

Rare Case of Disseminated Mucormycosis Presenting with Bilateral Pulmonary Artery Aneurysm

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ABSTRACT

Pulmonary artery aneurysm is an infrequent entity and is generally congenital in origin or secondary to pulmonary arterial hypertension. Infections causing pulmonary artery aneurysm are limited, in which tuberculosis and bacterial infections being the shared causative etiologies. There have been only scarce cases reported in the past, in which mucor caused pulmonary artery aneurysm. Pulmonary mucormycosis causing pulmonary artery aneurysm is an infrequent and almost fatal complication, as most of the diagnoses are made postmortem. Our case is the first case report in Indian literature that brings out a case of disseminated mucormycosis, causing bilateral segmental pulmonary artery aneurysm in a patient with uncontrolled diabetes. This patient was cured by timely treatment with antifungals.

KEYWORDS: Diabetic ketoacidosis, pulmonary artery aneurysm, pulmonary mucormycosis

INTRODUCTION

Pulmonary mucormycosis is a comparatively uncommon but imperative opportunistic fungal infection in immunocompromised hosts. The key risk factors are diabetes mellitus, hematologic malignancies, and organ transplantation. This relatively infrequent but often deadly disease should be considered in immunocompromised hosts who do not respond to antibacterial therapy. Mycotic pulmonary artery aneurysms are infrequent causes of acquired pulmonary artery aneurysm. These are mostly caused by tuberculosis, bacteria such as streptococcus and staphylococcus, syphilis, and only occasionally by fungus-like mucormycosis.^[1,2] There are only limited published cases where pulmonary mucormycosis was the causative organism of pulmonary artery aneurysm, with very few survivals as the antemortem diagnosis is difficult.^[3] Optimal therapy requires systemic antifungals, surgical resection, and control of the underlying disease. We report the clinicopathological features of a case of disseminated mucormycosis who presented with pulmonary mucormycosis along with

bilateral segmental pulmonary artery aneurysm and renal abscess.

CASE REPORT

A 60-year-old male, known case of uncontrolled Type 2 diabetes mellitus presented with complaints of intermittent fever, productive cough, and progressive breathlessness on exertion of 20 days duration and a single episode of hemoptysis of volume approximately 100 ml. Clinically, he had tachycardia, tachypnea, fever, and respiratory system examination revealed bronchial breath sounds in the right infraclavicular and mammary regions.

On evaluation, hemogram showed normocytic normochromic anemia, hyperglycemia (random blood glucose 426 mg/dl), arterial blood gas showed metabolic acidosis, urinalysis was positive for ketones, and sputum

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for Gram stain/Ziehl-Neelsen-stain/fungal stain: No organisms/fungal elements seen, Sputum for Gene Xpert: MTB not detected, HIV/HBsAg negative. Vasculitis workup was negative. Chest radiograph [Figure 1] revealed alveolar opacities in the right upper, middle, and lower zone. Computed tomography pulmonary angiogram [Figure 2] revealed large cavitating consolidation in both lower lobes and right upper lobe with aneurysmal dilatation of the pulmonary artery and hypodense filling defect in segmental pulmonary arterial branches of the right upper lobe. The patient was managed as a case of diabetic ketoacidosis and pulmonary thromboembolism with insulin infusion, intravenous fluids, and enoxaparin under cover of empirical broad-spectrum antibiotics. However, the general condition of the patient kept on deteriorating. The patient underwent fiberoptic bronchoscopy, which showed endobronchial nodularity in the left lower lobe bronchus, and endobronchial biopsy was taken from the left lower lobe bronchus. Endobronchial biopsy [Figure 3] showed histomorphologic features consistent with mucormycosis. The patient was immediately commenced on liposomal amphotericin along with culture-guided antibiotics. The patient continued to remain febrile after 2 weeks and repeat imaging showed an increase in the area of consolidation with areas of breakdown in the bilateral upper lobes, left lower lobe, pulmonary artery aneurysm in the apical segment of the left lower lobe, and right lateral basal segment and enlarged left kidney with large nonenhancing area occupying nearly 75% of renal volume with the perinephric collection. In view of slow clinico-radiological response to injection amphotericin B, syrup posaconazole was added. Follow-up ultrasonography of the kidneys, ureters, and bladder showed avascular region in the superior and mid-region of the left kidney 5.4 cm × 2.3 cm, suggestive of the left renal abscess and subcapsular collection. Renal biopsy was done, which showed histomorphological evidence of mucormycosis. The patient then underwent left radical nephrectomy, and histopathological examination was consistent with mucormycosis with angioinvasion.

