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Case Report

A rare presentation of aorto-bronchial fistula and the relevance of open repair in endovascular era

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ABSTRACT

Aorto-bronchial fistula is a rare but potentially fatal condition due to torrential bleeding from erosion of aneurysmal wall into bronchial wall or lung parenchyma. It is usually associated with infection, lung transplantation, pulmonary malignancies, atherosclerotic and mycotic aneurysms, and anastomotic pseudoaneurysms after open or endovascular surgery. The typical presentation usually manifested by haemoptysis, but rarely the patient also can be presented with hematemesis and this will delay the definitive treatment. Surgical or endovascular repair is mandatory because non-operative management will lead to a poor prognosis due to massive bleeding from the fistula. We would like to present a unique case of an elderly man presented with intermittent haematemesis, in which he later developed profound haemoptysis and diagnosed with ABF. Although in the era of endovascular, the ABF was successfully treated via open thoracotomy, resection of thoracic aorta aneurysm, left lower lobe lobectomy, and completed with aortic repair using Gelweave aortic graft.

Keywords: Aorto-bronchial, Aorta, Bronchus, Fistula, Aneurysm

INTRODUCTION

Aorto-bronchial fistula (ABF) is a rare but potentially fatal condition due to torrential bleeding from erosion of aneurysmal wall into bronchial wall or lung parenchyma. It is usually associated with infection, lung transplantation, pulmonary malignancies, atherosclerotic and mycotic aneurysms, and anastomotic pseudoaneurysms after open or endovascular surgery.[1]

Classically, ABF would manifest with life-threatening haemoptysis, but commonly most patients would experience small volume sentinel haemorrhage before progressing to haemoptysis.[2] Eventually, ABF diagnosis can be achieved through CT angiography or bronchoscopy. Surgical or endovascular repair is mandatory because non-operative management will lead to a poor prognosis due to massive bleeding from the fistula.

CASE REPORT

A 64 years old Malay, male with underlying diabetes mellitus, hypertension, with history of partial gastrectomy for bleeding peptic ulcer presented with an intermittent episode of vomiting of blood for two days. On examination, patient was hypotensive and responded to fluid resuscitation. He was in respiratory distress and required non-invasive ventilatory support. Lung

examination revealed reduced air entry at the left lower zone with crepitation. Other clinical examinations were unremarkable. The initial laboratory investigations showed anaemia with haemoglobin reading of 6.5 g/dl. The patient was subsequently transfused 2 pints of packed cell. The chest radiograph showed consolidation at left lower zone and abdominal radiograph was normal. The provisional diagnosis of upper gastrointestinal bleeding (UGIB) was made and an urgent esophagogastroduodenoscopy (OGDS) was performed. The OGDS findings revealed normal mucosa until second part of duodenum. Patient subsequently developed sudden massive haemoptysis with dropping of haemoglobin level to 5 g/dl. Urgent CT angiogram was performed and showed saccular aneurysm measuring 6×5 cm with possibility of ABF. The patient was referred to Cardiothoracic team and underwent operation of left thoracotomy, left lower lobe lobectomy, resection of thoracic aneurysm and repair of the aorta under femoral artery-femoral vein cannulation for heart and lung bypass. Intraoperatively, the aneurysmal sac was adhered to the lung near the hilar region, and it was resected together with left lower lobe.

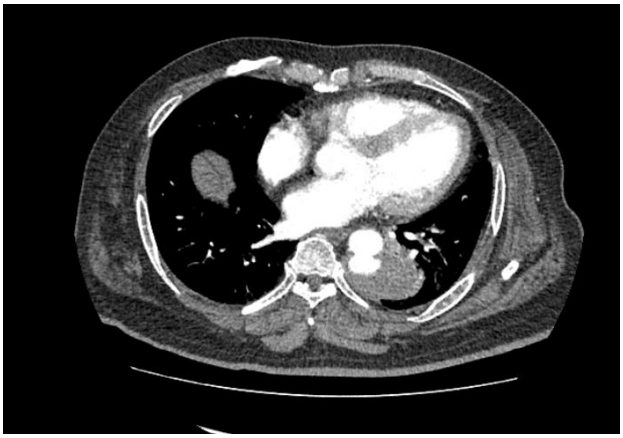

Figure 1: Coronal section of CT scan showing descending thoracic aneurysm with intramural hematoma.

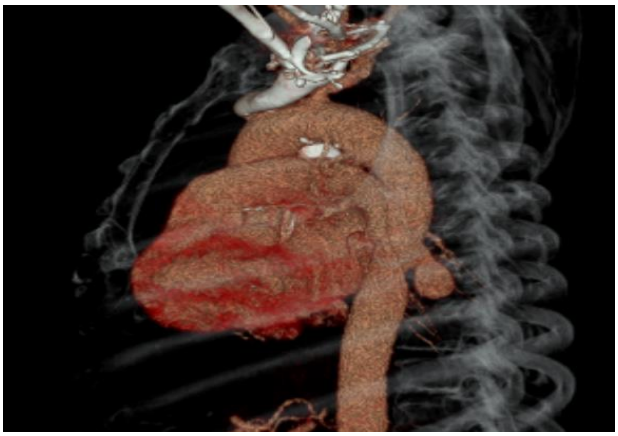

Figure 2: Reconstruction of CT scan showing descending thoracic saccular aneurysm.

