



Case report

Rasmussen's pseudoaneurysm- case report

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A B S T R A C T

Background: Pulmonary aneurysms and pseudoaneurysms have various etiologies; however, the term Rasmussen's pseudoaneurysm refers specifically to a focal dilatation of a branch of the pulmonary artery into adjacent tuberculous cavity. The incidence of such tuberculosis related pulmonary vascular complication is extremely rare, hence, under recognized by many physicians. Management of pulmonary pseudoaneurysms is challenging as they present by life-threatening hemoptysis. Furthermore, contrary to the most causes of massive hemoptysis their bleeding is of pulmonary rather than bronchial artery origin. Prompt diagnosis and early interventions are needed as a very high mortality rate is associated with this illness.

Case description: We are reporting on a case of a young male who was presented to our hospital with recurrent episodes of massive hemoptysis and was diagnosed to have pulmonary tuberculosis. Despite being actively treated, his hemoptysis persisted. We describe in this case the role of different diagnostic modalities and the available therapeutic options.

Conclusion: Rasmussen's pseudoaneurysm is rare and potentially lethal pulmonary vascular complication of tuberculosis. It should be considered in the differential diagnosis of hemoptysis in patients known or suspected to have pulmonary tuberculosis. In such cases, multidetector computed tomography (MDCT) scanning is the investigation of choice to confirm the diagnosis and to localize the source of bleeding prior to the therapeutic interventions. Head to head comparison between interventional radiology procedures and surgery in treatment of pulmonary pseudoaneurysms is lacking, thus, choice depend on the availability and local expertise.

1. Introduction

Pulmonary tuberculosis (PTB) poses tremendous diagnostic and therapeutic challenges as it can present by various forms of complications even after complete biological cure of the disease [1]. These complications may involve the lung parenchyma, airways, pleura, or pulmonary vessels. The mediastinum and chest wall can be involved as well. Cavitation, bronchiectasis and pleural disease are being the most common. On the other hand, vascular complications are rare sequelae of PTB; however, they carry a very high mortality rate. Aneurysmal dilatation of the pulmonary artery adjacent to PTB cavity (Rasmussen's pseudoaneurysm) is one of these vascular complications which estimated to occur in 0.25% of PTB patients [2]. It usually presents clinically by massive hemoptysis; thus carry extremely high mortality rate that exceeds 38% [2]. Massive hemoptysis in the vast majority of PTB cases is from bronchial circulation, however, in Rasmussen's pseudoaneurysm the bleeding is of pulmonary artery origin [3]. As a result of its rarity and under recognition by many physicians the diagnosis is usually delayed or missed. Rasmussen's pseudoaneurysm should be considered in the differential diagnosis of hemoptysis in PTB patients along with other causes of hemoptysis such as post PTB bronchiectasis, pulmonary embolism and bleeding from cavitary lesions.

In this report we present one such case of Rasmussen's aneurysm and review of the relevant literature.

2. Case report

We are reporting on a 28 years –nonsmoker- male, prisoner, who presented to the hospital complaining of coughing up a fresh blood for few weeks. His hemoptysis was associated with fever, loss of appetite and weight. He reported having contact with a prisoner diagnosed to have pulmonary tuberculosis. He had no risk factor of pulmonary embolism or bronchiectasis. He was not on regular medications for any chronic medical illness. On examination, he was fully conscious, alert, oriented and not in acute respiratory distress. However, he looked ill and underbuilt. His chest examination revealed bilateral upper lobes bronchial breathing with diffuse scattered inspiratory and expiratory crepitation. Apart from tachycardia, examination was normal. His chest X-ray revealed bilateral diffuse reticulonodular opacities with some fibrotic changes in both apices (Fig. 1). He was admitted and isolated into the negative pressure room. Sputum acid fast bacilli (AFB) and Nucleic Acid Amplification (NAA) Tests confirm the diagnosis of pulmonary tuberculosis. First line standard medications for PTB were immediately commenced. Despite he had culture proven pan-sensitive

