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Case Report

Bronchial artery to pulmonary artery fistula in a patient with cavitating tuberculosis—Demonstration with 4D CTA ☆

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ABSTRACT

We present a case of a bronchial artery to pulmonary artery fistula. This occurred in a 77-year-old male of Bangladeshi ethnicity with a new diagnosis of cavitating pulmonary tuberculosis. A 4D CTA protocol was required to elicit the nature of the vascular abnormality. Fistula between bronchial and pulmonary arteries is an uncommon phenomenon, with few published cases. This case demonstrates the difficulty with which these fistulae are imaged. 4D CTA is a technique that is increasingly being shown to be useful in characterizing vascular anomalies such as fistulae, with embolization as the mainstay of treatment.

Case presentation

Fistulae between bronchial and pulmonary arteries are relatively uncommon, as reflected by the limited available literature. The cause of these fistulae includes infection, malignancy, trauma, and congenitally acquired [1,2]. It is reported that tuberculosis (TB) is the most common infective cause [3]. These fistulae can appear like a Rasmussen aneurysm radiologically (a Rasmussen aneurysm is a pseudoaneurysm of a pulmonary artery branch in the wall of a tuberculous cavity [4]).

A 77-year-old male of Bangladeshi ethnicity was admitted with low volume hemoptysis, weight loss, and fevers in the

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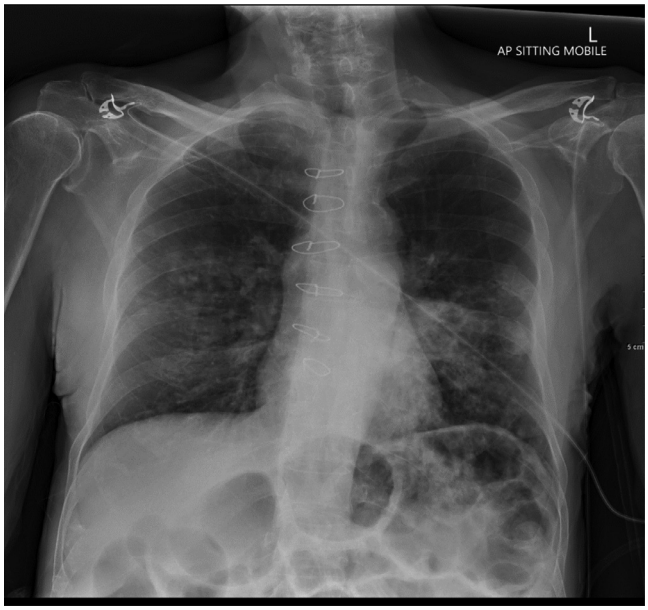

Fig. 1 – Chest X-ray AP showing mulifocal consolidation in the right midzone and left mid and lower zones, with more confluent consolidation/opacity in the left midzone. There is a likely cavitation in the left mid zone opacity.

context of a progressively worsening cough over the last 5 months. His background is significant for Alzheimer's dementia, Parkinson's, coronary artery bypass graft in 2013, insulin-dependent diabetes, and hypertension. He was taking clopidogrel 75 mg daily, rivastigmine 1.5 mg twice a day, metoprolol 50 mg twice a day, rosuvastatin 5 mg at night, telmisartan 80 mg at night, vildagliptin 50 mg at night, dapagliflozin 10 mg daily, levodopa/carbidopa, and insulin. The patient was afebrile with an oxygen saturation of 96% on room air, blood pressure of 138/53 mmHg, pulse rate of 68 bpm, and a respiratory rate of 18. He was cachectic with notable clinical findings of left thoracic posterior-basal inspiratory crepitations on auscultation (Fig. 1).

A CT pulmonary angiogram showed a 61 × 46 × 45 mm thick walled cavitating lesion in the superior segment of the left lower lobe. There was tree-in-bud nodularity in all lobes of the lung but most confluent in the left lower lobe. A CT thoracic aortogram showed a high-density lobulated structure of the same density as adjacent intra-arterial blood in the medial wall of the cavitating lesion. This was closely related to the pulmonary artery supplying the area.

He tested positive for pulmonary TB based on a mycobacterial polymerase chain reaction test on spontaneous expectorated sputum. This diagnosis was later confirmed on mycobacterial cultures. Human immunodeficiency virus antibodies were negative. At this juncture, the patient's hemoptysis was significant and further imaging was performed for potential angioembolization.

