Endovascular treatment of thoracic aortic pseudoaneurysm with aortobronchial fistula in the late postoperative period of surgical correction of the aortic coarctation

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Abstract

Aortobronchial fistula is an abnormal passage between the aorta and the bronchus, and even when recognized and treated promptly, it carries a high risk of fatality. It can develop after aortic operations, and it is usually the result of a pseudoaneurysm. Massive or intermittent hemoptysis is the main symptom. Conventional treatment of aortobronchial fistula is open surgery of the thoracic aorta with tracheobronchial reconstruction. Recently, endovascular repair has been proposed as an alternative. The authors report a case of successful endovascular treatment of thoracic aortic pseudoaneurysm with aortobronchial fistula, 22 years after surgical correction of the aortic coarctation.

Keywords: Aorta; heart defects, congenital; fistula; hemoptysis; aneurysm, false.

Introduction

Aortobronchial fistula is a connection between the aorta and the bronchus that can cause severe and fatal hemoptysis. Mortality is high because of massive hemoptysis and because of the difficulty of diagnosis, since only in very rare cases can the fistula be visualized on imaging studies.[1,2] It may develop from an aneurysm of the descending thoracic aorta, after trauma, or it may arise as a surgical sequela in the repair of congenital heart defects.[3] Even when promptly recognized and treated, aortobronchial fistula carries a high risk of death. Some treatment alternatives have been reported recently, including extra-anatomic bypass and cryopreserved homograft, but little is known about the subject because of its low frequency.[4] The authors present a case report of successful endovascular treatment of a thoracic

aortic pseudoaneurysm with aortobronchial fistula in the late postoperative period of surgery to correct aortic coarctation.

Case report

The patient was a 38-year-old woman with long-standing hypertension. At 16 years of age, she had undergone surgery to correct aortic coarctation by placement of an extra-anatomic bypass with a synthetic Dacron graft. The bypass had its proximal, end-to-side anastomosis at the juxtasubclavian left portion; distally, the anastomosis was end-to-side and was located in the middle third of the descending aorta.

Twenty-two years after the first operation, the patient had intense precordial pain for two days, radiating to the back and relieved by rest. From that episode on, she began to hear a wheezing sound during inspiration, which worsened in the supine position. Three months later, she was admitted to the emergency department with intense precordial pain radiating to the back, severe dyspnea, hypotension and cough accompanied by large-volume hemoptysis. The patient denies any history of trauma.

During hospitalization, computed tomography showed the presence of an aortic pseudoaneurysm (Figure 1). The aortobronchial fistula was not visualized on the examination, but the history of surgery to correct aortic coarctation 22 years earlier, the presence of a pseudoaneurysm, the precordial pain and the massive hemoptysis suggested the presence of a sealed aortobronchial fistula.[2] The diagnoses of pulmonary thromboembolism, pneumonia and lung abscess were ruled out, and emergency endovascular treatment was indicated.

Endovascular exclusion of the thoracic aortic pseudoaneurysm, extending from the juxtasubclavian left portion to the distal third of the thoracic aorta, at the level of T10, was performed with placement of two endografts, Zenith TX2® Z TEG 28-28-140 mm and 28-24-115 mm (Cook Incorporated, Bloomington, Ind), telescoped retrogradely through the right common femoral artery. The "clothesline" technique was used, with a 0.14” guidewire introduced through the right brachial artery and exteriorized at the right common femoral artery, because of the technical difficulty of primary retrograde catheterization, thus maintaining the stability and firmness needed to support the endograft. Angiographic control confirmed a patent aorta along its entire length with exclusion of the pseudoaneurysm and no leaks (Figure 2).

At follow-up after three months, the patient brought the result of computed tomography angiography of the thoracic aorta, showing a patent endograft with no signs of contrast extravasation. A small focal dissection of the abdominal aorta was noted slightly proximal to the origin of the celiac trunk, with displacement of an intimal flap from the posterior wall of the aorta, without thrombi (Figures 3 and 4). The celiac trunk, superior mesenteric artery and renal arteries were patent, without significant stenoses or evident aneurysmal dilatations. The patient was in good general condition, without complaints, denying abdominal pain, weight loss or change in bowel habits.

