

**Case  
Report**

# Aortoesophageal Fistula after Endovascular Repair for Aberrant Right Subclavian Artery Aneurysm

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**A 76-year-old woman with malignant rheumatic arthritis developed dysphagia and hoarseness secondary to an aberrant right subclavian artery aneurysm. We performed a hybrid endovascular repair with concomitant surgical treatment for the aberrant right subclavian artery aneurysm. One month after discharge, she was emergently admitted to our hospital because of chest pain and fever. We diagnosed aortoesophageal fistula and stent graft infection based on computed tomography, gallium scintigraphy, and esophagoscopy results. Esophagectomy, elimination of the infected stent graft, and muscle plom-bage were performed during several surgeries. However, she died of hemorrhagic shock secondary to an aortobronchial fistula.**

**Keywords:** aberrant right subclavian artery aneurysm, aortoesophageal fistula, endovascular procedures

## Introduction

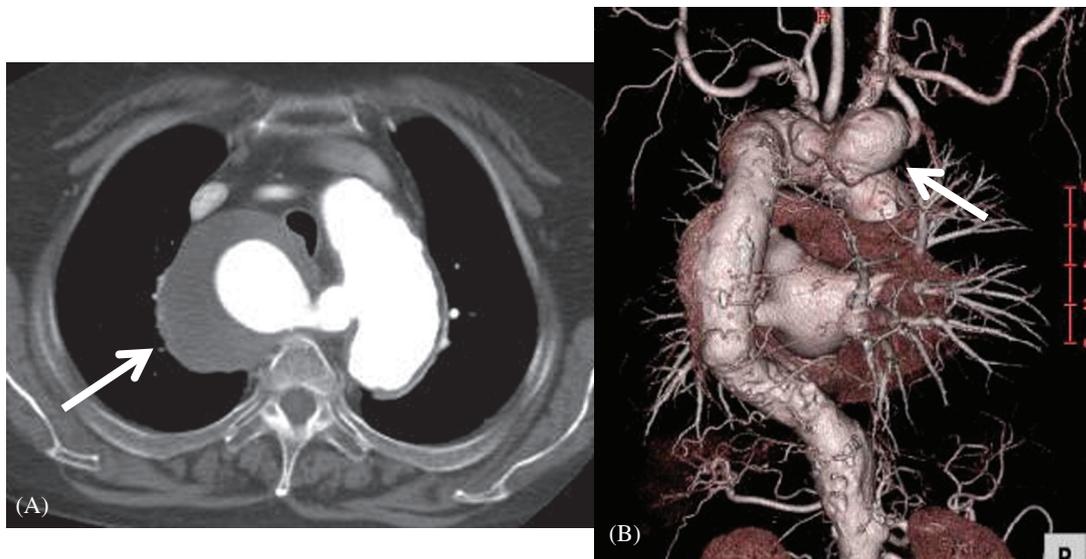
Aortoesophageal fistula (AEF) after stent grafting, a relatively rare condition, has become a problem because of patients' lethal clinical conditions and increasing numbers of stent grafting procedures.<sup>1-4)</sup> Recently, endovascular stent grafting combined with surgery has been performed for aberrant right subclavian artery (arteria lusoria) aneurysm (ASCAA).<sup>5,6)</sup> We herein describe a case of an AEF after hybrid endovascular repair with concomitant surgery for ASCAA.

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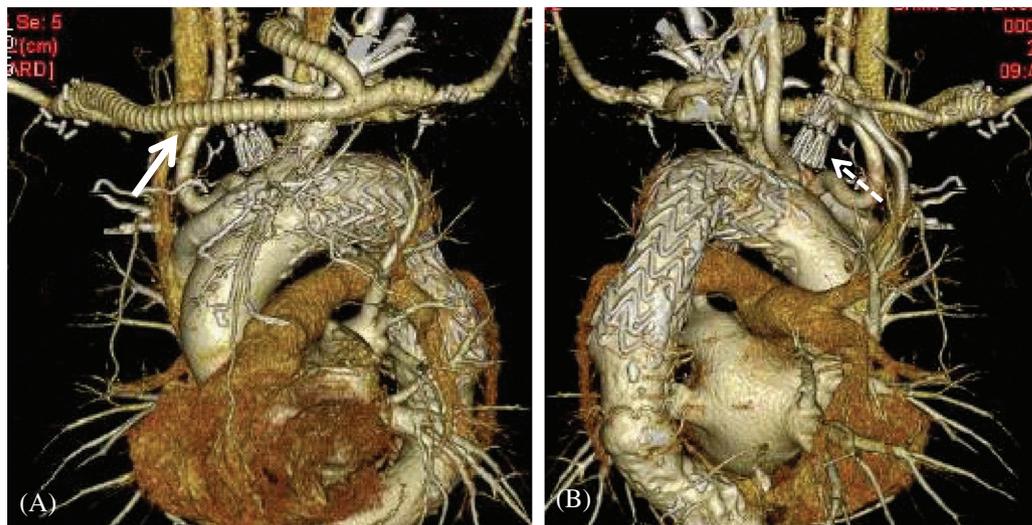
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## Case Report