The patient thereafter was continued on liposomal amphotericin and syrup posaconazole along with supportive therapy. The patient responded well to given treatment in the form of general well-being, resolution of symptoms, and radiological improvement on follow-up imaging [Figure 4].

DISCUSSION

Mucormycosis belongs to the class zygomycetes. Humans largely acquire infection through the respiratory

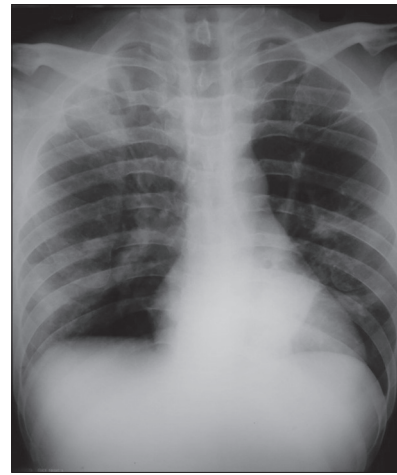


Figure 1: Chest radiograph showing alveolar opacities in the right upper, middle, and both lower zones

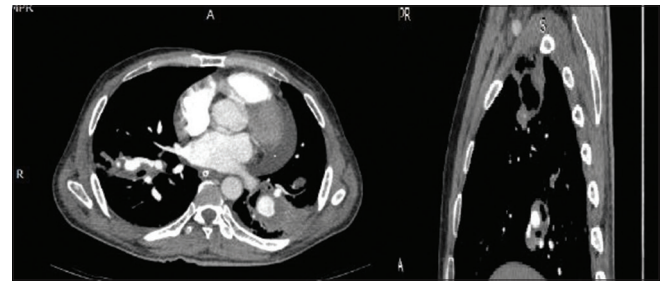


Figure 2: Computed tomography pulmonary angiogram revealed large cavitating consolidation in both lower lobes and right upper lobe with aneurysmal dilatation of the pulmonary artery and hypodense filling defect in segmental pulmonary arterial branches of the right upper lobe

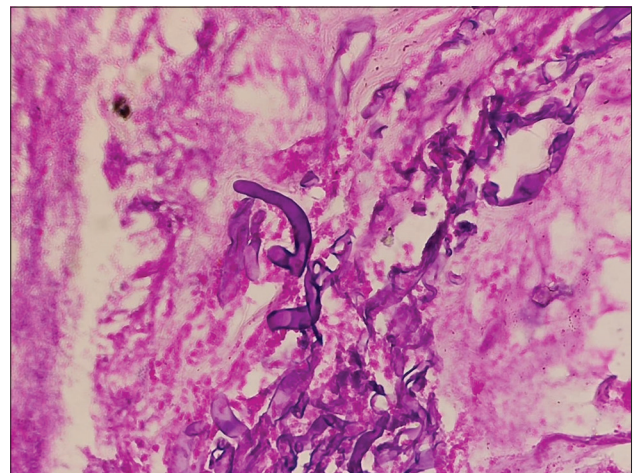


Figure 3: Photomicrograph of endobronchial biopsy showing broad aseptate hyphae of mucor (H and E, ×100)

route by inhalation of spores of the fungi.^[4] Pulmonary mucormycosis is the second most shared form of human mucormycosis after rhinocerebral variant and presents with fever, cough, breathlessness on exertion, chest pain, and occasionally life-threatening hemoptysis like in our case.^[5] Radiologically, it can present as lobar consolidation, cavitation, or an air crescent sign, which