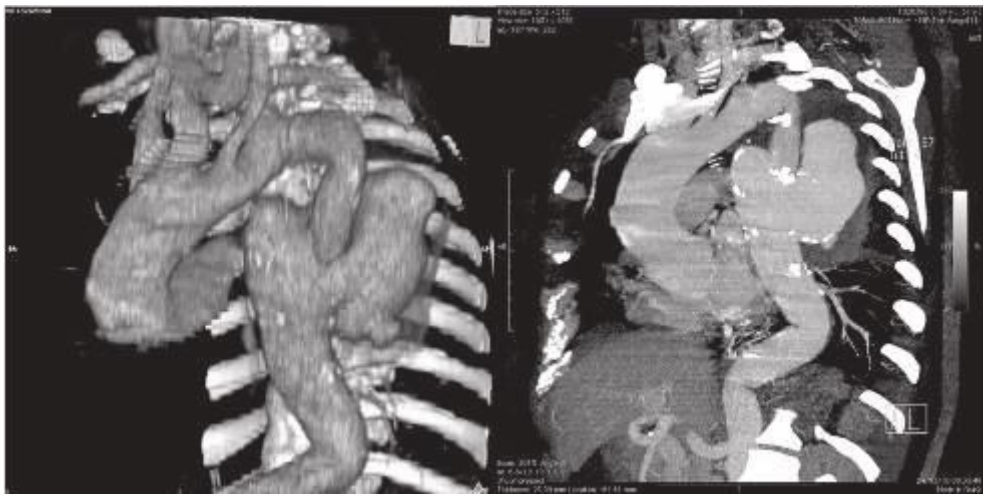


Figure 1. Three-dimensional volume reconstruction and multiplanar reconstruction showing a large pseudoaneurysm of the thoracic aorta, with contrast extravasation mainly at the distal anastomosis.

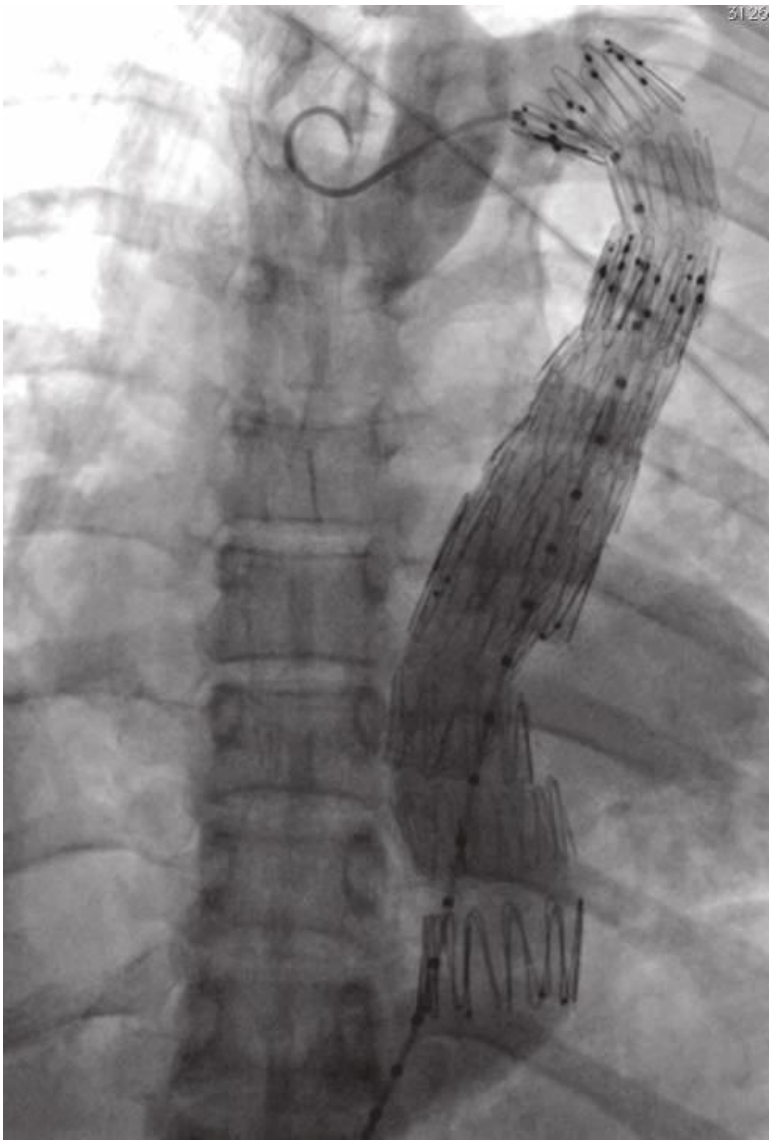


Figure 2. Intraoperative arteriography after deployment of the two endografts, showing patency and absence of leaks.