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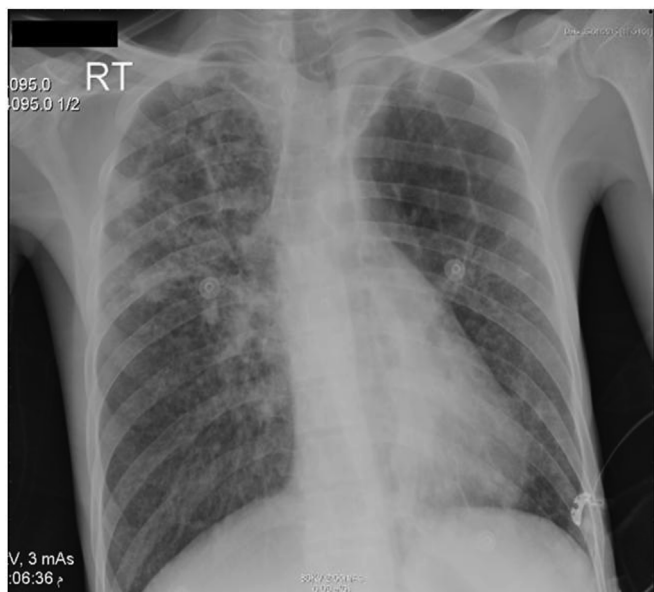


Fig. 1. Chest X-ray showing bilateral reticulonodular opacities with some fibrotic changes in both apices.

Mycobacterium he continued to have significant amount of hemoptysis and his AFB remained positive throughout his hospitalization. He received repeated packed red blood cells (PRBCs) transfusion to correct his severe anemia. A total of 3 units of (PRBCs) were required to maintain his Hemoglobin (Hb) around 9 g/dL. Fiberoptic bronchoscopy examination was performed to diagnose the cause of bleeding; inspection showed normal airways, no endobroncheal mass, vascular lesion or other obvious source of bleeding identified. Bronchoalveolar lavage (BAL), brush samples and transbroncheal biopsies were taken during the procedure. Cytology and microbiology results showed abundant

polymorph neutrophils and red blood cells consistent with acute inflammation, AFB was present. Histopathology examination showed no abnormal finding other than active chronic inflammatory processes. A computed tomographic (CT) pulmonary angiogram was done, and revealed a thick walled cavitary lung lesion filled with blood, and surrounded by fibrotic and bronchiectatic changes (Fig. 2). A three dimensional reconstruction of the vascular anatomy (MDCT) showed a very large dilatation of pulmonary arterial wall (Rasmussen's pseudoaneurysm) measuring $81 \times 65 \times 62$ mm in the craniocaudal, anteroposterior and transverse diameter respectively. The pseudoaneurysm was originating from a left lower lobe segmental pulmonary artery branch (Fig. 3). Either pseudoaneurysm embolization or lobectomy was discussed in a multi-disciplinary meeting with thoracic surgeons and interventional radiologists as he continued to have recurrent episodes of major hemoptysis. Pseudoaneurysm embolization and subsequent coil insertion were decided and planned. Unfortunately, the patient had hemodynamic collapse during the procedure, and was resuscitated in the angiography suit then shifted to intensive care unit (ICU) where he remained relatively in stable hemodynamic condition. Few days later he had a second sudden cardiovascular collapse and eventually succumbed to his illness.

3. Discussion

A focal dilatation of the pulmonary artery is a rare pathological complication of various pulmonary and systemic diseases. In cases where all three layers of pulmonary artery wall are involved the focal dilatation is termed aneurysm; pseudoaneurysm on the other hand, does not involve all arterial wall's layers [4]. Nevertheless, the term aneurysm and pseudoaneurysm are commonly used interchangeably in the literature [5]. Many etiologies are implicated in pulmonary aneurysms and pseudoaneurysm as post-infection as PTB, trauma, congenital, inflammatory (e.g. Behcet disease) and pulmonary hypertension. The term Rasmussen's pseudoaneurysm refers specifically to tuberculous etiology. It results from a PTB cavitary lesion that erodes