A 74-year-old centrally obese woman receiving corticosteroids for malignant rheumatic arthritis developed dysphagia and hoarseness. Chest X-ray revealed prominence of the right first curve of the cardiac shadow. A computed tomography (CT) angiogram showed an ASCAA 6.5 cm in diameter that compressed the esophagus and trachea (**Fig. 1**). Coagulopathy and inflammatory responses were not detected. We performed hybrid endovascular repair comprising left common carotid artery to bilateral subclavian artery bypass with a T-shaped polytetrafluoroethylene graft and subsequent insertion of a thoracic aortic endovascular stent graft (TEVAR; 37 mm–15 cm GORE TAG Thoracic Endoprosthesis, W. L. Gore & Associates Inc., Flagstaff, Arizona, USA) and a stent graft (16-mm Zenith Flex AAA Endovascular Graft Iliac Plug, Cook Medical Inc., Bloomington, Indiana, USA) between the ASCAA and origin of the right vertebral artery (**Fig. 2**). On postoperative day 7, she developed right homonymous hemianopia caused by a left occipital lobe infarction. She was



**Fig. 1** Computed tomography angiogram before hybrid endovascular repair shows aberrant right subclavian artery aneurysm (arrow). (A) Axial and (B) three-dimensional views.

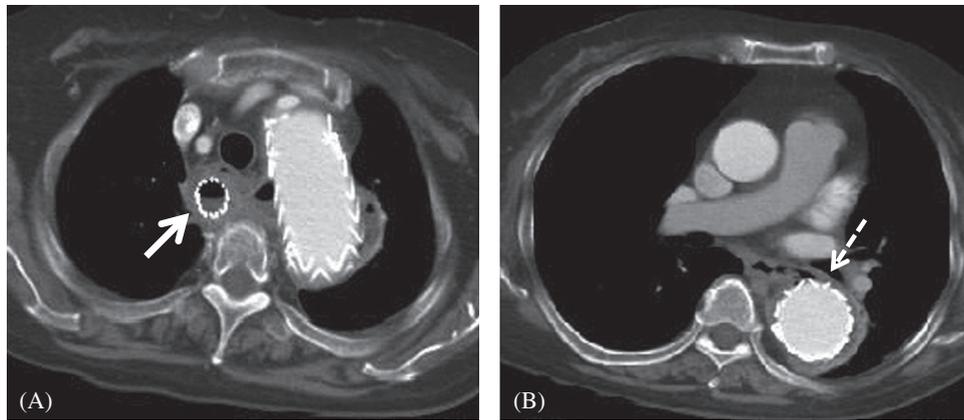


**Fig. 2** Hybrid surgical repair for aberrant right subclavian artery aneurysm. First, left common carotid artery to bilateral subclavian artery bypass was performed (A, arrow). Next, thoracic endovascular aortic repair and insertion of an occlusion stent graft (B, broken arrow) were performed.

discharged on postoperative day 42. Postoperative CT angiography showed a complete exclusion of the ASCAA without endoleakage.

One month after discharge, she was emergently admitted to our hospital because of chest pain with fever. Chest X-ray showed a disappearance of the ASCAA shadow. Blood tests showed elevated white blood cells (11,100/ $\mu$ L) and C-reactive protein (24.68 mg/dL) with coagulopathy. CT angiography, gallium scintigraphy, and esophagoscopy indicated an AEF, stent graft infection, and mediastinitis (**Fig. 3**). Esophagoscopy revealed

a mid-esophageal fistula with mucosal erosion and no evident bleeding. Empirical antimicrobials and proton pump inhibitors were administered. However, 2 weeks after admission, she developed sudden hematemesis, presumably from the AEF, and underwent insertion of a Sengstaken-Blakemore tube into the esophagus. Emergent angiography delineated leakage of contrast agent from the descending aorta at the distal aspect of the stent graft. Continuous TEVAR (31-mm and 37 mm–15 cm GORE TAG Thoracic Endoprosthesis) was performed. Twenty-four days after admission, we performed



**Fig. 3** Computed tomography angiogram after emergent admission reveals air in the stent (**A, arrow**), air and fluid around the thoracic stent graft and mediastinum with contraction of aberrant subclavian artery aneurysm, esophagus fistula (**B, broken arrow**), and consecutive dilatation of descending aorta.

esophagectomy, elimination of the stent graft in the right subclavian artery, and latissimus dorsi muscle plom-bage to the mediastinum through a right thoracotomy followed by cervical esophagostomy and enterostomy. The esophagus had two fistulas: one associated with the stent graft in the right subclavian artery (**Fig. 4, arrow**) and one associated with the stent graft in the descending aorta (**Fig. 4, asterisk**). Histopathological findings showed necrosis, disappearance, and thinning of the muscle layers of the esophagus; granulation accompanied by inflammatory cell infiltration and growth of capillaries around the fistula in right subclavian artery; and inflammatory cell infiltration and necrosis of all layers around the fistula with fibrosis in the descending artery. *Pseudomonas aeruginosa* was cultured from abscesses around the mediastinum and the stent.

