

Late-onset aorto-esophageal fistula after treatment of a chronic type B aortic dissection with a three-step approach

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ABSTRACT

Aorto-esophageal fistula is a rare but lethal complication after thoracic endovascular repair for thoracic aortic diseases. Extensive treatment is reserved for patients fit for surgery. Various technical approaches have been described; however, mortality rates are still high. Herein, we report a case of a 76-year-old woman with aorto-esophageal fistula treated by a three-step treatment approach, with close collaboration between cardiothoracic and general surgery specialists. The patient required tracheostomy after the first procedure, but this was closed at 15 days. She subsequently recovered and is doing well at 3 months after surgery. Staged treatment aims to shorten operative times, to reduce the risk of anesthesia complications, and to provide the patients the time to recover after each procedure. (*J Vasc Surg Cases and Innovative Techniques* 2018;4:50-3.)

Endovascular treatment of thoracic aortic diseases is the “gold standard” today. These procedures are often performed rapidly, with low complication rates. Nevertheless, adverse events are possible, and among these, the development of an aorto-esophageal fistula (AEF) is a rare but lethal complication.^{1,2} Several reports described different technical approaches to treat this complication; however, mortality rates are still high. We report a case of a 76-year-old woman who was treated with a three-step treatment approach. The patient gave consent for publication of this case.

CASE REPORT

A 76-year-old woman (weight, 72 kg; height, 168 cm) developed late-onset AEF 2 years after treatment of a chronic Stanford type B aortic dissection with progressive growth of the maximal aortic diameter (first diagnosis, January 2012; treatment, April 2014). A thoracic stent graft (Cook Medical, Bloomington, Ind) was implanted to cover the intimal tear located in Ishimaru zone 3. Because of covering of the left carotid artery, an in situ fenestration of the stent graft was necessary, with deployment of a bare-metal stent (Palmaz GENESIS stent; Cordis Europa NV, Johnson & Johnson Corp, Roden, The Netherlands). In addition, a carotid to subclavian artery Dacron bypass was implanted.

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Her medical history included arterial hypertension, chronic renal insufficiency (stage 3), secondary hyperparathyroidism, chronic obstructive pulmonary disease (Global Initiative for Chronic Obstructive Lung Disease stage 2), peripheral artery disease, and restless legs syndrome. Her left ventricular systolic cardiac function showed decreased systolic function (ejection fraction, 35%).

In December 2016, she was readmitted to the primary hospital, presenting with painful swelling in the left supraclavicular region. After clinical examination, an infection of the carotid to subclavian artery bypass was confirmed on computed tomography (CT) angiography. A reoperation was performed, consisting of bypass explantation, patching of the left carotid artery, and left subclavian artery ligation.

After 1 month, she was referred to our hospital because of recurrent hemoptysis. A CT scan demonstrated sudden growth in the aneurysm sac (from 55.1 mm to 68.3 mm), with a contained intrabronchial rupture of the thoracic aorta in the mediastinum and a type Ib endoleak, caused by degeneration in the distal sealing zone. Endovascular repair was successfully performed, with distal extension of the thoracic stent using a thoracic stent graft (42-32-165; Cook Medical). The patient was discharged on postoperative day 5.

After a symptom-free period of 2 months, the patient was again referred to our institution for repeated hemoptysis. CT angiography showed air bubbles in the aneurysm sac between the stent graft and the esophagus (Fig 1). Despite leukocytosis (leukocyte count, $17.5 \times 10^9/L$; normal range, $4.419-12.68 \times 10^9/L$), elevated C-reactive protein concentration (11.6 mg/dL; normal range, <0.5 mg/dL), and increased procalcitonin level (0.9 ng/mL; normal range, <0.5 ng/mL), no clinical signs of systemic sepsis were present. Wide-spectrum antibiotic therapy was started (meropenem, 1 g intravenously [IV] every 8 hours; daptomycin, 6 mg/kg IV every 24 hours; and micafungin, 100 mg/day IV), then modified according to the antibiogram. Blood cultures showed *Haemophilus influenzae*, *Candida tropicalis* and *Candida glabrata*, and *Enterococcus faecium*. Esophagogastroscopy and bronchoscopy were performed. The bronchial system



Fig 1. Computed tomography (CT) scan showing air collection between esophagus and stent graft.

