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Aortobronchial fistula formation following the rupture of penetrating aortic ulcer causing thoracic aortic pseudoaneurysm: A case report

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Abstract

Background: Aortobronchial Fistula (ABF) with hemoptysis is a rare but life-threatening disease with high mortality. The etiologies mainly include infections, trauma, post-surgery, and it is rarely reported that penetrating aortic ulcer causing thoracic aortic pseudoaneurysm is related to ABF. The purpose of this case report is to stress the importance of early identify and treat penetrating aortic ulcer and thoracic aortic pseudoaneurysm complications to reduce risks of the ABF formation.

Case presentation: Here, we reported a rare case of a 57-year-old man with intermittent cough and hemoptysis presented to the emergency department. A contrast computed tomography angiography revealed ABF formation following the rupture of penetrating aortic ulcer causing thoracic aortic pseudoaneurysm. Because he and his family refused any invasive operation, the patient experienced sudden death 8 hours later after admission.

Conclusions: More attention should be paid to hemoptysis patients with penetrating aortic ulcer causing thoracic aortic pseudoaneurysm and without any significant lung parenchymal disease so as to early identify and treat this scarce but disastrous ABF disease.

Keywords

Aortobronchial fistula; Penetrating aortic ulcer; Thoracic aortic pseudoaneurysm.

Background

Aortobronchial Fistula (ABF) is a rare but life-threatening disease with high mortality. The common symptoms of ABF includes intermittent or massive hemoptysis, dyspnea, hypotension, and even death [1]. Early diagnosis and repair can play a pivotal role in survival and better outcomes. However, in scarce situations, patients may present to the hospital due to atypical symptom and be prone to have delayed diagnosis and mismanagement. In this situation, recognizing the radiologic scenarios with better understanding of the anatomic pathology might be critical.

The etiologies can be degenerative, atherosclerosis, infections, trauma, post-surgery, and post-endovascular aortic repair [2,3]. However, it is rarely reported that penetrating aortic ulcer is related to ABF. We reported that one case of ABF caused by the rupture of penetrating aortic ulcer causing thoracic aortic pseudoaneurysm with intermittent hemoptysis.

Case Presentation

A 57-year-old man with a 10-year history of hypertension presented to the emergency department of an outside hospital with intermittent cough and hemoptysis for 6 hours. Chest contrast-enhanced computed tomography scan showed an intramural hematoma along the posterior wall of aortic arch. Then the patient was transferred to our hospital. On arrival in the emergency room, the patient began to hemoptysis with a total volume of 200 milliliter accompanied with dizziness and fatigue. Physical examination did not find any Marfan's stigmata, and no pulse deficit was revealed. His radial arterial pressure was 144/95 mmHg and heart rate was 131 beats/min, with oxygen saturation 96% on room air. A Contrast Computed Tomography Angiography (CCTA) revealed a 35 mm × 25 mm pseudoaneurysm arising from the proximal of descending thoracic aorta, with peri-aneurysmal hematoma and a 10 × 8 mm consolidation with haziness in the left upper lobe without pleural effusion or pericardial effusion (Figure 1 A,B). What is more, many penetrating aortic ulcers were found in the thoracic aorta CCTA (Figure1 C,D). Further laboratory investigations revealed moderate anemia (HGB 78 g/L) and mildly increased plasma D-dimer (4.65 mg/L). We diagnosed rupture of the penetrating aortic ulcer causing thoracic aortic pseudoaneurysm into the lung, which caused tracheobronchial compression, leading to ABF. A multidisciplinary team including cardiologist, thoracic surgeon, vascular surgeon and anesthesiologist was consulted, and emergency Thoracic Endovascular Aortic Repair (TEVAR) was suggested. However, the patient and his family refused any invasive operation. Unfortunately, the patient experienced sudden death 8 hours later after admission, which was supposed to be caused by rupture of the aortic pseudoaneurysm into the thoracic cavity.

Discussions & Conclusions

ABF is characterized by the development of a communication between the aorta and the bronchial tree. Its main clinical presentation is intermittent or massive hemoptysis, which depends on the characteristics of the fistula [1]. Once diagnosis is established, multidisciplinary approach and dynamic hemodynamic monitor are very important. Besides, emergency intervention is required. Except for traditional open surgical repair, TEVAR is an alternative, less invasive treatment with a lower morbidity and mortality in

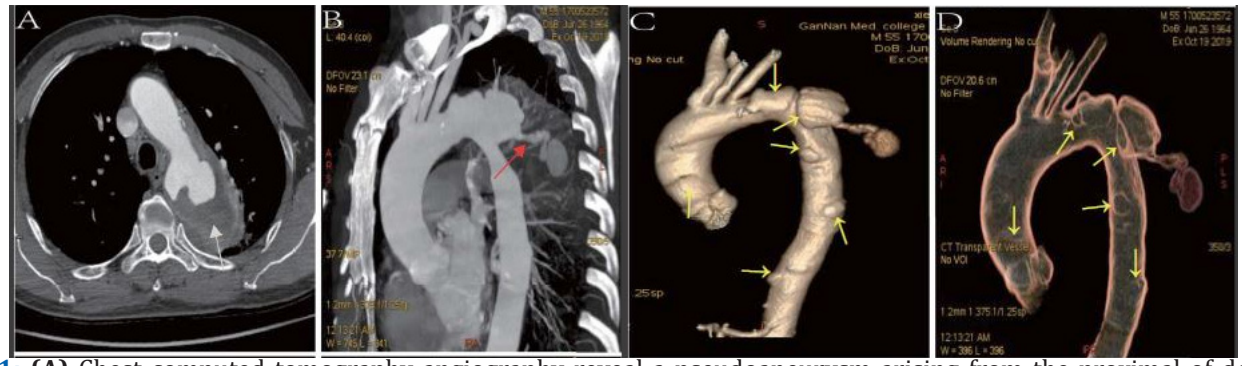

Figure 1: **(A)** Chest computed tomography angiography reveal a pseudoaneurysm arising from the proximal of descending thoracic aorta, with peri-aneurysmal hematoma (white arrow). **(B)** The pseudo-aneurysm cause extrinsic compression of the left upper lung and aortobronchial fistula formation following the rupture of thoracic aortic pseudoaneurysm (red arrow). **(C)** Three-dimensional computed tomography reconstruction(3-DCT) shows thoracic aortic pseudoaneurysm and multiple penetrating aortic ulcers. **(D)** Transparent aorta manifestation of the 3-DCT.

selected patients of ABF. Recent evidence showed TEVAR had good procedural success and favorable short and middle term outcomes [4].

The ABF etiologies mainly include post-surgery and post-endovascular aortic repair [1]. Besides although rare, brucella aortitis was associated with aorto-bronchial fistula [5]. In our case report a contrast computed tomography angiography of aorta showed many penetrating aortic ulcers in thoracic aorta, which indicated penetrating aortic ulcers as his underlying aetiology. However, it is rarely reported that penetrating aortic ulcer is related to aortobronchial fistula. So, more attention should be paid to hemoptysis with penetrating aortic ulcer and without any significant lung parenchymal disease so as to early identify and treat this scarce but disastrous disease. The mechanism in the presented ABF case might be associated with mechanical compression and secondary erosion with increased local inflammatory response by progression of the pseudo-aneurysm [6]. It's a pity that this patient refused to receive surgical or endovascular aortic repair operation and died during the hospital stay.

Declarations

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Consent for publication: We obtained written informed consent from the patient family for this case report publication.

Competing interests: The authors declare that they have no conflicts of interest.

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