

Arterio-oesophageal fistula caused by aberrant right subclavian artery aneurysm

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Abstract

A 63-year old man had dysphagia for 4 months and was admitted to our hospital with sudden haematemesis. Computed tomography revealed an aberrant right subclavian artery (ARSA) aneurysm and free air inside the aneurysm. Arterio-oesophageal fistula was diagnosed, and an emergency operation was performed. Before thoracotomy, a percutaneous transluminal angioplasty (PTA) balloon was inserted from the right brachial artery and placed at the orifice of the ARSA to control bleeding. Through a left thoracotomy, the aorta was excised and the orifice of the ARSA was exposed. There was no sign of infection in the operative field. Bleeding from the ARSA was controlled by balloon occlusion. The proximal portion of the right vertebral artery was ligated and blood backflow from the ARSA stopped. The ARSA was ligated proximal to the aneurysm, and the orifice of the ARSA to the aorta was closed. The infected aneurysm was not resected. The descending aorta was replaced by a rifampin-bonded artificial graft, and omentopexy was performed. Gastrostomy was performed for decompression of the oesophagus and enteric feeding. Three months after the operation, gastrointestinal endoscopy showed a healed oesophageal ulcer, and the patient was discharged uneventfully.

Keywords: Arterio-oesophageal fistula • Aberrant right subclavian artery aneurysm • Percutaneous transluminal angioplasty balloon • Gastrostomy

INTRODUCTION

Arterio-oesophageal fistula is a life-threatening disease that has been treated mainly by aneurysmectomy and oesophagectomy in simultaneous or staged procedures. Since these procedures often result in unfavourable outcomes, it is important to evaluate other treatment strategies. We describe the successful repair of an aberrant right subclavian artery (ARSA) aneurysm accompanied by arterio-oesophageal fistula by prosthetic replacement of the aorta without aneurysmectomy and gastrostomy for decompression and drainage of the oesophagus.

CLINICAL SUMMARY

A 63-year old man had dysphagia for 4 months and was admitted to the hospital with sudden haematemesis. He had slight fever (37.5°) and laboratory data showed leukocytosis (white blood cell count: 17 000/mm³) and an elevated C-reactive protein value (21.6 mg/dl). Computed tomography (CT) showed an ARSA aneurysm behind the oesophagus and free air inside the aneurysm (Fig. 1A and B). In the delayed phase of CT, the ARSA aneurysm wall had a contrast effect extending to the aorta. The left vertebral artery originated between the left subclavian artery and the ARSA (Fig. 2). From these findings, we suspected active infection extending from the aneurysm to the aorta and planned open surgery rather than endovascular repair.

Compression of the oesophagus by the aneurysm was thought to cause the arterio-oesophageal fistula, resulting in haematemesis. Before emergent thoracotomy, a percutaneous transluminal angioplasty (PTA) balloon, (9-mm diameter; Wanda®, Boston Scientific, Watertown, MA, USA) was inserted through a guiding catheter in the right brachial artery and advanced into the thoracic aorta under echocardiographic and radiographic guidance. The origin of the ARSA was temporally occluded by the PTA balloon to control bleeding. Femoro-femoral extracorporeal circulation was established for distal perfusion, and a left anterolateral thoracotomy was performed in the third intercostal space. The aorta was clamped proximal and distal to the ARSA, and the clamped segment was opened. The right subclavian artery, vertebral artery and mammary artery were exposed via a right supra-subclavian approach. After the PTA balloon was removed, the right subclavian artery was ligated proximal to the origin of the right vertebral artery. Absence of blood flow through the origin of ARSA convinced us that there was no collateral communication between the right vertebral artery and the aneurysm. There was no infectious thrombus in the aneurysm or the oesophagus. The ARSA was closed at the aortic orifice. Aortic replacement was performed with a rifampin-bonded, gelatin-sealed graft (Gelweave™, Vascutek Ltd., Scotland). The artificial graft was covered with omentum. After closing the thoracic cavity, gastrostomy was performed to reduce intra-gastric and intra-oesophageal pressures, and for early induction of enteric nutrition. Adequate blood supply to the right upper extremity was confirmed by monitoring

