Case Report

Solitary pulmonary nodule: A rare presentation of pulmonary mucormycosis in an immunocompetent adult

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ABSTRACT

Pulmonary mucormycosis is a rare opportunistic infection of immunocompromised individuals. Here, we report a case of 70-year-old male, smoker presenting with high-grade fever for 2 weeks and episodes of hemoptysis. Contrast-enhanced computed tomography (CT) thorax revealed a solitary pulmonary nodule measuring 2.3 × 1.6 cm in the right upper lobe. CT guided fine needle aspiration cytology and true cut biopsy showed plenty of typical fungal hyphae consistent with the diagnosis of mucormycosis. Fungal culture confirmed the organism as mucor. Positron emission tomography-CT scan showed a non- 18 fluorodeoxy glucose avid nodule ruling out possibility of malignancy. Investigation did not reveal any evidence of immunosuppression. Patient was treated with intravenous liposomal amphotericin B for 4 weeks. Follow-up chest X-ray and CT scan after 6 weeks were normal.

KEY WORDS: Immunocompetent host, mucormycosis, pulmonary, solitary pulmonary nodule

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INTRODUCTION

Pulmonary mucormycosis is relatively uncommon but life-threatening infection affecting mostly individuals such as diabetes mellitus, hematological malignancies, chronic renal failure, posttransplantation, and other immunocompromised states. Pulmonary mucormycosis first came into light by Furbinger *et al*., in 1876.[1] Mucormycosis of lung has a wide range of clinical and radiological manifestations. Few cases of pulmonary mucormycosis, presenting as fungal ball, cavity resembling tuberculosis, nonresolving, and recurrent pneumonias in patients with diabetes mellitus or other immunosuppressive conditions, were reported from India.[2,3] To the best of our knowledge, pulmonary mucormycosis presenting as solitary pulmonary nodule in an immunocompetent individual was not reported from India.

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CASE REPORT

A 70-year-old male, smoker for 4 decades presented with high-grade fever for 2 weeks and episodes of hemoptysis for 5 days. He was admitted to our institution with massive hemoptysis. General survey revealed temperature was 102°F; pulse rate was 104/min, regular; respiratory rate was 28/min and blood pressure was 130/76 mm of Hg. Systemic examination including respiratory system was essentially normal. Routine blood and biochemistry showed hemoglobin was 8.7 gm/dL, total white blood cell count was 4600/mm^3 with neutrophil comprising 77%, platelet count was 1,75,000/cmm, fasting sugar was 76 mg/dL, serum levels of urea was 28 mg/dL, and creatinine was 0.9 mg/dL. Blood for HIV I and II was negative, and there was no evidence of immunosuppression. His chest X-ray posterior-anterior view showed a nodular opacity in right upper zone. His hemoptysis was controlled with conservative management but fever persisted and we went for further investigation. Contrast-enhanced computed tomography (CT) thorax revealed an irregular, well-demarcated nodule (32 H.U) measuring 2.3 × 1.6 cm seen at anterior segment of right upper lobe with few areas of rarefaction within the nodule [Figure 1]. We suspected the nodule to be malignant and went for CT-guided fine needle aspiration cytology (FNAC) and true cut biopsy. CT-guided FNAC showed plenty of fungal hyphae in a necroinflammatory background with absence of granuloma

or malignancy. Giemsa-stained smear showed broad, irregular, nonseptate hyphe with right-angled branching consistent with the diagnosis of mucormycosis [Figure 2a]. Gomori's methenamine silver staining of CT-guided true cut biopsy also demonstrated the typical hyphae suggestive of mucormycosis. Hematoxylin and eosin staining of the true cut biopsy material showed wide areas of necrosis and inflammatory cell infiltration [Figure 2b].

Fungal culture of the specimen inoculated on Saboraud's Dextrose Agar medium yielded white colonies within 3 days and organism was identified as mucor. Fibreoptic bronchoscopy revealed bleeding coming out of right upper lobe bronchus along with congested mucosa. Bronchoalveolar lavage showed absence of malignant cells along with no growth of mycobacterium and fungus. Positron emission tomography-CT scan showed a non-FDG avid nodule with spiculated margin and calcification in anterior segment of right upper lobe [Figure 3]. The diagnosis of pulmonary mucormycosis was confirmed. As patient refused surgery, we started treatment with intravenous liposomal amphotericin B at a dose of 5 mg/Kg for a period of 4 weeks. Fever subsided after 4 days of starting liposomal amphotericin B and follow up chest X-ray and CT scan after 6 weeks were normal [Figure 4].

