Endovascular management of an acquired aortobronchial fistula following aortic bypass for coarctation

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Abstract

Aortobronchial fistula (ABF) in the setting of aortic coarctation repair is very rare but uniformly fatal if untreated. Endovascular stenting of the descending aorta is now the first-choice approach for ABF presenting with haemoptysis and offers a less-invasive technique with improved outcomes, compared with open repair. We report a case of late ABF occurring following bypass for aortic coarctation. Management focused on two key manoeuvres: use of a covered endovascular stent to occlude the aortic bypass thus controlling the fistula and dilatation and stenting of native coarctation.

Keywords: Aorta • Endovascular • Fistula • Coarctation

CASE REPORT

A 46-year old man was referred from another institution following an episode of massive haemoptysis requiring immediate transfusion. He underwent aortic coarctation as a neonate with subsequent revision and an aortic bypass performed using a Dacron graft at the age of 13 for restenosis. He experienced three smaller episodes of haemoptysis over the preceding 12 months which were attributed to haematemesis in another institution and investigated with upper gastrointestinal endoscopy. ABF had not been considered.

Upon arrival, he was complaining of severe central chest pain and shortness of breath. He was hypertensive and tachycardiac. Haematological investigations revealed haemoglobin of 8.8 g/dl and type I respiratory failure. Septic parameters were normal. Resuscitation was commenced.

A computed tomography angiogram (CTA) was performed, which demonstrated multifocal, diffuse ground-glass changes consistent with pulmonary haemorrhage predominantly in the left upper lobe (Fig. 1A). CTA also demonstrated a coarctation of the aorta distal to the left subclavian artery, with a surgical graft bypassing the coarctation. The graft extended from the distal arch of the aorta to the distal descending thoracic aorta, and the left subclavian artery was reimplanted into the proximal bypass graft. There was an aneurysmal dilatation at the site of the reimplanted left subclavian artery.

The patient was transferred to a high-dependency unit for stabilization. He was commenced on broad-spectrum antibiotic prophylaxis, which was continued into the postoperative period. A diagnostic angiogram was performed to more clearly delineate the anatomy and further assess the pseudoaneurysm (Fig. 1B). There was significant stenosis at the origin of the left subclavian artery.

An endovascular approach was agreed upon. Initially, primary stenting through the bypass graft with a covered stent was considered. The angles were considered unfavourable at either end of the bypass graft to safely land the stent with good anatomical coverage additionally, the risk of the stent kinking was considered high.

Under general anaesthesia, the right common femoral artery (CFA) was surgically dissected and controlled. On the contralateral side, a 6-Fr sheath was also percutaneously placed in the left CFA under ultrasound guidance.

Via the left CFA, first a 6-mm Amplatzer 2 plug was deployed in the proximal left subclavian artery (Fig. 2A). Subsequently, following intra-arterial systemic heparinization, via the right CFA, a 45-mm covered metal stent (CP) was deployed across the coarctation and expanded with a balloon (Mullins X) with good angiographic expansion of the coarctation (Fig. 2B and C). Following this, a 22-Fr, 28-mm covered thoracic stent (Valiant Medtronic) was deployed proximal to the left common carotid artery and distal to the lower end of the bypass graft. Post-thoracic stent angiogram demonstrated good stent positioning with complete exclusion of the surgical bypass graft (Fig. 2D).

The patient recovered well and CTA 1 week later demonstrated good stent positioning with no endoleak and no contrast seen in the bypass graft. There was no evidence of sepsis throughout the post-procedural period and prophylactic broad spectrum antibiotic therapy was continued until discharge. The patient was advised to continue this but declined.

DISCUSSION

This case is best summarized as an ABF at the site of a left subclavian artery aneurysm following complex aortic coarctation repair in childhood, whereby the original coarctation and subsequent ABF were managed using endovascular techniques. Our patient presented in the usual manner for an ABF with atypical...
chest pain and dyspnoea, followed by sentinel bleeds leading to a life-threatening haemorrhage [1]. In the setting of previous thoracic aortic surgery, suspicion of ABF should have been high. We opted to perform contrast-enhanced CT, which is the gold standard diagnostic tool as more invasive methods (e.g. bronchoscopy) have been known to result in blood clot instability and substantial blood loss in this setting [2]. CT can detect a pseudoaneurysm, periaortic haematoma and consolidation in the adjacent pulmonary tissues; however, visualization of the aortobronchial fistula (ABF) is possible only in 17% of cases and although the other
radiographic features were highly suggestive, visualization of the ABF itself was not possible in this case [3].

This case demanded extensive pre-procedural planning. The potential for sepsis as a cause of fistulation was a consideration throughout management and for that reason prophylactic antibiotic therapy was instituted early despite normal haematological septic parameters and the absence of fever. While we considered open surgical repair, on balance we felt that the benefits offered by an endovascular approach were superior and after full explanation of the options was preferred by the patient.

The use of endovascular stenting for ABF is relatively new and the largest published case series consists of 26 patients [4]. There were 4 in-hospital mortalities, but overall the technique was well tolerated with minimal risk and deemed the likely first-choice treatment for ABF presenting with haemoptysis, considering that conventional open surgery in ABF is associated with mortality rates as high as 24% and considerable morbidity primarily due to pulmonary infection, prolonged ventilation and neurological sequelae. The use of endovascular techniques is not appropriate in all cases; however, a case report of life-threatening sepsis developing at an interval post-endovascular treatment of ABF serves as a reminder that stenting may not adequately manage ABF secondary to sepsis and can result in poor outcomes [5].

CONCLUSION

This is a presentation of ABF in a patient with an uncommonly seen aortic coarctation repair and complex surgical history. Haemoptysis in the setting of previous aortic surgery should be investigated and managed with emergency, as it typically heralds a life-threatening bleed and CT angiography is the investigative modality of choice.

Conflict of interest: none declared.

REFERENCES

[1] Hacker S, Langenberger H, Plank C, Gorlitzer M, Ehrlich M, Dolak W. Parametrical stenting of an acquired aortobronchial fistula (ABF) using endovascular technology [1]. The Author 2013. Published by Oxford University Press on behalf of the European Association for Cardio-Thoracic Surgery. All rights reserved.


eComment. Late complications after aortic coarctation repair

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We have read the article by O’Sullivan et al. and we would like to add a comment on the complications of this pathological entity [1].

Aortic coarctation (AC) carries an incidence of 4 per 10 000 live births (5-8% of all congenital heart disease) and it can occur in association with patent ductus arteriosus, ventricular septal defect, bicuspid aortic valve (present in nearly two-thirds of patients with coarctation), concomitant hypoplastic aortic arch as well as parachute mitral valve stenosis, circle of Willis cerebral aneurysms and other congenital heart diseases (transposition of the great vessels, Tausig-Bing anomaly, hypoplastic left heart syndrome etc). Finally, AC is the commonest cardiac anomaly present in females with Turner syndrome [2, 3].

More importantly, the mean survival without treatment is 35 years and the vast majority of patients (90%) die before their 50th [2].

The choice of treatment has to be decided by a multi disciplinary team meeting with relevant specialists [Recommendation Class I, Level of Evidence C] [3].

Despite successful outcomes of its surgical treatment, late survival is compromised. For patients who are operated at a mean age of 16 years, the 10-, 20- and 30-year survival rates are 91%, 84% and 72% respectively [2].

Late complications can be subdivided into 8 categories namely: (i) re-coarctation, (ii) aortic aneurysm formation/aortic dissection, (iii) related to coexisting bicuspid