A 4D CTA protocol was performed. Dynamic intermittent axial/sequential scans through the region of interest in the left lower lobe were performed for 20 seconds after administration of intravenous contrast. The axial/sequential slab had 16 cm coverage in the Z-direction. This resulted in pre-contrast, pulmonary arterial and thoracic aortography phases being acquired. This demonstrated an enhancing focus on the medial aspect of the cavity during the systemic arterial phase which appeared to be supplied by a small left bronchial artery. Furthermore, there was systemic arterial phase enhancement of the adjacent pulmonary artery branch (which appeared dilated) and associated laminar flow enhancement extending into the left lower lobar pulmonary artery. Review of the pulmonary arterial phase images showed the draining pulmonary artery branch and unenhanced laminar flow artefact in the lower lobe pulmonary artery (Fig. 2).

The imaging demonstrated a high-density structure in the wall of the cavity and given the patient's hemoptysis; this was thought to be the culprit lesion. Based on the density, location, and the pattern of enhancement, we proposed the differential of Rasmussen pseudoaneurysm of either a bronchial or pulmonary artery or a bronchial-pulmonary arterial fistula. The 4D CTA showed concurrent enhancement in different vascular filling phases which was characteristic of a bronchial-pulmonary arterial fistula.

His pulmonary TB was initially treated with pyrazinamide, ethambutol, rifampicin, and isoniazid. This was complicated by asymptomatic hepatitis and pancreatitis which resulted in suspension of all TB therapies until there was sustained improvement in liver enzymes and lipase. Isoniazid was thought to be the culprit. He was later recommenced on moxifloxacin, ethambutol, pyrazinamide, and rifampicin with no further complications. His hemoptysis was treated successfully with cessation of clopidogrel and initiation of oral tranexamic acid. The patient's hemoptysis resolved completely after 5 days of tranexamic acid. Clopidogrel was never reinitiated during his admission, with cardiology advice to consider aspirin on subsequent outpatient review. He was discharged and returned to Bangladesh 2 weeks later.

The cause for arteriovenous fistula secondary to TB is uncertain but considered to be multifactorial, with a combination of chronic inflammation, architectural change, and angiogenesis [5]. Reported cases highlight the difficulty in imaging these systemic to pulmonary shunts. This provides a clinical dilemma if hemodynamic instability and large hemoptysis is present [1,6]. Catheter angiography may be insufficient for diagnosis depending on the size and origin of the feeding bronchial artery, the size of the draining pulmonary artery, and the amount of shunting.

In the absence of an acute event, bronchial-pulmonary artery fistulae can be subclinical. Long-term clinical effects of these fistulae are often rare and require multiple fistulae to manifest. However, if the fistulae are large enough, they can cause sufficient left to right shunting to result in pulmonary hypertension, as reported by Jahcec et al. [2]).

First-line treatment for hemoptysis is supportive measures (including oxygen), correction of any coagulopathy, and nebulized tranexamic acid. Early bronchoscopy and CT should be performed to localize bleeding, with bronchoscopy having the potential to directly treat the cause. Angiography has a significant role, with bronchial arteriography performed prior to pulmonary angiography because of the higher likelihood and risk of bronchial bleeding (as the systemic pressure is higher than pulmonary pressure). A systematic review of bronchial artery