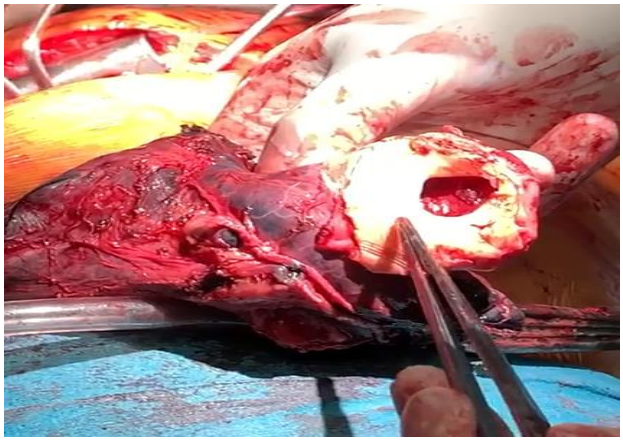

Figure 3: The sac of aneurysm is opened and showing the fistula connection with the lung.

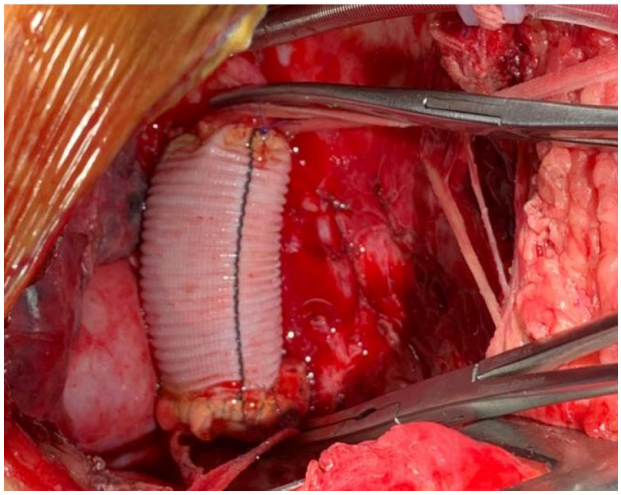

Figure 4: Repair of descending aorta using Gelweave aortic graft 28 mm.

This was followed by repairing of the aorta using Gelweave aortic graft size 28 mm. His post-operative recovery was challenging as this patient developed multiple bouts of lung infections with prolonged ventilation requiring tracheostomy. He was discharged home in good condition eight weeks post operation. Histopathological examination revealed the aneurysm has thickened and degenerated tunica media. The aortic wall was seen in continuity with bronchial wall, lined by respiratory type epithelium with area of squamous metaplasia. There was also chronic inflammation with underlying fibrosis together with former findings suggestive of ABF.

DISCUSSION

ABF overall incidence is unknown and data are only limited to case reports or case series. More than one-half of primary ABFs are caused by thoracic aortic aneurysm. While secondary ABF can be caused by traumatic manipulation during aortic surgery, infections or graft pseudoaneurysms.[2]

For patients presented with haemoptysis, the severity of the haemoptysis depends on the size of the fistula where smaller fistulae would cause mild haemoptysis and maybe briefly occluded by thrombus. This will result in transient symptom resolution. As the fistula is getting bigger or the clot dislodged, haemoptysis will become severe with greater frequency and volume until the underlying condition is treated. Patients may also present with haematemesis during initial period resulting from swallowing expectorated blood.[2] This explains why our case report's initial clinical presentation suggestive of UGIB and managed accordingly followed by OGDS. This mimicry characteristic of ABF misled to a more common diagnosis of UGIB and delays definitive treatment.

ABF diagnosis can be achieved through CT angiography or bronchoscopy. CT angiography findings that might be suggestive of ABF would include aneurysm or pseudoaneurysm of the aorta with localised surrounding ground-glass opacities in the parenchyma of the lung.[3] These findings correlate with the CT angiography of our patient. Bronchoscopy may detect active haemorrhage or stigmata of recent haemorrhage, but it carries the risk of thrombus dislodgment and may not be diagnostic if the fistula is present in the peripheral lung parenchyma.[4] Therefore, CT angiography is generally considered the preferred diagnostic test. However, it should be noted that in patient who presented early with haemoptysis or haematemesis while being haemodynamically stable, CT angiography may not demonstrate active extravasation. But the presence of active extravasation on imaging is indicative of a worse prognosis.[5]

Surgical or endovascular repair is imperative because non-operative management is universally fatal.[4] Open repair has a high mortality rate, reported at 15-41%. In comparison with thoracic aortic stent-graft placement via endovascular repair with or without subsequent lung tissue repair has at least similar rates but generally better outcomes.[6] Although endovascular surgery alone may be acceptable, studies has shown that there is a reduced risk of fistula recurrence with subsequent pulmonary resection, and the stent graft is covered with intercostal interposition muscle flap.[7]

CONCLUSION

The case presented here demonstrates several important clinical pearls. Patient with ABF may experience intermittent but progressively worsening haematemesis or haemoptysis over days because thrombus can temporarily occlude small ABF communications in the initial course of the disease. Clinicians should have high index of suspicions for ABF for patients with thoracic aortic aneurysm with history of haemoptysis even though CT angiography may not show evidence of active extravasation of contrast. Despite the current shift towards endovascular repair owing to its overall better outcome, there is still a role of open surgical repair for ABF especially in emergency settings where there is a need for simultaneous haemostasis, repair, and lung resection, and when there is no endovascular service available.

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