Respiratory Medicine Case Reports 16 (2015) 71–73

Contents lists available at ScienceDirect

Respiratory Medicine Case Reports

journal homepage: www.elsevier.com/locate/rmcr

Case report

Mucormycosis causing pulmonary artery aneurysm

Lakshman Ramachandran [a, *], Sandeep Dewan [b], Vishal Kumar [b], Bhushan Wankhade [b]

[a] Jaypee Hospital, Critical Care Medicine, Sec 129, Noida, Uttar Pradesh, India
[b] Fortis Memorial Research Institute, Gurgaon, India

ARTICLE INFO

Article history:
Received 27 June 2015
Received in revised form
23 July 2015
Accepted 27 July 2015

Keywords:
Mucormycosis
Pulmonary artery
Aneurysm

ABSTRACT

Pulmonary artery aneurysm (PAA) is an uncommon entity and is usually congenital in origin or secondary to pulmonary arterial hypertension. Infections causing PAA are few, tuberculosis and bacterial infections being the common causative organisms. There have been few cases reported previously, in which the organism causing PAA was found to be a rare fungus called mucor. Pulmonary mucormycosis causing PAA is an infrequent and almost fatal complication as most of the diagnosis was made post mortem.

This report brings out a case of pulmonary mucormycosis causing ruptured PAA in a patient with diabetes. This patient was cured by a timely treatment of a combination of surgery and medical therapy.

1. Introduction

Pulmonary artery aneurysm (PAA) is an uncommon entity in a medical ICU as compared to the more prevalent aortic and intracranial aneurysms [1,2]. Most of them are found incidentally during radiological investigations in adults and are commonly congenital in origin. Mycotic or infectious pulmonary artery aneurysms are rare cause of acquired PAA. These are most commonly caused by Tuberculosis, syphilis, bacteria like streptococcus and staphylococcus and rarely fungus like actinomycosis and candida [3,4]. There are few scattered published cases describing pulmonary mucormycosis as causing PAA, with only very few survivals as ante mortem diagnosis is difficult [5,6]. Pulmonary mucormycosis is a rare opportunistic infection in patients with severe defects in any one of the arms of their immune system like diabetes, chronic renal failure, haematological malignancies, etc. We present a case report of a successful management of a ruptured PAA caused by mucormycosis in a diabetic patient.

2. Case description

A 58 yr old male, diabetic since 7yrs was presented to the Emergency Room (ER) with fever, breathlessness, cough and haemoptysis for the last two months which had increased in severity since two days.

One month back the patient had been evaluated for the above complaints and a diagnosis of right sided tubercular empyema (Fig. 1) was made, for which a chest tube drain was inserted and was prescribed anti tubercular treatment. The inter-costal drain was removed subsequently a week later and patient was discharged. But, his symptoms recurred and he was brought to our hospital with worsening of his condition.

In ER, on receiving, the patient was found to be pale, drowsy and had obvious respiratory distress. He also had features of circulatory shock with a blood pressure of 60/40 mm of Hg and increased heart rate of around 150 beats per minute. Immediate resuscitation was started with intravenous fluids, inotropes and oxygen supplementation by face mask. On examination, he was found to have a right-sided tension pneumothorax and an ICD was immediately inserted to decompress it. His general condition gradually improved. The patient was stabilised and shifted to Medical Intensive Care Unit (MICU).

A Computed Tomography (CT) of the chest done subsequently showed a large area of consolidation in the apical segment of right lower lobe (Fig. 2). He was put on broad spectrum intravenous antibiotics and his ATT (Anti Tubercular Treatment) was continued. His condition however did not improve. He continued to remain tachypneic and had recurrent episodes of massive haemoptysis (4–5 cupful). Subsequently Computed Tomography Pulmonary Angiography (CTPA) was done which revealed a saccular aneurysm involving the distal part of right lower lobar pulmonary artery measuring 3.1 × 2.3 cm with a neck diameter of 4.2 mm suggestive of Pulmonary Artery Aneurysm (PAA) (Fig. 3).