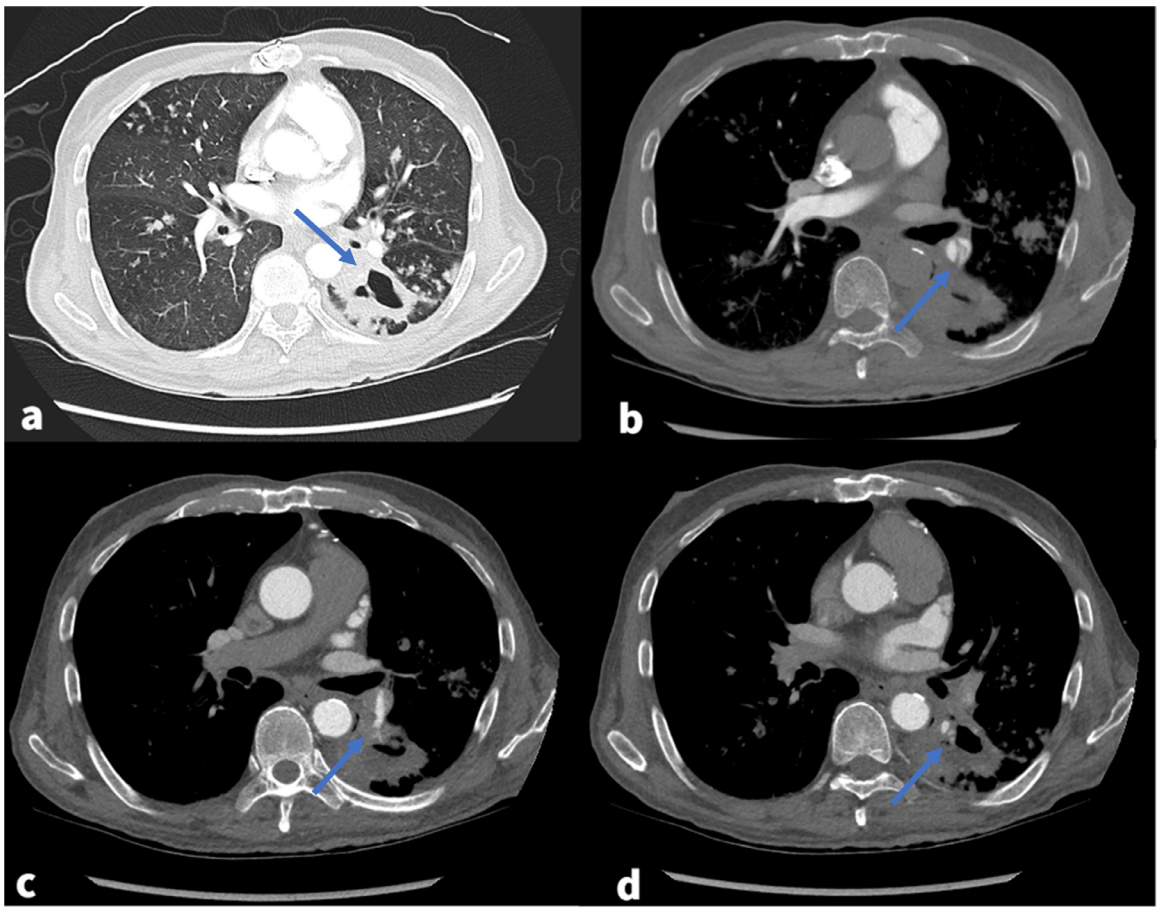

Fig. 2 – 4D CTA showing (a) a thick-walled cavitating lesion in the left lower lobe with surrounding tree-in-bud nodules indicative of active TB, (b) a jet of unopacified blood in the left pulmonary artery from the bronchial to pulmonary artery fistula, (c) from later in the 4D series a jet of opacified blood from the bronchial artery flowing into the unopacified pulmonary artery, and (d) adjacent bronchial artery pseudoaneurysm.

embolization by Panda et al. [7] found success rates in hemoptysis to vary between 70% and 99% with recurrence 12%-57%. Surgery is an important alternative option, depending on the cause of the bleeding [8].

There is much less literature on treatment of hemoptysis due to bronchial-pulmonary artery fistulae. Options include bronchial artery ligation, lobectomy, and embolization [9]. Bronchial artery embolization in massive hemoptysis is shown to have a high rate of success in controlling symptoms [3], with minimal complications. Kato et al. [9] discussed the role of lobectomies as an alternative to arterial embolization in the context of a bronchial-pulmonary artery fistula from a pulmonary hemangioma. They described surgery to be more definitive than embolization albeit more invasive. The published cases favor angioembolization as the first-line treatment.

Bronchial to pulmonary artery fistulae, like other vascular anomalies, are difficult to image. Radiologically, the difficulty in imaging stems from numerous factors. These include the dynamic enhancement of various portions of the vascular anomaly at differing timepoints, the complex—often tortuous—morphology of the lesions, and the constantly moving background of the lungs and nearby heart [10,11]. The traditional gold standard for bronchial-pulmonary artery fistula is digital subtraction angiography (DSA). 4D DSA is an established technique for imaging in vascular malformations in interventional neuroradiology [10]. This allows the vascular architecture to be viewed from any projection at any point in time and allows treatment of the malformation to be more easily performed and planned. It was also reported that the likelihood of repeat imaging is lower than in 2D and 3D modalities. 4D CTA is being increasingly used for nervous system arteriovenous malformations. A review by Wang et al. [11] shows that 4D CTA provides similar accuracy of imaging to 4D DSA, with some minor limitations.

We are not aware of any prior reports showing the utility of 4D CTA in imaging bronchial artery to pulmonary artery fistulae.

In conclusion, bronchial-pulmonary arterial fistulae are radiologically difficult to image. We show a case where dynamic 4D CTA was used to confirm this diagnosis where traditional CT pulmonary angiography and CT aortic angiography were both unclear. Basic supportive measures are the

first-line treatment when hemoptysis occurs. If these fail, angioembolization is the preferred intervention.

Learning points

1. CT of patients with hemoptysis can demonstrate arterially enhancing lesions in the wall of pulmonary cavities. Differential for these include Rasmussen aneurysms and fistulae.
2. This case demonstrates the difficulty with which bronchial-pulmonary artery fistulae are imaged. 4D CTA is a technique that is increasingly being shown to be useful in characterizing vascular anomalies such as fistulae.
3. Embolization is the mainstay of treatment for bronchial artery to pulmonary artery fistulae with uncontrolled bleeding and has shown satisfactory results in the literature.

Patient consent

Complete written informed consent was obtained from the patient for the publication of this study and accompanying images.

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