After the operations, she underwent a tracheotomy because of bilateral recurrent nerve paralysis, and long-term administration of antimicrobial drugs was performed because of sustained elevation of inflammatory markers. However, 5 months after the emergency admission, stent graft infection in the thoracic aorta was still present, and an aortobronchial fistula to the left B6 developed with hemoptysis. Therefore, she underwent partial removal of the infected thoracic aortic stent graft, replacement of the descending aorta, and placement of a drainage tube in the mediastinum through a left thoracotomy. Histopathological findings of the explanted descending aorta revealed aortic infection as evidenced by abscess formation with inflammatory cell infiltration mainly composed of neutrophils, deposition of fibrin, and edema in the intima, which revealed atherosclerosis and



**Fig. 4** Two fistulas at the mid and distal esophagus.

cholesterol cleavage. Although the C-reactive protein level gradually decreased to 1.31 mg/dL, subsequent CT angiography revealed a pseudoaneurysm around the residual proximal thoracic aortic stent graft that could not be removed because of severe adhesion. Finally, an open stent-grafting technique (home-brew stent graft comprising a woven graft combined with a bronchial Z stent) was performed through a median sternotomy 7 months after the emergency admission. However, she died of hemorrhagic shock secondary to a new aorto-bronchial fistula 11 months after the first operation.

## Discussion

We first reported a case of an AEF after hybrid endovascular repair for ASCAA. The incidence of an AEF after TEVAR is relatively rare (approximately 1.9%).<sup>1,2)</sup> However, increasing numbers of endovascular stent grafting procedures, including those performed for ASCAA, increase the problems of high mortality and difficulty in AEF treatment.

The causes of an AEF after stent grafting are considered to be as follows: (1) direct erosion of the stent graft into the esophagus, (2) pressure necrosis of the self-expanding endoprosthesis, (3) ischemic esophageal necrosis due to disruption of arteries that feed the esophagus, (4) stent graft infection, (5) pseudoaneurysm development, and (6) endoleakage into the residual aneurysmal sac.<sup>1,2)</sup> In our patient, endoleakage and pseudoaneurysm formation were not detected. Although stent graft infection continued, we suggest that graft infection was not the cause of an AEF and that the infection may have developed after AEF formation in our patient. Histopathological findings of the esophagus showed esophageal necrosis and inflammatory changes with no apparent infectious changes. Thus, direct erosion of the stent into the esophagus or pressure necrosis of the self-expanding endoprosthesis may have caused the AEF in this patient. Findings of the resected esophagus support this interpretation. Furthermore, we suggest that the arterial wall may have been damaged by the stent graft because of vulnerability of the arterial wall due to long-term corticosteroid therapy.

Recent reports have shown that an AEF from a thoracic aortic aneurysm may result from pressure necrosis by aneurysmal expansion and inflammatory destruction by aneurysmal infection or hematoma reabsorption from the ruptured aneurysm.<sup>3,7)</sup> Our patient had dysphagia due to esophageal compression by the large ASCAA. An AEF may have been present before the operation in this patient. Histopathological findings of the AEF in the right subclavian artery also suggested a preoperative AEF. It is possible that the aneurysm itself caused pressure necrosis and chronic inflammatory changes in the esophagus. If an AEF is suspected based on symptoms and CT findings before surgery, preoperative esophagoscopy should be performed, and another strategy involving management of the aneurysm with esophagectomy should be considered.

Treatment for an AEF involves radical surgery comprising exclusion of the infected stent graft, aortic graft replacement, and esophageal fistula repair or esophageal resection.<sup>1,2,7)</sup> TEVAR has recently been performed in patients with an AEF, but it is controversial because of insertion into a potentially contaminated or infected

field.<sup>4)</sup> Based on our experience in the present case, TEVAR may be a palliative treatment to prevent hemorrhagic shock because the descending thoracic stent graft infection continued despite esophagectomy, muscle plomage, and partial removal of the stent graft. Therefore, radical removal of infected prostheses may be needed to treat an AEF.

## Conclusion

We experienced a case of an AEF after hybrid endovascular repair for ASCAA. Preoperative esophagoscopy may be necessary when an aneurysm compresses the esophagus.

## Disclosure Statement

The authors have no conflicts of interest to declare.

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