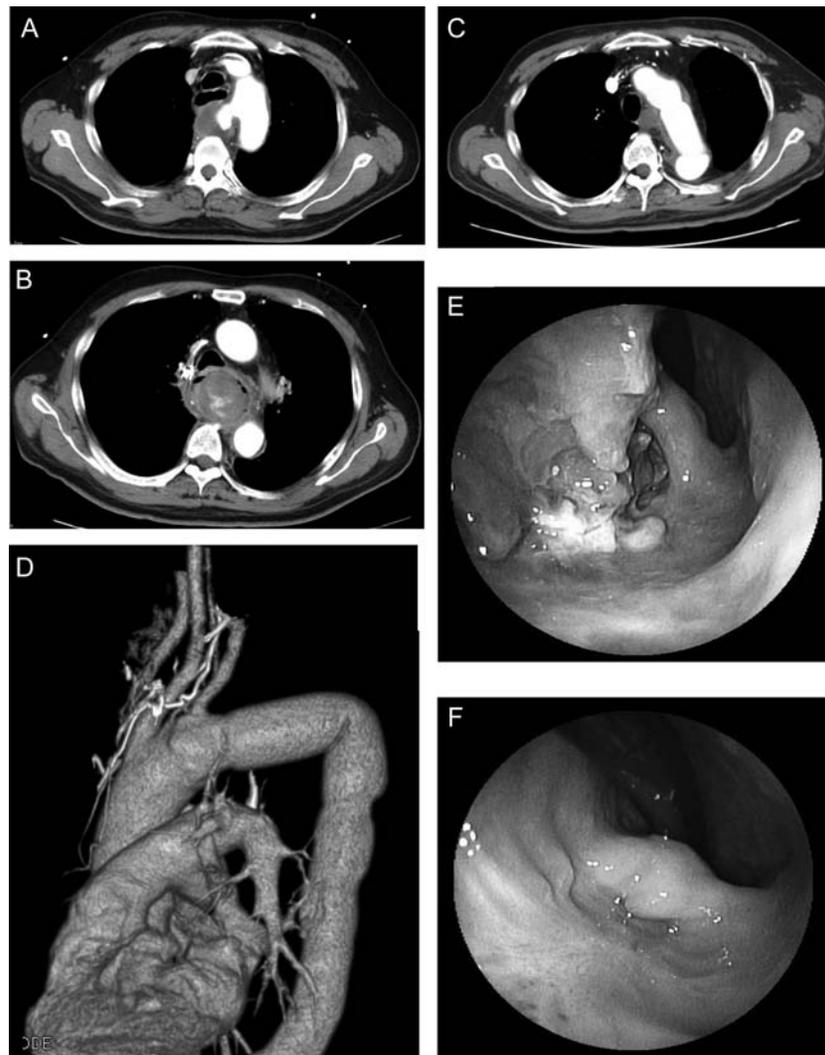


Figure 1: Preoperative CT shows an ARSA aneurysm behind the oesophagus (A) and with free air inside the aneurysm (B). Postoperative CT and a three-dimensional reconstruction image showed shrinkage of the ARSA aneurysm. (C and D) On postoperative day 4, an upper gastrointestinal endoscopy showed that the oesophageal ulcer was covered with fibrinous tissue (E). Three months after surgery, endoscopy showed complete healing of the oesophageal ulcer (F).

the arterial blood oxygen saturation. Carbapenem (6 g/day) was administered for 14 days. Postoperative laboratory data improved immediately, and he had no fever or inflammatory signs. On the fourth postoperative day, upper gastrointestinal endoscopy confirmed that the oesophageal ulcer was covered with fibrinous tissue, and transgastric feeding was started (Fig. 1D). Gastric drainage was continued for 1 month. Antibiotic treatment was continued for 3 weeks until all laboratory data were normal. Oral feeding was started at 3 months, when gastro-oesophageal endoscopy showed complete healing of the oesophageal ulcer (Fig. 1E). A postoperative CT showed shrinkage of the ARSA aneurysm (Fig. 1C). The patient was discharged 4 months after surgery.

DISCUSSION

Arterio-oesophageal fistula is a serious and fatal complication of thoracic aortic aneurysm. In our case of arterio-oesophageal fistula accompanied by an ARSA, the aorta was replaced without aneurysmectomy, and the oesophagus was preserved using gastric tube drainage.

Aberrant subclavian artery is a rare anomaly and occurs in about 0.5–2.0% of the population [1]. Aneurysm of the origin of the aberrant subclavian artery is called ‘Kommerell diverticulum’ and arises from a remnant of the primary aorta [2]. Since a Kommerell diverticulum compresses the surrounding structures, the most common symptom is dysphagia, occurring in 33–38% of all cases. In addition, dyspnoea, hoarseness, chest pain or shoulder pain may also occur. However, 21–50% of patients are asymptomatic.