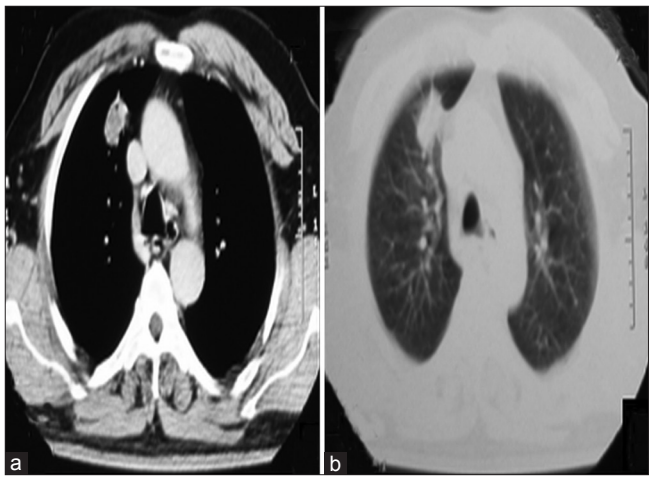

Figure 1: (a and b) Contrast-enhanced computed tomography thorax showing a solitary pulmonary nodule measuring 2.3 × 1.6 cm with few areas of rarefaction seen at anterior segment of right upper lobe

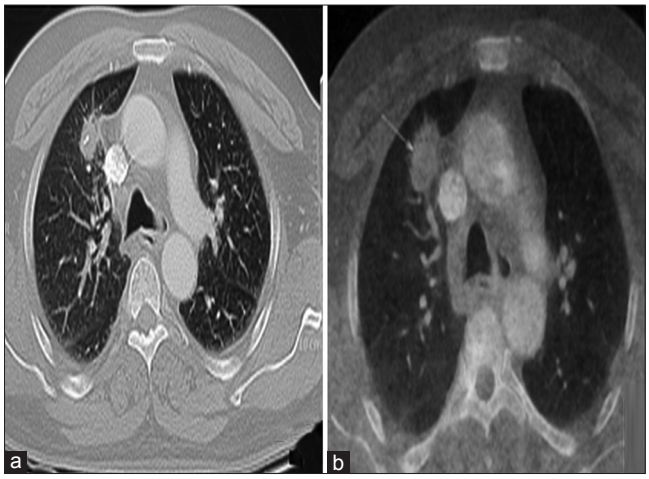

Figure 3: Positron emission tomography-computed tomography scan showing (a) Nodule with spiculated margin and calcification, (b) Non-FDG avid nodule seen in anterior segment of right upper lobe

DISCUSSION

Mucormycosis is defined as infection caused by fungi of the order Mucorales belonging to the class Zygomycetes. Rhizopus is the most commonly identified genus among Zygomycetes followed by Mucor and Cunninghamella.[4] Mucormycosis is a life-threatening fungal infection that mostly affects immunocompromised adults. Mucormycosis is caused predominantly by inhalation of spores caused by inhalation of spores with lung and paranasal sinuses being the most commonly affected organs.[5] Pulmonary mucormycosis is a disease predominantly affecting males with M:F ratio ranging from 2.4:1-3:1.[6] Predisposing individuals are those with diabetes mellitus,

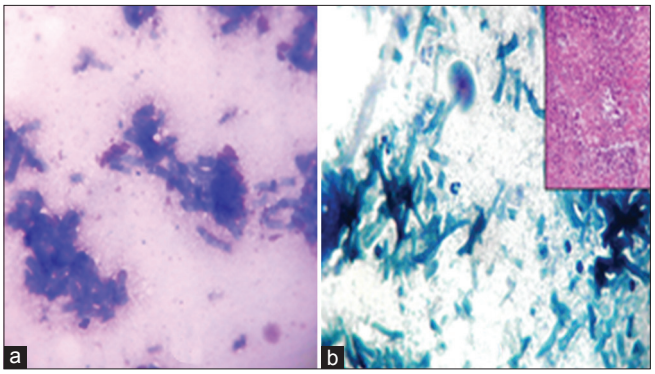

Figure 2: (a) Giemsa-stained smear, (b) Methenamine silver-stained smear showing broad, irregular, and nonseptate hyphae of mucormycosis. H and E staining of the biopsy material showing wide areas of necrosis and inflammatory cell infiltration (inset)

