

Aortoesophageal Fistula after Thoracic Endovascular Aortic Repair Diagnosed and Followed with Endoscopy

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Abstract

A 70-year-old man reported dysphagia two months after undergoing thoracic endovascular aortic repair (TEVAR). An endoscopic examination revealed a fistula between the esophagus and the thoracic aortic aneurysm, and computed tomography (CT) showed that the thoracic aortic aneurysm had increased in size. The patient was diagnosed with an aortoesophageal fistula (AEF), and surgical replacement of the thoracic aorta was performed. AEFs are a rare but typically fatal complication after TEVAR. Physicians should consider a diagnosis of AEF and perform endoscopic examinations and CT in patients who undergo TEVAR and subsequently complain of dysphagia.

Key words: aortoesophageal fistula (AEF), thoracic endovascular aortic repair (TEVAR), endoscopy

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Introduction

Aortoesophageal fistulas (AEF) are a rare disorder that can occur as an unusual complication after thoracic endovascular aortic repair (TEVAR). Worldwide, TEVAR has become a standard treatment for aortic aneurysms and dissections because it is associated with lower rates of postoperative morbidity than conventional open surgery. There is no established treatment for AEFs, and the mortality rate associated with the disorder is high. Performing endoscopic examinations is helpful in making a prompt and definite diagnosis of AEF. However, there have been few case reports in which endoscopic follow-up was performed after surgery for AEF. We herein present a case of AEF that developed after TEVAR in which the patient was diagnosed on an endoscopic examination and computed tomography (CT) and treated with thoracotomy.

Case Report

A 70-year-old man who had undergone TEVAR in December 2009 reported dysphagia two months after the pro-

cedure. CT showed that the aneurysm of the thoracic aorta had increased after TEVAR, thus forming a false lumen and compressing the esophagus (Fig. 1). The patient was admitted to the department of cardiovascular surgery at Hirosaki University Hospital. The man was a heavy smoker (40 cigarettes a day for 50 years). A blood test revealed an elevated level of C-reactive protein (6.2 mg/dL) (Table). An endoscopic examination performed after admission revealed a protrusion (similar to a submucosal tumor) with an associated defect of the esophageal wall that exposed the coagula and necrotic tissue at the upper section of the esophagus (20 cm from the incisor) (Fig. 2). Even a thin scope (Olympus GIF-XP260) could not pass through the portion due to stenosis. Subsequent angiography revealed an extravasation from the proximal side of the stent placed at the descending thoracic aorta (Fig. 3). The patient was diagnosed as having an AEF, and removal of the stent-graft, replacement of the descending aorta with a prosthetic graft and omental implantation were performed. A proton-pump inhibitor (PPI) was administered during the therapeutic term.

Two weeks after surgery, endoscopic examinations showed a reduction in the size of the coagula and necrotic tissue (Fig. 4a), and further reductions with regenerating

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mucosa were observed four weeks after surgery (Fig. 4b). The patient showed no complications after taking liquid food and was discharged on the 79th day postsurgery. Five

months after surgery, CT confirmed the reduction in size of the thoracic aortic aneurysm (Fig. 5a), and an endoscopic examination showed that the brownish irremovable necrotic mass was still present in the esophagus (Fig. 4c). CT performed 10 months after surgery revealed the almost complete disappearance of the thoracic aneurysm (Fig. 5b). The patient remains well without any significant complications, and attends our hospital for regular examinations.

Table. Laboratory Data on Admission

Peripheral blood		Blood chemistry	
WBC	7,560 / μ L	TP	8.2 g/dL
Neutro	66.3 %	Alb	4.3 g/dL
Baso	0.7 %	T-bil	1.3 mg/dL
Eosino	2.1 %	AST	18 IU/L
Lympho	23.9 %	ALT	12 IU/L
Mono	7.0 %	LDH	159 IU/L
RBC	486×10^4 / μ L	ALP	222 IU/L
Hb	14.9 g/dL	γ -GTP	159 IU/L
Ht	44.4 %	AMY	62 IU/L
Plt	28.6×10^4 / μ L	BUN	20 mg/dL
		Cr	1.0 mg/dL
		CRP	6.2 mg/dL

WBC: white blood cell, RBC: red blood cell, Hb: hemoglobin, Ht: hematocrit, Plt: platelets, Alb: albumin, T-bil: total bilirubin, AST: aspartate aminotransferase, ALT: alanine aminotransferase, LDH: lactate dehydrogenase, ALP: alkaline phosphatase, γ -GTP: γ -glutamyl transpeptidase, AMY: amylase, BUN: blood urea nitrogen, Cr: creatinine, CRP: C-reactive protein

