

Incidental Bronchial Artery Aneurysm with Associated Bronchial Artery to Pulmonary Venous Fistula: Treatment By Embolization

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ABSTRACT

BACKGROUND: Bronchial artery aneurysm (BAA) is a rare vascular phenomenon with potentially life-threatening complications, especially since most cases are asymptomatic. Symptoms may vary depending on the aneurysm's size and position.

CASE PRESENTATION: Here we present the case of a 40 year old healthy gentleman who developed right sided chest pain following an exercise stress test for insurance purposes. CT of the chest showed an aneurysmal accessory artery arising from the thoracic aorta. Focused CT of the thoracic aorta showed a 21 mm arterial aneurysm posterior to the left main bronchus with a long, tortuous efferent forming a fistula with the pulmonary venous circulation. The patient underwent prophylactic embolization and coiling of the aneurysm. Complete occlusion was confirmed on angiography.

DISCUSSION: Size of bronchial artery aneurysm does not appear to be related to risk of rupture. Many are asymptomatic or present with rupture and haemorrhagic shock. BAAs should be treated immediately regardless of size and symptoms. Embolization is an acceptable treatment modality as it is minimally invasive however treatment is complex when concomitant fistula is present, especially in those with fistulation to the pulmonary venous circulation, due to risk of emboli entering the systemic circulation.

Key Words: Bronchial artery aneurysm, BAA, bronchial artery to pulmonary venous fistula, BA-PV, embolization

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Abbreviations and Units

BAA: Bronchial artery aneurysm; BA-PA: Bronchial artery to pulmonary artery; BA-PV: Bronchial artery to pulmonary vein; BP: blood pressure; CT: Computer tomography; ECG: Electrocardiography; EST: Exercise stress test; LRTI: Lower respiratory tract infection; mm: Millimetre; mmHg: Millimetres of mercury; VATS: Video-assisted thoracoscopy.

CASE REPORT

Mr RP, a 40 year old gentleman, was referred for an exercise stress test for insurance purposes, which was normal at high workload.

The day after the test he presented with right sided chest pain, radiating to the upper limb. The patient also gave a history of long-standing dry cough. BP 140/80 mmHg; ECG was normal sinus rhythm at 67 beats per minute. He was admitted with a working diagnosis of a chest infection and CT thorax was performed. This showed an aneurysmal accessory artery arising from the thoracic aorta.

The patient also underwent echocardiography, which showed normal left ventricular dimensions, normal ejection fraction and tissue doppler imaging. Trace mitral insufficiency was seen. Figure 1 shows the aneurysmal accessory artery arising from the descending thoracic aorta.

CT thoracic aorta was performed to further characterize the findings on CT thorax - a 21mm × 17mm arterial aneurysm was evident posterior to the left main bronchus, anterior to the descending thoracic aorta and lateral to the oesophagus (Figure 1). The afferent supply from the descending thoracic was also aneurysmal, however it also had a stenosed origin. The efferent was long, tortuous and divided into several small branches which fistulated with the pulmonary venous circulation, particularly the right upper lobe pulmonary vein. The aneurysm was also in communication with the inferior surface surface of the aortic arch.

The patient underwent embolization of the aneurysm two months later using a right femoral artery approach. Angiography revealed a hypertrophied, tortuous right bronchial artery with a large aneurysm of the middle third - this had increased in size compared to the measurements on CT. Selective catheterization using a Direxion catheter and GlideGold microwire was carried out. The efferent vessel was then embolized whilst the aneurysm itself was densely packed with metallic coils. Repeat angiography showed complete occlusion of the aneurysm and repeat CT at one month follow up confirmed this. Figure 2 shows the aneurysm full of contrast at angiography whilst figure 3 shows the aneurysm after coiling.

DISCUSSION

Bronchial artery aneurysms (BAAs) are rare vascular abnormalities. Diagnosis by selective bronchial angiography occurs in less than 1% of cases^[1]. BAA with concomitant fistula is even more uncommon^[2].

BAA may be intrapulmonary, mediastinal or both. Symptoms depend on size, location and comorbidities. They are usually asymptomatic and may present with haemodynamic instability following rupture. Intrapulmonary BAAs typically present with haemoptysis^[2,3]. Mediastinal BAAs tend to present with symptoms of compression or fistulation into surrounding structures e.g. dysphagia and haematemesis respectively^[3].