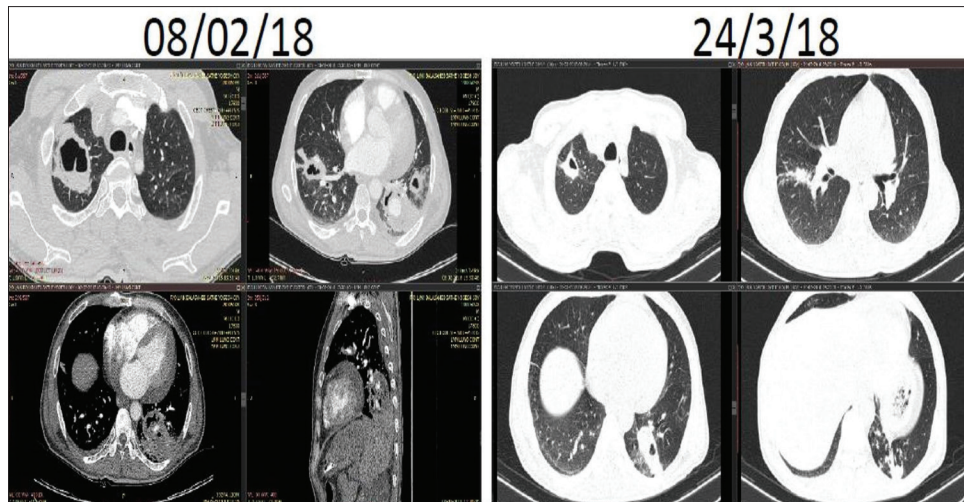


Figure 4: Computed tomogram of the chest showing significant regression of radiologic opacities after 6 weeks of treatment

indicates a vascular invasion by mucor.^[6] Pulmonary artery aneurysms are of diverse etiology ranging from congenital, trauma, vasculitis, pulmonary hypertension, and rarely secondary to infections such as tuberculosis and fungi.^[7,8] A mycotic aneurysm is one which is caused by the infectious involvement of the vessel wall.^[8] These show a distinctive peripheral distribution of the aneurysm.^[8] In our case, mucor was unswervingly accountable for the development of pulmonary artery aneurysm. The mechanism of aneurysm formation was most likely due to invasion of the vessel wall by mucor from adjacent parenchyma, causing inflammation and consequent weakening of the vessel wall. Antifungal therapy, along with surgical options of aneurysectomy, lobectomy, or pneumonectomy is the favored modality.^[2] Conservative options include endovascular coiling or embolotherapy.^[9] However, in our case, since the patient had bilateral pulmonary artery aneurysm, the surgical procedure was precluded; and the patient was managed conservatively with amphotericin B and posaconazole. However, the patient had undergone nephrectomy to reduce the fungal load. In our literature review, pulmonary mucormycosis has rarely been reported to present as a bilateral segmental pulmonary artery aneurysm in the background of immunosuppression, most common being uncontrolled diabetes mellitus. However, there is no report of such a case in Indian literature with disseminated mucor and bilateral segmental pulmonary artery aneurysm diagnosed antemortem. The other differentials that were considered were tuberculosis, malignancy, and vasculitis. They were ruled out through bronchoscopic studies, negative vasculitis workup, and later with

the good resolution of the pulmonary lesions with antifungal therapy.

CONCLUSION

Pulmonary mucormycosis can rarely cause pulmonary artery aneurysm which is associated with a high mortality. Early diagnosis of this complication and timely intervention can be life saving.

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Conflicts of interest

There are no conflicts of interest.

REFERENCES

- Kim HS, Oh YW, Noh HJ, Lee KY, Kang EY, Lee SY. Mycotic pulmonary artery aneurysm as an unusual complication of thoracic actinomycosis. *Korean J Radiol* 2004;5:68-71.
- Ramachandran L, Dewan S, Kumar V, Wankhade B. Mucormycosis causing pulmonary artery aneurysm. *Respir Med Case Rep* 2015;16:71-3.
- Loevner LA, Andrews JC, Francis IR. Multiple mycotic pulmonary artery aneurysms: A complication of invasive mucormycosis. *AJR Am J Roentgenol* 1992;158:761-2.
- Baker RD. Pulmonary mucormycosis. *Am J Pathol* 1956;32:287-313.
- Spellberg B, Edwards J Jr, Ibrahim A. Novel perspectives on mucormycosis: Pathophysiology, presentation, and management. *Clin Microbiol Rev* 2005;18:556-69.
- Lee FY, Mossad SB, Adal KA. Pulmonary mucormycosis: The last 30 years. *Arch Intern Med* 1999;159:1301-9.
- Trell E. Pulmonary arterial aneurysm. *Thorax* 1973;28:644-9.
- Bartter T, Irwin RS, Nash G. Aneurysms of the pulmonary arteries. *Chest* 1988;94:1065-75.
- Hammad AM, Al-Qahtani SM, Al-Zahrani MA. Huge pulmonary artery aneurysm. *Can Respir J* 2009;16:93-5.