* Corresponding author.
E-mail address: laxman2104@gmail.com (L. Ramachandran).

http://dx.doi.org/10.1016/j.rmcr.2015.07.007

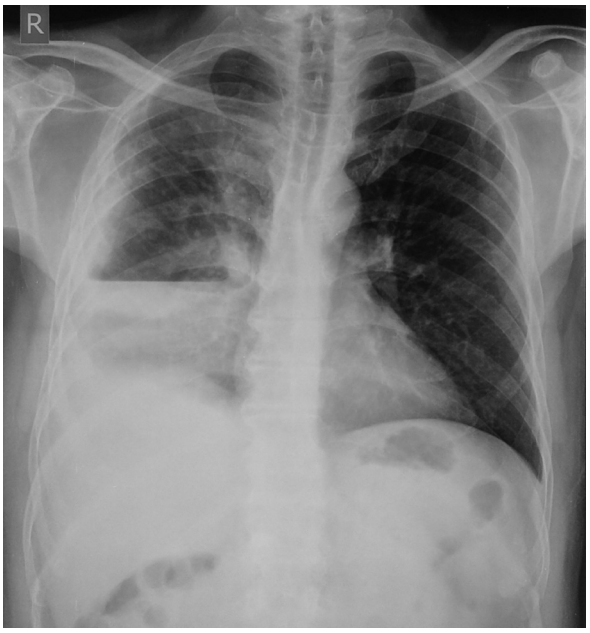

Fig. 1. Chest X-ray showing right hydro pneumothorax.

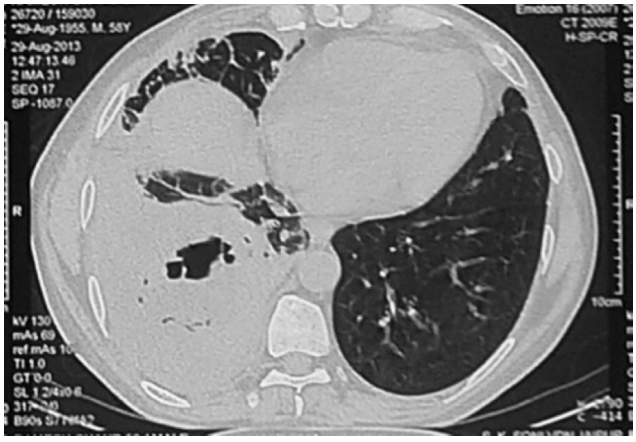

Fig. 2. CT showing collapse consolidation in right lower lobe with cavitation in the apical segment.

Few hours later, the patient suddenly went into respiratory distress and shock. His trachea was immediately intubated and mechanical ventilation was initiated. CT chest was done which showed right sided complete air space opacification probably haemorrhage. Suspecting it to be a ruptured PAA, patient was immediately shifted to the OT, where laparotomy and right lower lobe lobectomy with PAA clipping was done. The lobectomy specimen was sent for Histo Pathological Examination. (HPE).

Post operatively, he continued to have fever with rising TLC (Total Leukocyte Count) counts. He also had frequent recurrent right middle lobe and upper lobe collapse for which frequent bronchoscopic suctioning for mucus plug removal was done. Meanwhile, the HPE report of the lobectomised lung tissue was reported as invasive mucormycosis (Fig. 4) which was sensitive to amphotericin. Liposomal Amphotericin B was added at a dose of 200 mg per day (4 mg/kg/day). He responded well to the antifungal and became afebrile. His TLC counts normalised, oxygenation

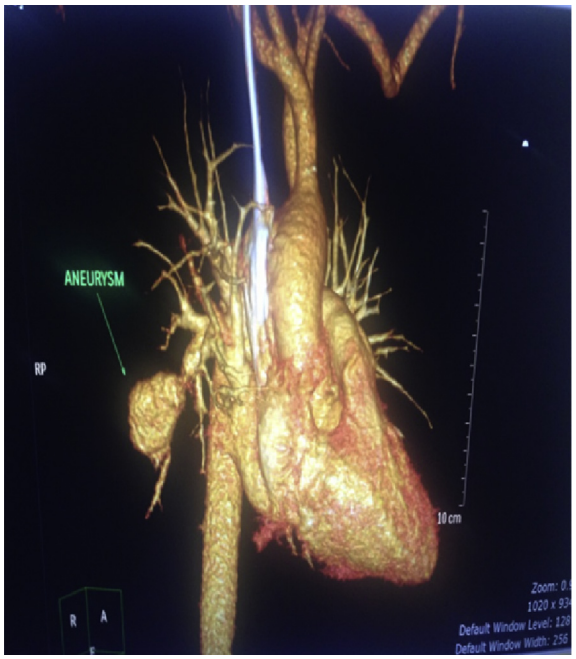

Fig. 3. A saccular aneurysm in the right lower lobar pulmonary artery.

improved and there was significant radiological Clearance. He was rapidly weaned off the ventilator and extubated. Post extubation, he was unable to clear secretions adequately because of his respiratory muscle weakness and poor cough reflex. So, on third day of extubation, he underwent minitracheostomy for adequate clearing of the secretions. Gradually, with aggressive chest physiotherapy and regular suctioning, the patient improved and was subsequently 'discharged on request' in a stable condition (with minitracheostomy in situ) to a primary health care centre. Unfortunately, Patient was lost in follow up.