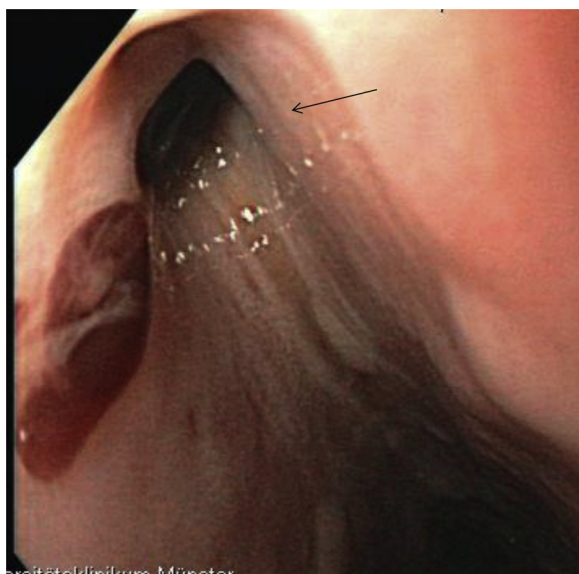


Fig 2. Esophagoscopy image showing the aorto-esophageal fistula (AEF; arrow).

was intact; however, a penetrating esophageal ulcer was found, and an AEF with concomitant stent graft infection was confirmed (Fig 2).

As an alternative palliative treatment, a multidisciplinary surgical multistep approach was proposed, with the cooperation of vascular, cardiothoracic, and general surgeons. The patient agreed to the extensive treatment plan, with informed consent. Because of her high mortality risk (European System for Cardiac Operative Risk Evaluation [EuroSCORE] II, 23.44%), a three-step treatment approach was planned as follows.

The first step consisted of deployment of an intraluminal vacuum system in the esophagus to enhance the esophageal healing process, using an Endo-SPONGE system (B. Braun Melsungen Ltd, Melsungen, Germany). The system consists of a sheath that is placed inside the esophageal ulceration or

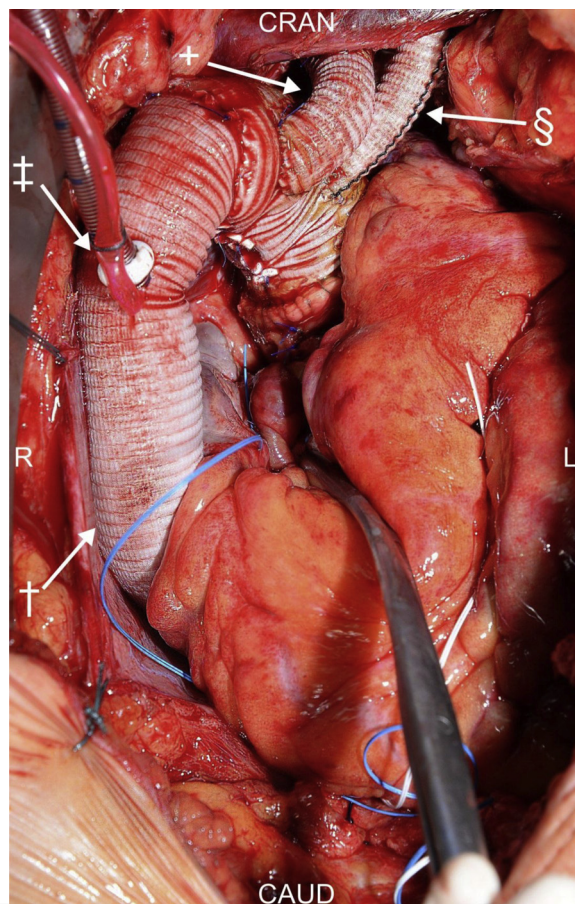


Fig 3. Intraoperative image: §, left carotid artery; +, truncus brachiocephalicus; ‡, aortic arch; †, descending aorto-aortic bypass.

esophageal lumen under endoscopic control. After removal of the endoscope, the sponge was delivered in the cavity using a pusher device over the overtube. The position of the sponge was checked endoscopically after removal of the overtube, and suction was applied using a negative pressure pump (Info-V.A.C., Kinetic Concepts Inc, San Antonio, Tex) after the tube was repositioned through the nose. Sponge changes were performed every 3 days. Meanwhile, the patient received parenteral nutrition. After a week, the vacuum system was withdrawn, and the esophageal leakage was covered with a Polyflex self-expanding covered stent (Willy Ruesch GmbH, Kernen, Germany) as this has been reported to be helpful in controlling sepsis and allowing early oral intake.³

The second step consisted of an aortic arch replacement through a median sternotomy, with a gentamicin-impregnated 28-mm Dacron conduit (Vascutek, Sulzer Medica, Austin, Tex), and reimplantation of the brachiocephalic trunk and left carotid artery with an interposition graft to the aortic graft. The chronically occluded (but asymptomatic) left subclavian artery was not revascularized. In addition, a retrocardiac tunneled aorto-aortic bypass, with a 20-mm Dacron conduit (Vascutek) between the ascending and descending aorta, was performed under deep hypothermia and circulatory arrest.

