Case Reports

Endovascular Exclusion of Aortobronchial Fistula

and Distal Anastomotic Aneurysm after Extra-Anatomic Bypass for Aortic Coarctation

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The treatment of choice for aortic coarctation in adults remains open surgical repair. Aortobronchial fistula is a rare but potentially fatal late sequela of surgical correction of isthmic aortic coarctation via the interposition of a graft.

The endovascular treatment of aortobronchial fistula is still under discussion because of its high risk for infection, especially if the patient has a history of cardiovascular prosthetic implantation. Patients need close monitoring, most notably those with secondary aortobronchial fistula. We discuss the case of a 65-year-old man who presented with the combined conditions, and we briefly review the relevant medical literature. ***(Tex Heart Inst J 2017;44(1):55-7)***

Key words: *Aorta, thoracic/abnormalities/radiography/surgery; aortic aneurysm/etiology; aortic coarctation/complications/diagnosis; endovascular procedures/instrumentation; fistula, aortobronchial; postoperative complications; treatment outcome*

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When aortic coarctation (AC) presents during adulthood, it most often is a case of overlooked native coarctation or of recurrent coarctation. Traditional treatment consists of open surgical repair. A rare but life-threatening late sequela of graft interposition for AC is aortobronchial fistula (ABF). Most ABFs that follow AC correction develop from an anastomotic aneurysm of the descending thoracic aorta (DTA). In recent years, the incidence of ABF secondary to surgical or endovascular repair of pathologic aortic conditions has increased roughly in accordance with the increase in aortic interventions. Thoracic endovascular aneurysm repair (TEVAR) of ABF is still under discussion, mainly because of the risk of graft infection. We describe our treatment of a patient who had AC, the correction of which progressed to ABF; and we place his case within the context of the relevant medical literature.

Case Report

In August 2014, a 65-year-old man presented at our emergency department with hemoptysis. His medical history was consistent with hypertension in medical treatment (atenolol 100 mg/d and amlodipine besolate 10 mg/d). He also had right hemiparesis, secondary to infantile cerebral palsy, and an extra-anatomic 20-mm Dacron graft placed 17 years earlier (as a bypass from the subclavian artery to the DTA for isthmic AC). Clinical examination showed normal vital signs and no fever. Anemia and leukocytosis were absent. A chest radiograph aroused suspicion of a DTA aneurysm. Computed tomographic (CT) scans confirmed the presence of a 21-mm distal anastomotic aneurysm of the extra-anatomic bypass (Figs. 1 and 2), associated with a "ground-glass" opacity at the level of the left inferior pulmonary lobe compatible with recent intra-alveolar bleeding. The patient was transferred to our department because an ABF was suspected.

Because of the high operative risk, an endovascular graft exclusion was planned as a bridge solution. Institutional Review Board approval was obtained. After giving his written informed consent, the patient underwent general anesthesia with cerebrospinal fluid drainage because of the risk of paraplegia associated with the procedure: in fact, the need of AC embolization to avoid a type 2 endoleak was unclear preoperatively.

Intraoperative bronchoscopy revealed blood at the level of the left inferior bronchus. Through bilateral femoral surgical access, 2 Lunderquist® extra-stiff guidewires (Cook Medical Inc.; Bloomington, Ind) were placed: one in the left subclavian artery through the bypass, the other in the aortic arch through the AC. A 30 × 30 × 150-mm Valiant® Captivia® aortic stent-graft (Medtronic, Inc.; Minneapolis, Minn) was in-

troduced through the left common femoral artery and deployed between the aortic bypass and DTA to exclude the aneurysm. Angiographic monitoring revealed the perfusion of the aneurysmal sac from the native aorta (Fig. 3) and a peak-to-peak transcoarctation pressure gradient <20 mmHg. Therefore, through left humeral access, we occluded the DTA at the level of the AC by means of a 10-mm AMPLATZER™ 2 Vascular Plug II (St. Jude Medical, Inc.; St. Paul, Minn). The completion angiogram showed the total exclusion of the anastomotic aneurysm, and this was later confirmed by a postoperative CT scan (Fig. 4).

The patient's postoperative course was uneventful, and he was discharged from the hospital on the 5th postoperative day with instructions to take long-term antibiotic agents, with the prospect of a later complete surgical repair in a sterilized field. Fifteen months later, CT revealed no signs of infection, a minimal type 2 endoleak, and a reduction in the diameter of the aneurysmal sac.

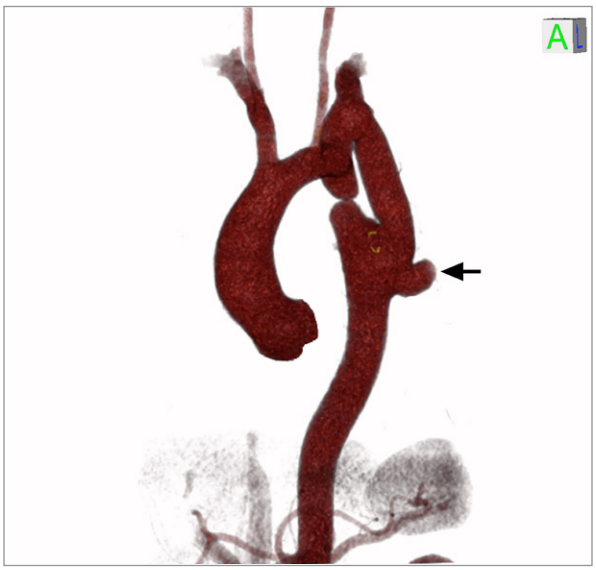

Fig. 1 *Computed tomogram (3-dimensional reconstruction) shows a distal anastomotic aneurysm (arrow) of the Dacron extra-anatomic bypass between the left subclavian artery and the descending thoracic aorta.*