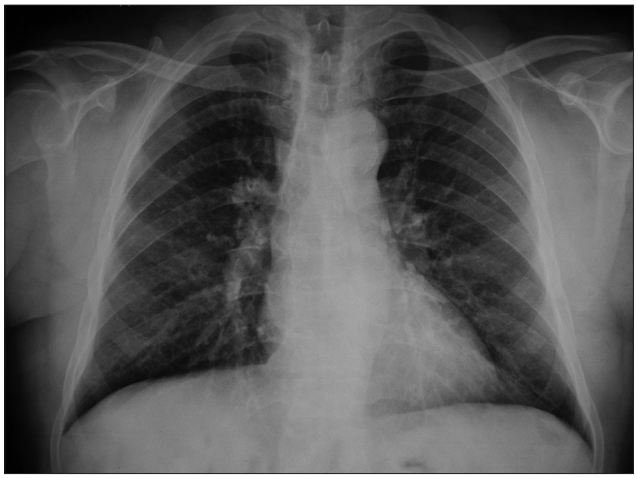

Figure 4: Follow-up chest X-ray done after 6 weeks showing clearance of the lesion

hematological malignancy, renal insufficiency, and solid organ transplantation.[7] Lee *et al.*,[6] in their study found only 11 out of the 87 patients had no underlying condition. Pulmonary mucormycosis has an acute onset and a wide range of clinical manifestations, such as fever (most common), cough, hemoptysis, dyspnea. Haemoptysis was found in about 25%-30% of the patients.[6] Radiological manifestations include infiltrates, consolidation, cavitation, focal masses, or nodules.[8] Reverse halo sign (RHS) is a diagnostic radiological clue toward diagnosis of mucormycosis. It is usually found in invasive mucormycosis in immunocompromised individuals either in the early state or during immune recovery phase. Histological examination of the pulmonary lesion of the patient with invasive pulmonary mucormycosis presented RHS, consolidation area was comprised of triplet structure; liquefaction, consolidation, and organization.[8] All these features are not found unless surgical excision is performed contrary to the true cut biopsy performed here. As mucormycosis is an angioinvasive organism, it has a tendency to grow toward blood vessels, and a mass or nodule encroaching toward the great vessels of mediastinum in successive radiographs suggests the possibility of mucormycosis. The typical bronchoscopic findings are bronchial stenosis or obstruction, erythematous mucosa, gelatinous or mucoid secretions, fungating or polypoid masses, and mucosal ulceration.[6] Transthoracic needle aspiration as a diagnostic tool of mucormycosis was used only in a small minority of patients (10%-20%).[6] Fungal culture of the specimens was found to be positive in half of the patients making it as a relatively insensitive tool. Up to 32% of patients presenting with zygomycosis have been observed to have a concurrent infection that is usually bacterial in origin.[6] Combined surgical and medical treatment of zygomycosis has a reported mortality of about 30%, compared with medical treatment alone which has a mortality of 55%.[3,6] Furthermore, treatment with liposomal amphotericin B was associated with a 67% survival rate compared with 39% survival with amphotericin B ($P = 0.02$) among patients with cancer who experienced mucormycosis.[9] Treatment of zygomycosis consists of the prompt administration of amphotericin treatment, reversal of the underlying condition preferentially combined with surgical resection of the necrotic tissue.[10]

In our case, we confirmed the diagnosis by CT-guided FNAC and true cut biopsy, an uncommon diagnostic tool, and by demonstrating the typical fungal hyphae and detecting the colonies of mucor by culture.

Our case had a unique presentation as solitary pulmonary nodule, not reported from India. Our patient was immunocompetent and that is contradictory to the common belief that mucormycosis is an opportunistic infection in immunosuppressed host. Moreover, no case of mucormycosis in immunocompetent adult has been reported in India. Although combined medical and surgical approach is recommended, our patient was treated successfully with medical treatment alone.

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