3. Discussion

Pulmonary mucormycosis which was previously found only in immunocompromised individuals like solid organ transplant recipients, patients on immunosuppressant, haematological malignancies etc., has now emerged as a life-threatening infection in patients with diabetes in medical ICU [7–9]. Most of the cases are diagnosed post-mortem, hence early suspicion and detection can curb the high mortality rate.

Mucormycosis belongs to the genus mucor, order mucorales and class zygomycetes which survive on decaying organic matter. Humans are mainly infected through inhalation of the spores [10].

Pulmonary mucormycosis is the second most common form of human mucormycosis after the rhino-orbital-cerebral form and presents with fever, chest pain, cough and rapidly progressing breathlessness and occasionally as life-threatening massive haemoptysis [8]. Rarely, it can also present with vocal cord palsy [9]. Radiological examination may show a wedge-shaped lobar consolidation, cavitation or an air crescent sign which indicates a life threatening vascular invasion by mucor and requires a prompt intervention [11]. Its uniqueness lies in its ability to invade the vessel wall causing thrombophlebitis and thrombosis leading to large parenchymal infarctions and occasionally, aneurysm and pseudo aneurysm formations in the pulmonary vascular system [4].

Pulmonary artery aneurysm is the focal dilatation of pulmonary vessels beyond 29 mm of diameter on CT scan [2,12]. A true

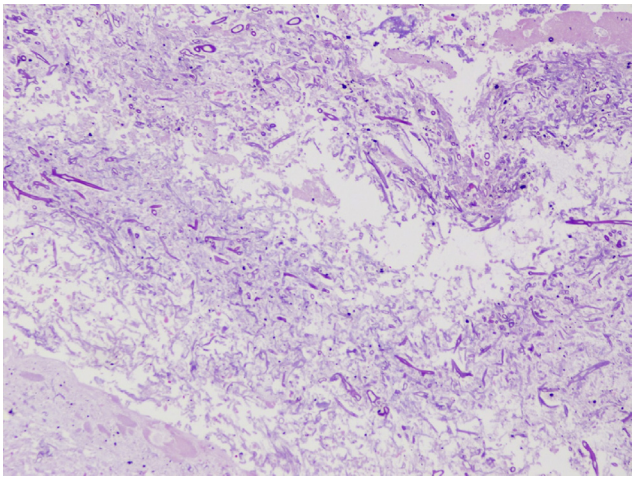

Fig. 4. H & E stained section showing sparsely septate irregular obtuse fungal hyphae (broken arrows) in a necrotic background (solid arrows). X10X.

aneurysm involves all three layers of the vessel wall whereas, a pseudo aneurysm is just a leaking haematoma layered by surrounding soft tissues. PAA are mostly congenital. Acquired forms are less common and are usually secondary to pulmonary hypertension associated with chronic pulmonary emboli or obstructive airway disease, mitral stenosis and other veno-occlusive diseases [13]. Mycotic aneurysms due to infections are very rare and are mainly associated with tuberculosis (Rossenmuesen aneurysm) which shows a characteristic peripheral distribution of aneurysms [14,15]. We believe, in the present case, mucor was directly responsible for the development of PAA. Mucor had encroached upon the vessel wall from the surrounding parenchyma causing inflammation and weakening of the vessel wall leading to the aneurysm formation.

Pulmonary CT angiography is the gold standard to diagnose PAA [12]. Combination of antifungal with surgical resection of the involved tissue is the best approach advocated [16]. Surgical resection is necessary as medical treatment alone has higher mortality because of the angioinvasive nature of the fungus. Surgical intervention is preferred earlier if the patient presents with haemoptysis. Surgical modalities now include aneurysectomy, lobectomy or pneumectomy [17]. If the aneurysm does not involve the main pulmonary trunk, nonsurgical techniques like embolotherapy or endovascular coiling is preferred [18].

Liposomal Amphotericin B is the recommended antifungal agent. The dose and duration of therapy is individualised depending on the clinical response, clearing in the radiological findings and negativity of the cultures. Posaconazole is the alternative drug against mucorales species for patients who are refractory or intolerant to amphotericin therapy [15].

In a nutshell, PAA is a deadly complication of pulmonary mucormycosis. For early diagnosis, keep a high index of suspicion in high risk patients like diabetes, chronic renal failure, haematological malignancies, etc. who are not responding to conservative management. Lung biopsy and pulmonary angiography to confirm infection and aneurysm respectively are necessary, when routine investigations are inconclusive. Multimodality approach combining medical therapy and surgical or endovascular intervention is the best approach observed although data available is limited. In our view, a good post-operative rehabilitation care also contributes to improved results.

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