Discussion

Recently, the number of patients who undergo TEVAR for thoracic aortic disease has increased due to the minimally invasive nature of this technique in comparison with open repair (1). However, TEVAR is associated with several complications, including paraplegia, stroke, post-implantation syndrome, device migration and AEF formation (2). AEF formation is the most lethal of these complications. Indeed, in an investigation of the previous literature, Muradi et al. found that only six of 24 patients (25%) who developed AEFs after undergoing TEVAR could be rescued (3). Egge-

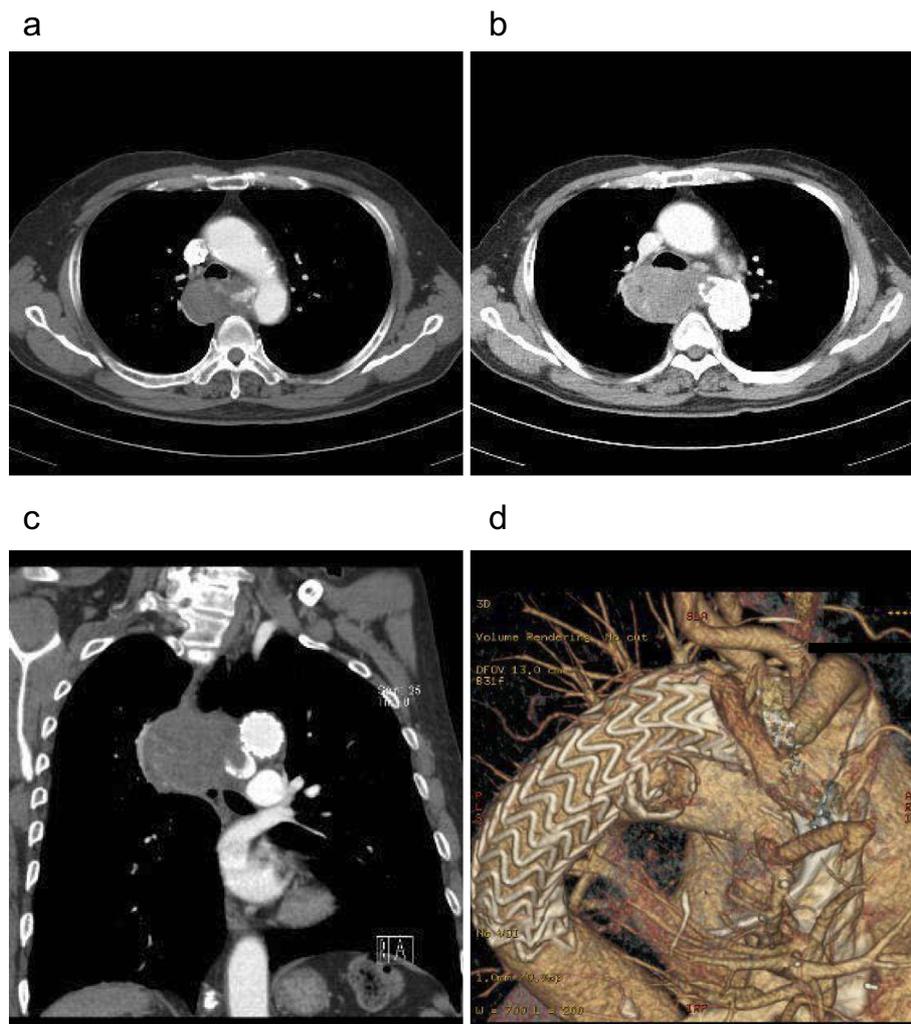


Figure 1. CT images showing the thoracic aortic aneurysm before TEVAR (a) and the increased aneurysm two months after TEVAR (b). A coronal section (c) and a 3D thoracic aortic image (d) were constructed.

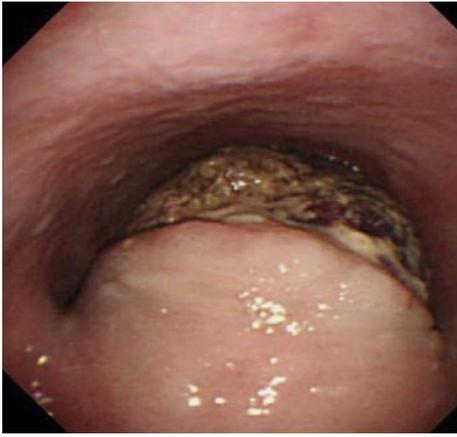


Figure 2. Endoscopic findings showed a protrusion similar to a submucosal tumor with ulceration at the posterior wall of the upper section of the esophagus.