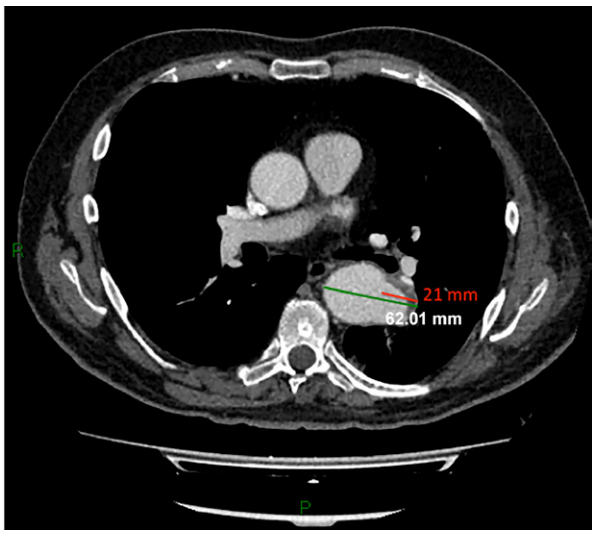

Fig. 2 *Preoperative computed tomographic angiogram shows the transverse aortic diameter (21 mm) at the level of the distal anastomotic aneurysm. The green line indicates the aortic diameter (62.01 mm).*

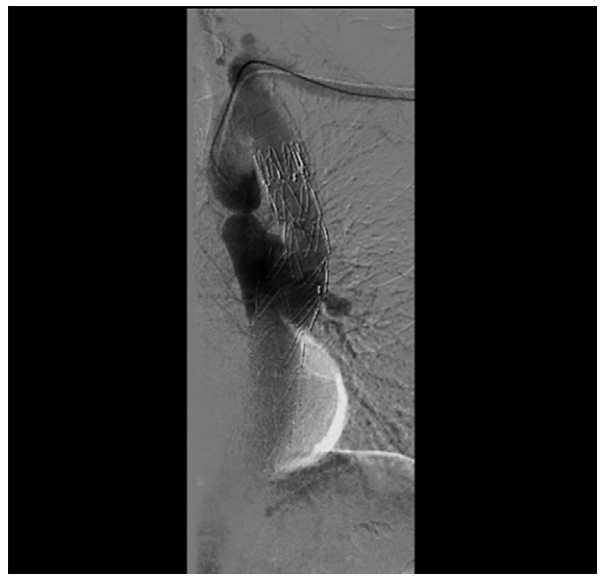

Fig. 3 *Intraoperative aortogram shows the aneurysmal sac's persistent perfusion after thoracic endovascular aneurysm repair.*

Fig. 4 *Postoperative computed tomogram (3-dimensional reconstruction) shows complete exclusion of the distal anastomotic aneurysm and embolization of the aortic coarctation with a vascular plug.*

Discussion

Surgical repair of AC was first performed in 1944 by Crafoord,[1] and various subsequent techniques were developed, such as resection with end-to-end anastomosis, subclavian flap aortoplasty, bypass grafting across the area of coarctation, and prosthetic patch aortoplasty. In adults, Dacron bypass grafting is the usual solution: it can be used when the AC is too complex for a graft-conduit interposition, when the collateral vessels are extensive, or when other significant intracardiac or aortic abnormalities must be treated. The reported rate for repeat intervention after surgical correction of AC ranges between 4% and 20% (average, ~10%).[2] Repeat coarctation is an important long-term sequela that occurs in up to 9% of patients. Another long-term sequela after surgical repair is aneurysm formation (prevalence, 5%–13%).[3] Aortoesophageal and aortobronchial fistulae are late sequelae of aortic bypass for AC.[4] Aortobronchial fistula presents with hemoptysis and can appear from 3 weeks to 25 years after surgery. Diagnosis is difficult and clinical suspicion is often unconfirmed. Treatment is almost always surgical and must be performed immediately because of the risk of fatal massive hemoptysis. Thoracic endovascular aneurysm repair is a less invasive (albeit less effective) possibility. Probably, the advantages of endovascular treatment of ABF are safer and more rapid treatment of unstable patients, who might later undergo definitive open operation in a more stable elective setting, with less associated risk of morbidity and death. In our experience, no intraoperative complications or deaths occurred and no signs of infection were detected during follow-up evaluation.[5-7] We think that the low rate of stent-graft infection probably was linked to minimal tissue trauma and to our placement of the stent-graft in the center of the aneurysmal sac, well away from the ABF and source of contamination. We have observed only one previous patient with ABF secondary to previous thoracic cardiovascular surgery,[5] and we think that in his case TEVAR was a life-saving but bridge solution, because of possible infection of the previously implanted aortic prostheses.

Although many recent reviews indicate that TEVAR is the treatment of choice for ABF, we conclude that severe sequelae can occur and that such patients need close monitoring—especially those with secondary ABF.

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