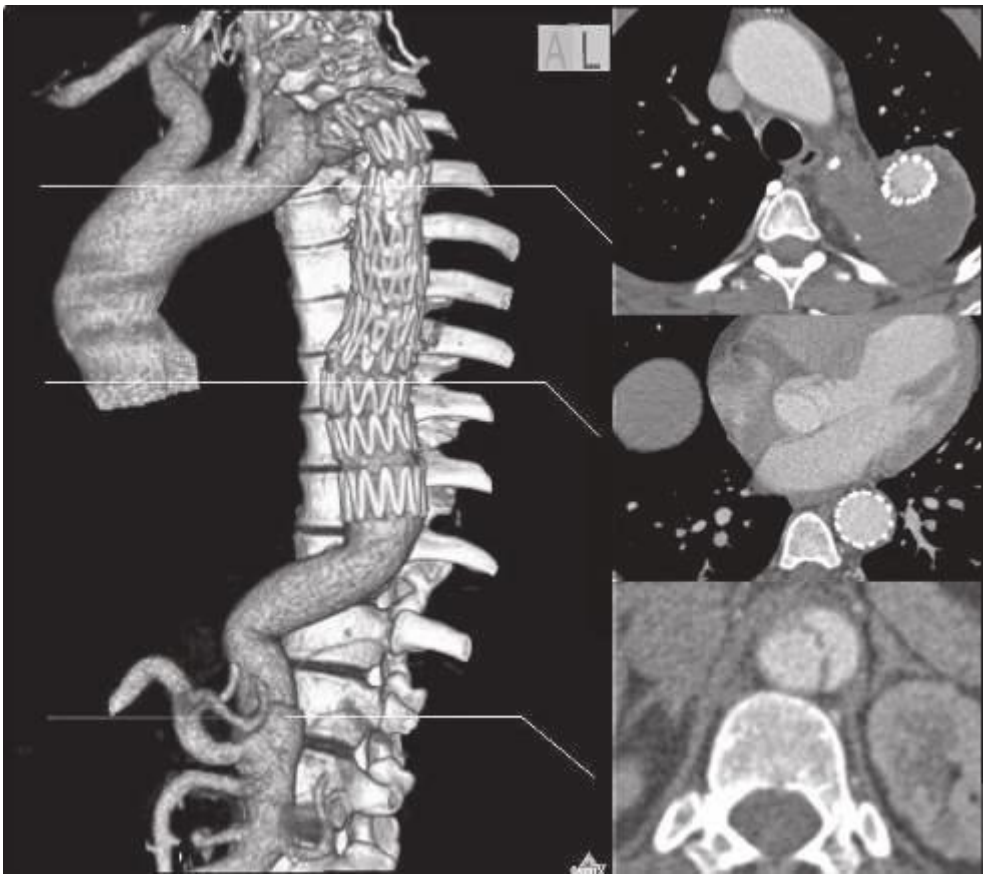


Figure 3. Three-dimensional volume reconstruction and key axial images of control computed tomography angiography: patency of the endografts and absence of leaks. Image consistent with focal dissection of the abdominal aorta slightly proximal to the origin of the celiac trunk, with displacement of an intimal flap from the posterior wall of the aorta, without thrombi.

