Ambiguous presentation of *Mycobacterium avium* complex-associated Rasmussen aneurysm

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Keywords

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**Abstract**

A 77-year-old man with a progressively dry cough (two months duration) was admitted with hemoptysis. Chest computed tomography (CT) revealed left lingular lobe consolidation and one thick-walled cavity lesion over the left lower lobe, which was accompanied by satellite micro-nodules in a tree-in-bud pattern. CT-guided biopsy confirmed mycobacterial infection, and subsequent culture yielded *Mycobacterium avium* complex (MAC). Unremitting hemoptysis was present despite treatment (14 days) with ethambutol, rifampin, clarithromycin, and streptomycin. Initial CT angiography (CTA) to determine the source of the hemoptysis revealed a suspected aneurysm in the consolidated left lingular lobe; however, this could not be localized via catheter angiography during the pulmonary and bronchial arterial phases. Two weeks later, a massive hemoptysis episode led to hemodynamic instability and serious consequences. Follow-up CTA confirmed the previously detected aneurysm, and glue embolization was performed successfully. This case report highlights a rare but catastrophic MAC-associated pseudoaneurysm and relevant treatment options.

**Introduction**

Rasmussen aneurysm is quite rare, with an estimated prevalence of 5%. It is a known complication of pulmonary tuberculosis (TB). To the best of our knowledge, this is the first report of non-tuberculosis mycobacterium (NTM)-related Rasmussen aneurysm. Dilatation of the pseudoaneurysm resulted from weakening of the pulmonary artery wall by the adjacent tubercular cavity [1]. The arterial wall was replaced by granulation tissue and fibrin, which subsequently led to rupture. Hemoptysis is a common presentation that can be fatal when massive.

Minor hemoptysis occasionally occurs in cases of pulmonary TB; however, it is mostly self-limiting and remitted by anti-tubercular therapy (ATT). Life-threatening hemoptysis generally originates arterially [1]. It is defined as >250 mL of blood coughed up within a 24-h period, with mortality of approximately 80% if left untreated [2]. Urgent angiographic embolization or surgical resection is required for the management of major bleeding. In cases of NTM infection, more intensive management is required owing to difficult diagnosis and poor treatment response.

**Case Report**

A 77-year-old Asian man was admitted with a three-day history of episodic hemoptysis. The patient was a retired janitor, a non-smoker, and had experienced a chronic dry cough for two months prior to admission. He denied any systemic disease, but hyperglycaemia was detected during hospitalization. At initial hospitalization, chest radiography revealed left lower lung field opacity with a positive silhouette sign over the left heart border. Chest computed tomography (CT) showed left lingular lobe consolidation and a thick-walled cavity lesion over the left lower lobe accompanied by satellite micro-nodules in a tree-in-bud pattern (Fig. 1A). CT-guided biopsy indicated chronic granulomatous inflammation with caseous necrosis, multinucleated histiocytes, and acid-fast bacilli, consistent with
Figure 1. (A) Chest computed tomography (CT) showing left lingular lobe consolidation and a cavity in the lower lobe surrounded by satellite micro-nodules. (B) Chest CT angiography showing a nodule (arrow) with delayed enhancement. (C) No aneurysm was detected on either pulmonary angiography (left) or bronchial artery angiography (right).

Figure 2. (A) Blood trickled down from the left lingular division of the bronchus after the removal of retained blood clots at the position of the left second carina (top left). Blood slowly oozed from the left lingular division of the bronchus (top right) with narrowed orifice during external compression. No bleeding was observed from the left superior division of the bronchus (bottom left) or the left inferior lobar bronchus (bottom middle and right). (B) Contrast between the aneurysm and adjacent feeding vessel (arrow) demonstrated via glue infusion.
mycobacterial infection. Four sputum smears were positive for acid-fast bacilli, but polymerase chain reaction testing for TB showed negative results. After treatment with empirical broad-spectrum antibiotics for suspected necrotizing pneumonia, the hemoptysis improved and the patient was discharged. Two days later, the patient revisited our emergency department because of relapsed hemoptysis. Chest radiography showed increased ground-glass opacity around the previously identified area of lung consolidation. Chest CT angiography (CTA) showed an increased alveolar opacity and a suspected aneurysm (Fig. 1B, arrow) in the consolidated left lingular lobe, which was enhanced to a greater extent in the aortic phase (Fig. 1B, arrow). However, the aneurysm could not be localized via catheter angiography in either the pulmonary or the bronchial arterial phase (Fig. 1C). All sputum and biopsy tissue cultures yielded *Mycobacterium avium* complex (MAC). ATT with ethambutol, rifampin, clarithromycin, and streptomycin was initiated. However, repeated massive hemoptysis led to hemodynamic instability, and cardiopulmonary-cerebral resuscitation was required two weeks post-ATT. Bronchoscopy revealed the source of the hemoptysis—at the left lingular division of the bronchi; external compression had caused narrowing of the orifice (Fig. 2A). Bronchial biopsy culture over the lingular lobe also yielded MAC. A second CTA showed a more evident aneurysm, with a clearly identified feeding vessel. Glue embolization was then performed (Fig. 2B). Despite cessation of hemoptysis, hypoxemic insult-related encephalopathy developed despite timely recovery of spontaneous circulation after resuscitation.

**Discussion**

TB-related hemoptysis can result from bronchiectasis of bronchial artery origin or, more rarely, from Rasmussen aneurysm of pulmonary artery origin. ATT is the main component of treatment; additional treatment options include conservative medical treatment, angiographic embolization, and surgical resection. Surgical resection is reserved for localized disease with refractory bleeding, or in cases refractory to ATT [3].

Diagnosis of NTM infection is based on mycobacteriological findings, as well as radiographic and clinical evidence. Eradication of NTM infection is far more difficult than that of TB. Recurrent infection by different strains of mycobacteria or relapse of the original organism is quite common [3]. More intensive evaluation of NTM infection-related complications, such as Rasmussen aneurysm in the present case, may lead to earlier detection and prevention of poor outcome.

In cases where an aneurysm is detected on CTA but is not observed during subsequent catheter angiography, several explanations are possible. These include partial thrombosis of the feeding vessel causing delayed and weak contrast enhancement in angiography, and rendered enhancement from digital subtraction for reduction of motion artefact. Delayed enhancement of the pseudoaneurysm in the aortic phase may result from partial thrombosis of the feeding vessel, low-pressure pulmonary arterial flow combined with low-contrast dose usage, or may be a feature of NTM-associated Rasmussen aneurysm.

This is the first reported case of a NTM-associated Rasmussen aneurysm; it highlights the fact that there should be increased awareness of pseudoaneurysm formation in NTM infection. A pseudoaneurysm may be more evident in the late arterial phase, or even in the aortic phase on CTA imagery. Chest CTA is more sensitive regarding the detection of pseudoaneurysm than catheter angiography.

**Disclosure Statements**

No conflict of interest declared. Appropriate written informed consent was obtained for publication of this case report and accompanying images.

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