Little is known about the aetiology, although it is considered to be caused by weakening of the vessel wall, usually in the context of an inflammatory process and increased blood flow to the lungs^[4,5]. It is possible that fistula formation occurs to relieve pressure and reduce risk of rupture^[2].

Embolization is a well-established modality, minimally invasive, with shorter hospital stays^[6]. Catheter angiography is also useful as CT alone may not identify fistulae. Embolization in BA-PV has a theoretical risk of emboli or the coil migrating to the systemic circulation^[7]. This has been documented in 3% of cases done for PA-PV fistula but there is no data for BA-PV fistula. The most common complaint post-procedure is pleuritic chest pain. Several materials are available to perform embolization. Coils are useful in fistula with aneurysm but because they can cause an overly proximal occlusion, their use precludes re-intervention^[7].

Size of BAA does not appear to be related to risk of rupture and because of the catastrophic complications it is recommended that they are treated on identification, with embolization being the treatment of choice, however multidisciplinary discussion regarding therapeutic modality. Surgical approaches include open, via thoracotomy or by video-assisted thoracoscopy (VATS). Surgical backup should be present when doing embolization in the event that intra-procedural

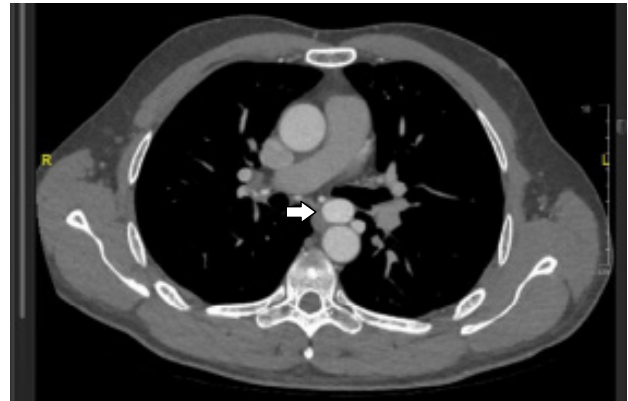


Figure 1 Axial CT of the thoracic aorta after intravenous contrast administration, showing the aneurysmal accessory artery arising from the descending thoracic aorta (white arrow).



Figure 2 Appearance of aneurysm at angiography showing the afferent and efferent vessels.

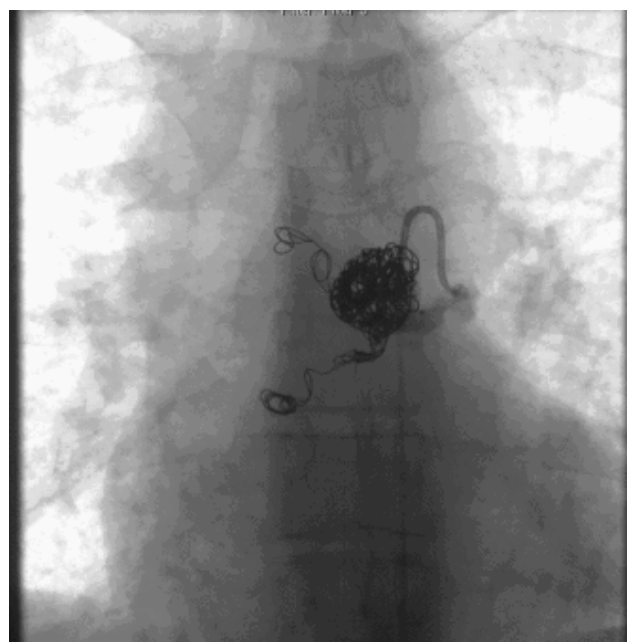


Figure 3 Bronchial aneurysm after coiling.

complications occur. Discussion with the patient about the risks and benefits of each is also recommended^[2,3].

Learning Points

(1) Rarity of BAA, in particular those associated with fistulae to the pulmonary circulation; (2) BAA is an important condition as it is usually asymptomatic and can present with haemorrhagic shock and haemodynamic instability; (3) Despite this, little is known about its aetiology, particularly with respect to associated fistulation; (4) BAA with BA to PV is particularly challenging to treat with embolization due to the risk of migration of embolizing material or emboli to the systemic circulation.

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