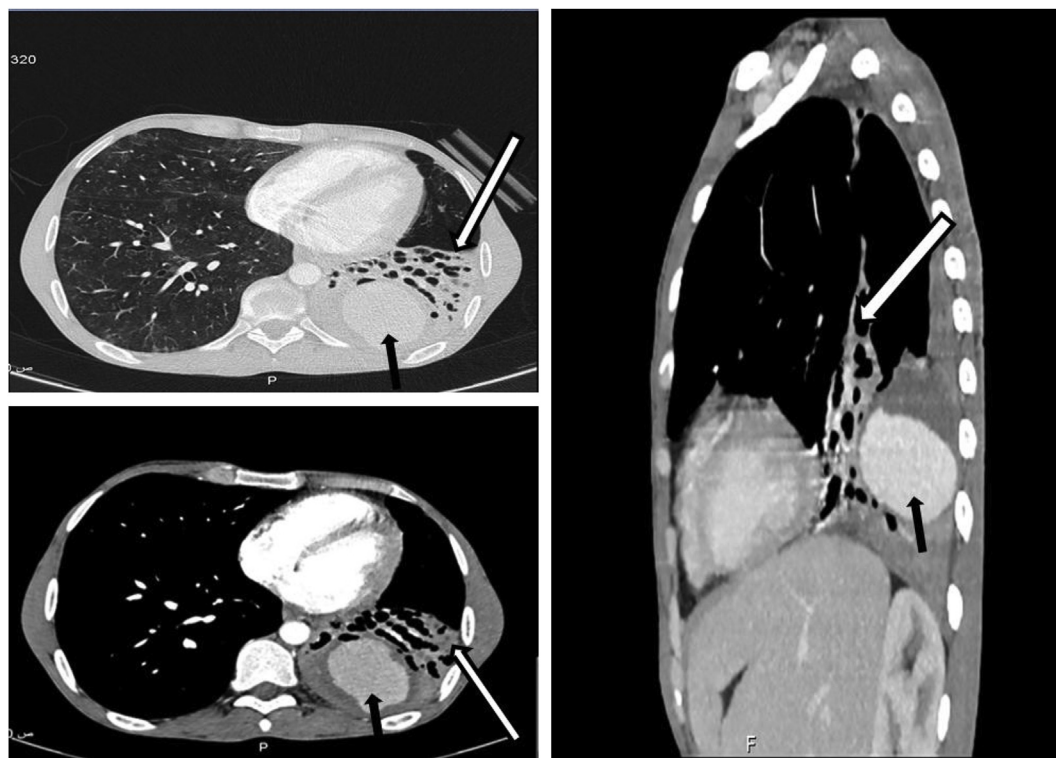


Fig. 2. Chest CT showing blood-filled cavitary lung lesion (black arrows). The cavity is surrounded by fibrotic and bronchiectatic changes (white arrows).

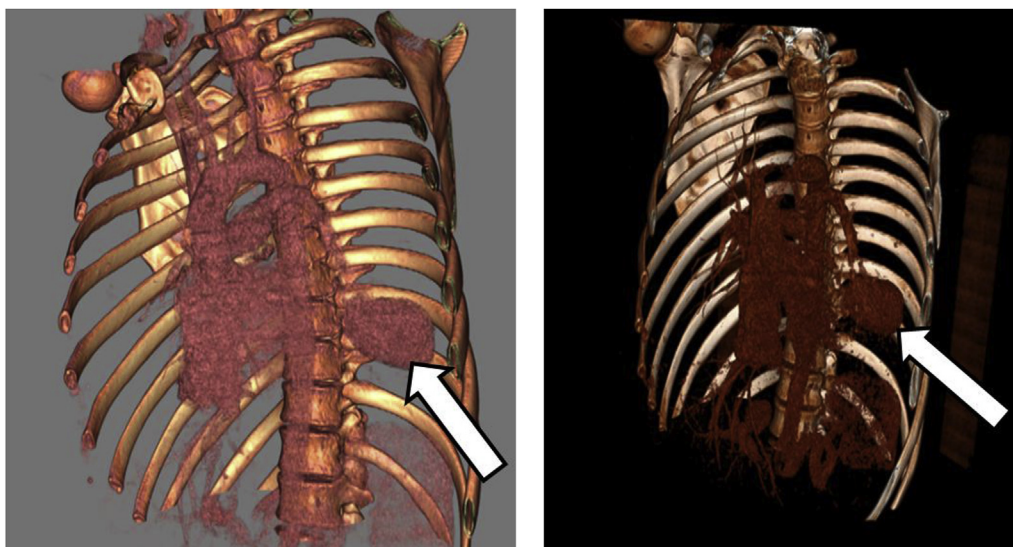


Fig. 3. A three dimensional reconstruction of the vascular anatomy (MDCT) showing details of the lung vascular. A pseudoaneurysm that measures $81 \times 65 \times 62$ mm and originates from a left lower lobe segmental pulmonary artery branch is indicated by a white arrow.

the adjacent structures in the lung, when a branch of the pulmonary artery come adjacent to or within the cavity, its wall progressively weaken as granulation tissue replaces both the adventitia and the media layer. Thereafter, the artery wall expands until it bursts resulting in pseudoaneurysm formation. These pseudoaneurysms can potentially rupture producing massive hemoptysis [6,7]. Rasmussen's pseudoaneurysm was reported in literature as early as the 19th century [8,9]. It was not well characterized until a detailed pathological description of 11 cases detailed by Dr. Fritz Rasmussen in 1868 and named after him [10].