There are several reports of aneurysm rupture in patients with arterio-oesophageal fistula. From their review of 32 patients, 19% presented with fatal aneurysm rupture [1]. When an ARSA is accompanied by arterio-oesophageal fistula, most of the patients have haematemesis or tarry stools, but they often die before or during the operation because of uncontrollable bleeding [3]. Miller *et al.* [4] reported that ARSA oesophageal fistula induced by a nasogastric tube could be repaired using intra-oesophageal balloon tamponade to control the bleeding.

The treatment of arterio-oesophageal fistula accompanied by ARSA aneurysm has been controversial. The standard strategy has been excision and reconstruction of the aorta and

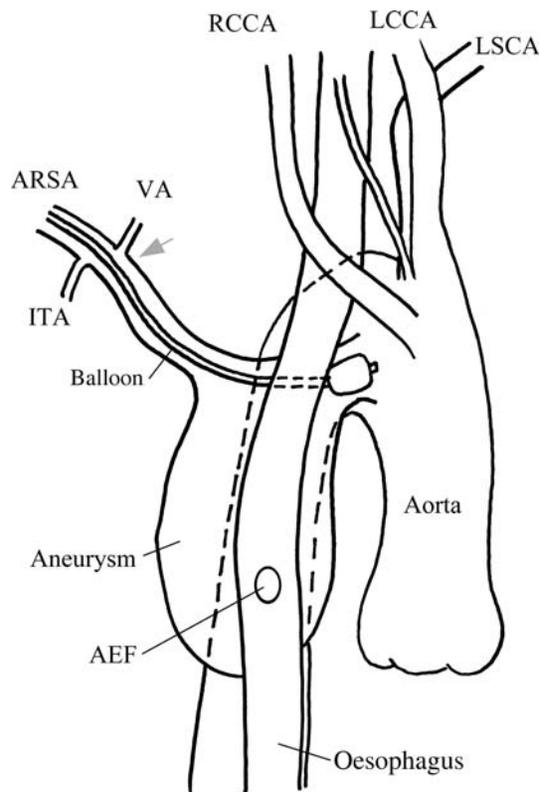


Figure 2: Schema of the operation. A PTA balloon was placed at the ARSA origin. The ARSA was ligated at the proximal side of the vertebral artery origin (grey arrow). Absence of blood flow through the origin of ARSA convinced us that there was no collateral communication between the right vertebral artery and the aneurysm. AEF: arterio-oesophageal fistula; LSCA: left subclavian artery; RCCA: right common carotid artery; LCCA: left common carotid artery; VA: vertebral artery; ITA: internal thoracic artery.

oesophagus in simultaneous or staged operations. Omentopexy is often performed, and antibiotics are given. In Japan, a rifampicin-bonded artificial graft is usually used in infected aneurysm because of the lack of homografts.

Recently, endovascular stent grafting for arterio-oesophageal fistula, which immediately stopped the bleeding from the aorta and achieved a successful outcome, has been reported. However, several cases developed intractable infection that required secondary prosthetic aortic replacement and oesophagectomy [5]. In our case, one of the reasons for the successful result was the fact that the compartment of the arterio-oesophageal fistula was isolated from the thoracic cavity and the aorta by closing the aneurysmal wall, and the contents of the aneurysm discharged via gastrostomy. This strategy should be considered in future cases of ARSA with arterio-oesophageal fistula.

Conflict of interest: none declared.

REFERENCES

- [1] Kouchoukos NT, Masetti P. Aberrant subclavian artery and Kommerell aneurysm: surgical treatment with a standard approach. *J Thorac Cardiovasc Surg* 2007;133:888-92.
- [2] Kommerell B. Verlagerung des Oesophagus durch eine abnorm verlaufende Arteria subclavia dextra (Arteria lusoria). *Fortschritte auf dem Gebiete der Rontgenstrahlen* 1936;54:590-5.
- [3] Edwards BS, Edwards WD, Connolly DC, Edwards JE. Arterial-oesophageal fistulae developing in patients with anomalies of the aortic arch system. *Chest* 1984;86:732-5.
- [4] Miller RG, Robie DK, Davis SL, Cooley DA, Klish WJ, Skolkin MD, Kearney DL, Jaksic T. Survival after aberrant right subclavian artery-oesophageal fistula: case report and literature review. *J Vasc Surg* 1996;24:271-5.
- [5] Jonker FH, Heijmen R, Trimarchi S, Verhagen HJ, Moll FL, Muhs BE. Acute management of aortobronchial and aorto-oesophageal fistulas using thoracic endovascular aortic repair. *J Vasc Surg* 2009;50:999-1004.