trial of anti-tuberculosis treatment was started. However, at the end of 2 months of treatment, his induced sputum and pleural fluid again stained negative for AFB. Culture of the sputum sample was also negative for AFB. Despite treatment and drainage with a broad intercostal drainage tube (ICD) his symptoms failed to resolve. The sputum and pus were then subjected to fungal smears and culture. The sputum smears were negative, but pus smears showed septate fungal hyphae and grew *A. fumigatus* in culture [Figure 2]. Smears from the culture showed septate hyphae and conidiophores confirming pathogenic role of the mould. Flexible bronchoscopy was done and bronchial wash was subjected to microbiological studies. Both fungal smear and culture were negative, indicating lack of pulmonary involvement. Patient was thus diagnosed as pleural *Aspergillosis* in the absence of invasive pulmonary *Aspergillosis* or any other obvious predisposing factors. He was started on voriconazole 300 mg twice-a-day. Within 4 weeks, the patient showed signs of recovery with reduction in cough and chest pain [Figure 3]. The pyopneumothorax showed signs of resolution and the ICD was removed after 6 weeks of starting treatment. Patient was maintained on oral voriconazole for 9 months and showed no signs of relapse 1 year later.

We conducted a thorough search for the source of the *A. fumigatus* infection in our patient and found that the boy had a carious tooth. On culture in blood agar, *Streptomyces spp*. was grown. In addition, the carious material was cultured in Sabouraud's Dextrose Agar medium and it grew *A. fumigatus*. We thus hypothesize that at some point in time the patient could possibly have aspirated the carious material, which would have infected the pleura subsequently without direct invasion of lung parenchyma. We thus, report this case in view of the extreme rarity of pleural *Aspergillosis* in patients with otherwise healthy lungs.

DISCUSSION

Pleural *Aspergillosis* was first described by Cleland in 1924.[6] It is a rare entity and usually occurs in the presence of disruption of local pulmonary defence mechanisms, most often due to tuberculosis. Krakowka *et al*. found that an established empyema and a bronchopleural or

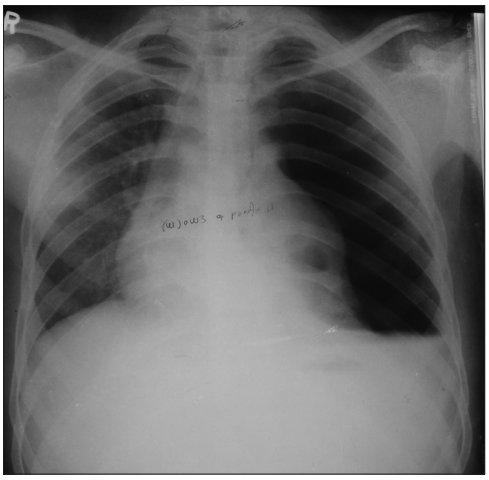

Figure 1: Chest radiograph anteroposterior view showing left pyopneumothorax with collapsed left lung

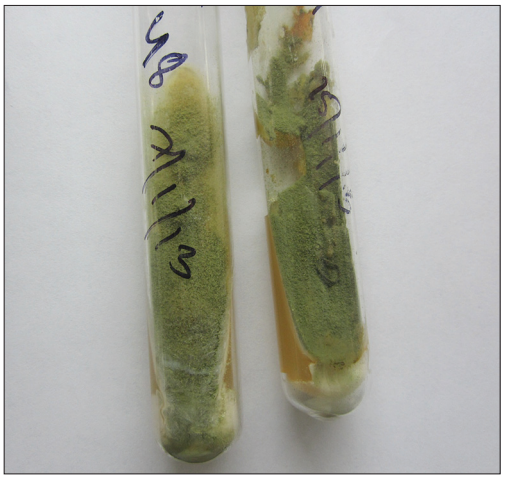

Figure 2: Photograph of culture of the pus in Sabouraud's Dextrose Agar in duplicate at 25°C and 37°C. Both test tubes showed luxurious bluish-green growth with a suede-like surface diagnostic of *Aspergillus fumigatus*

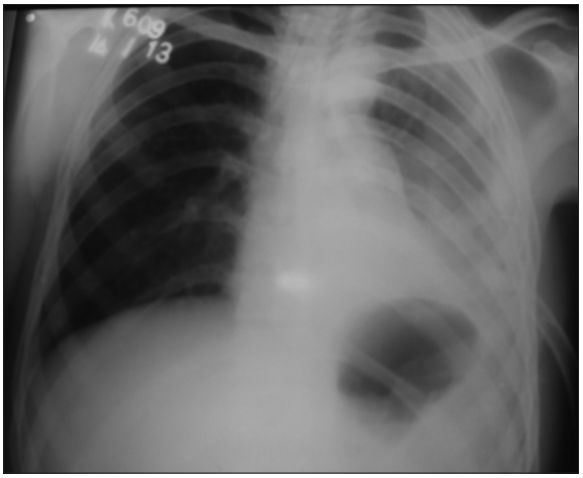

Figure 3: Chest radiograph taken 1 month after treatment initiation showing well-expanded left lung with intercostal tube *in situ*

pleurocutaneous fistula were the two clinical settings most commonly associated with pleural *Aspergillosis*.[7] In our case, infection of the pleura occurred in the absence of any lung pathology in an otherwise healthy individual. The growth of *A. fumigatus* in the culture of the carious material from the tooth is a useful clue to the origin of the infection and the sequence of events, which could have followed. We hypothesize that micro aspiration of the fungal elements led to direct seeding of the pleura (as no lung pathology was seen on the chest radiograph or the bronchial wash microscopy). An extensive PubMed search revealed no case in English literature, which did not have evidence of previous injury to the pleura. Karthik and Sudarsanam reported a case of pleural *Aspergillosis* caused by *Aspergillus terreus* in an Indian patient who was an old case of pulmonary tuberculosis.[2] They treated the patient with intercostal drainage and amphotericin-B (AMB) (1 mg/kg for a total of 1.5 g). However, the patient did not significantly improve and subsequently underwent a left lung decortication surgery.

Many treatment modalities have been tried including antifungals and surgical procedures. Early administration of antifungal agents and pleural drainage are helpful in improving the outcome of patients.[4] AMB has been the mainstay conventionally, but the major limitation has been the toxicity associated with its administration (nephrotoxicity, phlebitis, hypokalemia, hypomagnesemia, anemia) in nearly 80% of patients.[8] Liposomal AMB is seen to be safer. Voriconazole is a broad spectrum triazole that is active against all species of *Aspergillus*. It is better tolerated than AMB, with fewer drug related adverse events.[8] Stern *et al*. used voriconazole 200 mg twice-a-day for 2 weeks in a 70 kg patient of pleural *Aspergillosis*, which was later increased to 300 mg twice-a-day. They demonstrated that voriconazole penetrated pleural fluid and sterilized the pleural cavity well wherein, their patient was completely cured after 6 months.[9]

Hence, clinical suspicion and early detection of fungal cause of a non-responsive empyema may improve the clinical outcome of such patients. We report this case in view of the extreme rarity of its occurrence in previously normal lungs.

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