In a study were 21,532 patients with confirmed PTB underwent multidetector computed tomography (MDCT) scanning or pulmonary angiography, Rasmussen's pseudoaneurysms were found in only 54 cases (0.25%) [2]. In PTB cases presenting with massive hemoptysis the incidence is higher, as it was shown to be present in 7% in one autopsy series of 80 patients who had PTB and massive hemoptysis [11]. In another series of 8 patients presented with severe hemoptysis secondary to cavitary PTB and failed arterial embolizations, selective pulmonary angiography diagnosed three patients (38%) to have Rasmussen's pseudoaneurysms [12]. These pulmonary artery pseudoaneurysms are the source of bleeding in this rare tuberculosis related vascular complication; nevertheless, bronchial artery is by far the commonest source of life-threatening hemoptysis even in PTB patients [13,14].

Flexible fiberoptic bronchoscopy is advocated as initial procedure of choice to investigate massive hemoptysis cases as it can be performed at the bedside and has potentially diagnostic as well as therapeutic benefit. It can localize the bleeding site in up to 93% of cases [15]. Moreover, bronchoscopy could localize the bleeding or diagnose its cause when chest x-ray and/or CT fail to do so [15,16]. In the other hand, bronchoscopic examination of the airways does not have specific findings in many important causes of hemoptysis such as bronchiectasis as an example [17]. In our case, bronchoscopic examination was not informative, it merely confirmed the present of AFB in the airways. In centers were urgent fiberoptic bronchoscopy is not available, thoracic angio CT scan is being used as alternative diagnostic modality which have high sensitivity to guide the therapeutic intervention [14]. CT scan has been shown in clinical studies to be the most sensitive diagnostic test when employed alone [16]. The Highest diagnostic sensitivity can be achieved by combining a CT scan with a bronchoscopy [16].

The therapeutic roles of the flexible fiberoptic bronchoscopy include

localization; ensure hemostasis or isolation of the bleeding segment using different modalities [18]. Such an intervention help prevent asphyxiation and usually used as a bridge for the definite intervention. The superiority of either interventional radiology procedures or surgical treatment of pulmonary aneurysms and pseudoaneurysms has never been established as head to head comparisons between these modalities are lacking. Generally, graft placement or aneurysmorrhaphy are performed in aneurysm involving pulmonary trunk [19]. In the other hand, Rasmussen's pseudoaneurysms rise from peripheral branches of pulmonary artery, and thus require lobectomy or even pneumonectomy [20]. Endovascular aneurysm repair (EVAR) a less invasive technique which does not require open surgery has been described in a few case reports as a compassionate release indication [21,22]. Many centers reserve surgery for cases refractory to angiographic embolization as patients who undergo emergency surgical resection during active massive hemoptysis have a high morbidity and mortality rate [23]. Moreover, as it was the case in this reported patient, many patients presenting with severe hemoptysis are not surgical candidates due to the presence of extensive PTB causing insufficient respiratory reserve [12]. Such cases mandate minimally invasive techniques and thus the interventional radiology procedures are preferred. The success rate of angiographic embolization for massive hemoptysis (of all causes) is reported to exceed 90% [24]. Bronchial artery embolization constitutes the main bulk of the published literature being the commonest source of bleeding in massive hemoptysis cases. Very few cases have been described in literature to have successful interventional angiographic occlusion of the pseudoaneurysms using different techniques [12,13,20,25].

4. Conclusions

Our case highlights many important points; firstly, the disease rarity hinders its suspicion and hence early investigation and diagnosis. Secondly, it poses diagnostic challenges as despite it causes massive hemoptysis, bronchoscopic examination which is the initial diagnostic tool of choice in cases of massive hemoptysis is usually unrevealing. Furthermore, PTB commonly causes cavitary lung lesions and bronchiectasis; both present with hemoptysis and mimic Rasmussen's pseudoaneurysm on standard chest CT images. Finally, it shows the therapeutic options and highlights the extremely high mortality risk of this illness.

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