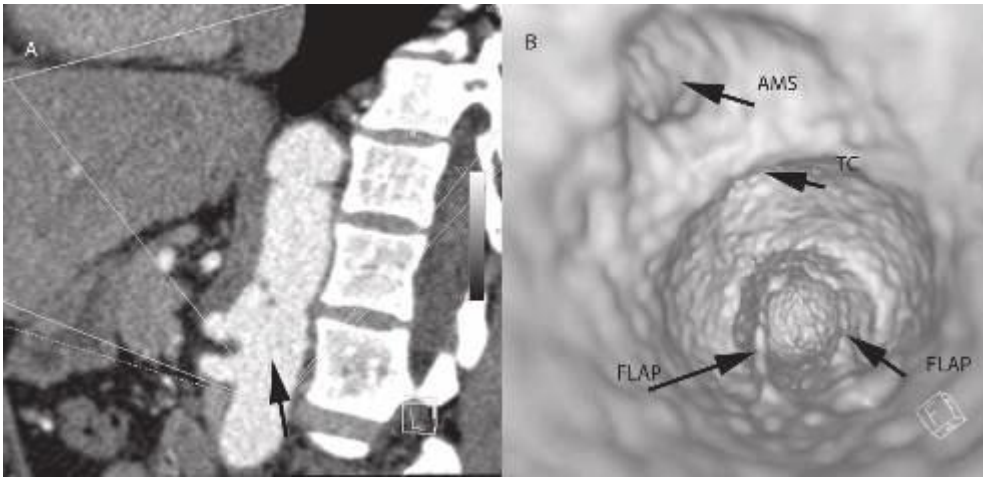


Figure 4. (A) The arrow shows, on the multiplanar reconstruction, the position of the camera in the reconstruction of image B. (B) Three-dimensional reconstruction of the internal view of the intimal flap, showing patency of the ostia of the superior mesenteric artery (AMS) and celiac trunk (TC).

Discussion

Aortobronchial fistulas may develop at an indeterminate time after aortic surgery and are usually a consequence of a pseudoaneurysm. Hemoptysis, massive or intermittent, is the main presenting symptom.[5,6] Different diagnostic modalities can visualize the tract of an aortobronchial fistula, but this visualization is achieved only in very rare cases,[2] as in the present case, in which the fistula was not

visualized on computed tomography. Hemoptysis is a sign that is present in countless diseases. For this reason, the clinical history is extremely important for the differential diagnosis. In the presence of hemoptysis, previous aortic surgery is highly suggestive of an aortobronchial fistula, especially when accompanied by a pseudoaneurysm.[2]

The conventional treatment of aortobronchial fistula is open surgery of the thoracic aorta with tracheobronchial reconstruction. Although some alternative strategies have been reported in the literature, including the use of extra-anatomic bypass and repair with a cryopreserved homograft, there is no consensus on the best type of treatment. The mortality rate of open surgery is related to the complications of hemorrhage and sepsis, which reach 78%. Endovascular repair of the thoracic aorta has been proposed as an alternative, because it is a less invasive procedure. However, the endovascular technique has limitations in the treatment of aortobronchial fistula, mainly because of the high risk of contamination of the prosthesis.[4]

Treatment of the thoracic aorta with endovascular prostheses has significantly reduced the morbidity associated with thoracotomy and aortic cross-clamping and has also reduced in-hospital mortality and postoperative morbidity; recent studies show encouraging mid-term results.[7] By analogy with cases of traumatic thoracic aortic pseudoaneurysm, in which open surgery still has high mortality rates, recent reports of success place the endovascular technique as a possible substitute for conventional treatment.[8] Despite the use of endovascular techniques, the rate of spinal cord ischemia, which results in paraparesis and/or paraplegia, remains unchanged when compared with open surgery.[7]

Little is known about the late postoperative results of the use of endografts in the treatment of aortobronchial fistulas. This is due to the low incidence, the relatively recent introduction of endovascular techniques and the scarcity of studies on the subject in the literature.[4]

Conclusion

This report shows the feasibility of the endovascular technique in the treatment of thoracic aortic pseudoaneurysm with aortobronchial fistula in the late postoperative period of surgery to correct aortic coarctation.

Declarations

Study conducted at Universidade de Santo Amaro (Unisa), São Paulo (SP), Brazil. Submitted: September 7, 2010. Accepted: January 21, 2011.

Author contributions. Study conception and design: ACMA, AAGN. Data analysis and interpretation: ACMA. Data collection: AAGN, FCPM, AGNMO, RTB. Writing of the article: ACMA, AAGN. Critical revision of the text: ACMA, RVS, RTB. Final approval of the article*: ACMA, AAGN, FCPM, RTB. Statistical analysis: ACMA, AAGN. Overall responsibility for the study: ACMA, RVS, RTB. Funding information: N/A.

*All authors read and approved the final version submitted to J Vasc Bras.

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