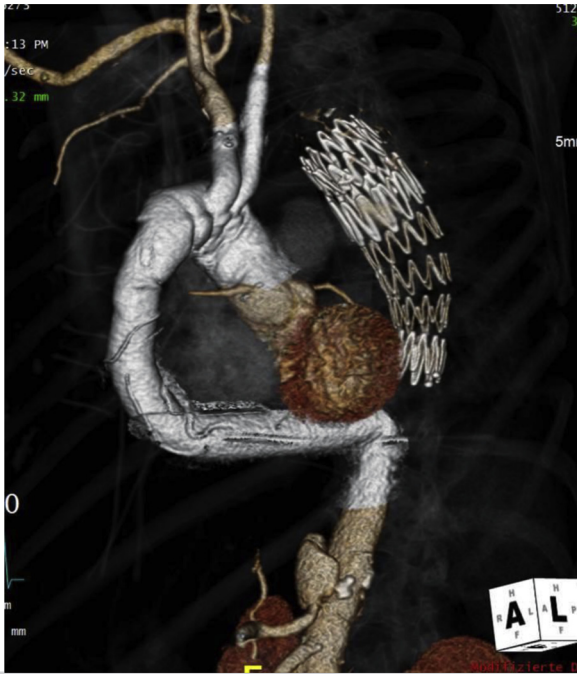


Fig 4. Postoperative (stage 2) computed tomography (CT) scan showing the reconstruction of the aortic arch and descending aortic bypass. The remaining thoracic stent graft is seen on the left.

with an additional explantation of the aortic arch stent graft and carotid stent (Figs 3 and 4). The proximal descending and distal descending aortic stumps were closed with direct suture. After 1 month of intensive care therapy, she had partially recovered, and the third step was carried out. A left thoracotomy and explantation of the remaining thoracic stent graft, extended débridement and repair of the AEF with direct suture of the wall, and wrapping with the remaining uninfected aortic wall were performed. Because of respiratory insufficiency and a prolonged weaning procedure, the patient underwent tracheotomy. Nevertheless, the patient recovered quickly, and after 3 days in the intensive care unit, the patient was transferred to the intermediate care unit. The tracheostomy was closed at 15 days, and the second operation took place 30 days after the first. Twenty days after the last surgical procedure, the patient was discharged. The total time between the first operation and the discharge after the last operation was 68 days.

After 3 months of follow-up, another gastroscopy was performed, and the esophageal stent was removed. The wall was completely healed, and she is still doing well. The broad-spectrum antibiotic therapy was continued for 6 weeks after discharge.

DISCUSSION

Infections after endovascular procedures represent challenging complications for the treating physician and are related to high mortality rates. AEF is a rare but well-described complication after thoracic endovascular aortic repair.² Promising results have been shown with explantation of the infected graft and surgical repair of

the fistula or esophageal resection with reanastomosis; however, for patients unfit for surgery, alternative palliative procedures with antibiotic therapy and parenteral nutrition have been proposed.^{4,5}

In this case, because of a high mortality risk, we preferred a three-step treatment approach, with close collaboration between vascular, cardiothoracic, and general surgery specialists. This contributed to reduced surgical and anesthesia risks related to longer operative times and allowed a partial recovery between each procedure. Of note, in cases of aortobronchial fistula, endovascular treatment has shown encouraging results; however, in cases of AEF, it is a temporary solution.⁶ Our experience confirms these statements. Unfortunately, we did not initially recognize the AEF, and a simple endovascular covering of the aortic rupture site was performed. Most reports have proposed an all-in-one treatment, encompassing explantation, in situ reconstruction, and repair of the AEF in the same session, albeit with long procedural times, high mortality rates (78%), and risk of reinfection.^{2,7}

In our case, the proximal part of the endovascular graft reached the origin of the brachiocephalic trunk, and a proximal clamping for a single-stage procedure would have been highly risky. We preferred a multistep approach as it allowed extra-anatomic aortic reconstruction, thus potentially reducing the risk for reinfection of the prosthesis and improving the esophageal healing process, which can be a challenge in such cases. Some authors have suggested an extensive resection of esophagus and an esophageal-gastric anastomosis with a gastric pullup or with a colon interposition; others have suggested the interposition of a muscle or peritoneal flap, given its effect in cases of surgical site infections.⁸ In our case, reconstruction was performed, and an esophageal stent was placed to protect the esophageal wall and to allow normal enteral nutrition.³ Reconstruction with direct suture of the esophageal wall was chosen, given the intraoperative finding of a barely visible and localized infection.

AEF remains a highly lethal clinical entity, and a clear, standardized therapeutic approach is still lacking. We believe that our three-step approach should be considered in patients deemed fit for surgery. Collaboration among experts in different disciplines is pivotal in establishing tailored treatments. A staged treatment aims to shorten operative times, reducing the risk of anesthesia complications and providing patients the time to recover after each procedure.

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