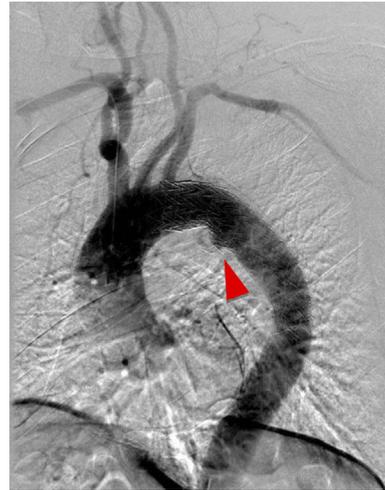


Figure 3. Angiography revealed an extravasation from the proximal side of the stent placed at the descending thoracic aorta (arrow).

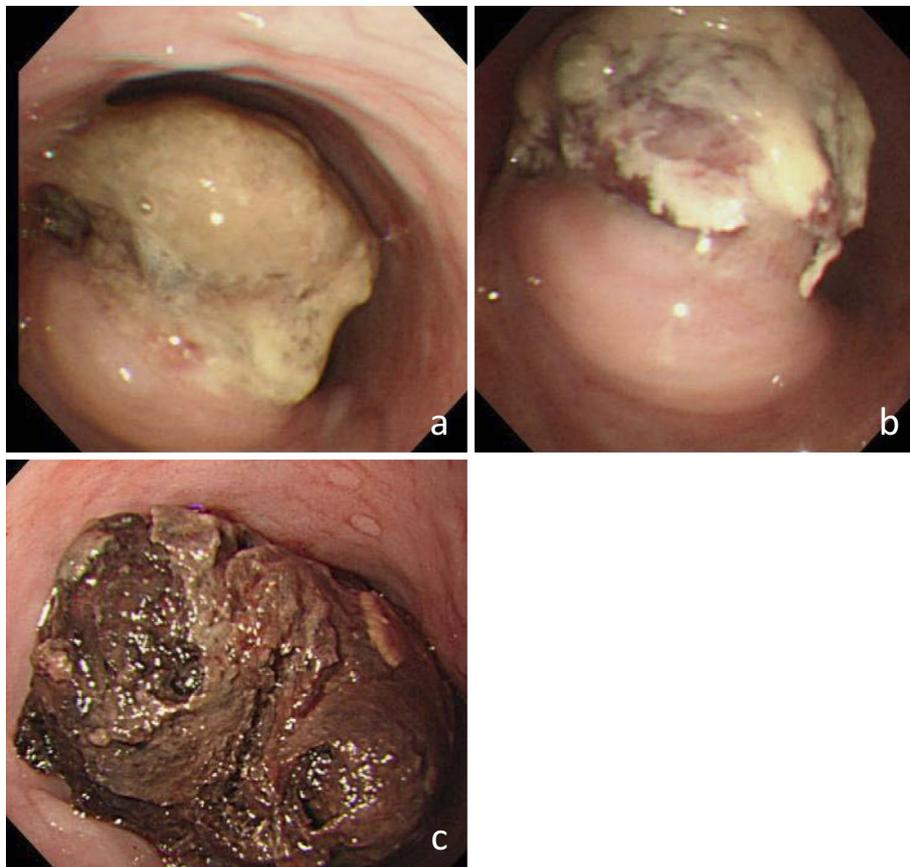


Figure 4. Endoscopic examinations were performed two weeks (a), four weeks (b) and five months (c) after surgery. The scope (GIF-XP260) was able to be passed through the stenotic part of the esophagus four weeks after the surgery.

brecht et al. also reported that six of 268 patients who had undergone TEVAR developed AEFs (incidence 1.9%) and that all of these patients died due to fatal bleeding or mediastinitis (4).

Mechanical damage caused by arterial pulsation, stent-graft rigidity and interruption of the circulation of the esophageal wall by enlarged aneurysms are all expected to

play roles in the development of AEFs (4). In our case, a false lumen of the thoracic aortic aneurysm gradually formed after TEVAR due to blood flow entry between the stent and the aneurysm (perigraft leak). As a result, the esophageal wall was compressed by the aneurysm. The clinical symptoms of AEFs generally consist of Chiari's triad: midthoracic chest pain, sentinel arterial hemorrhage

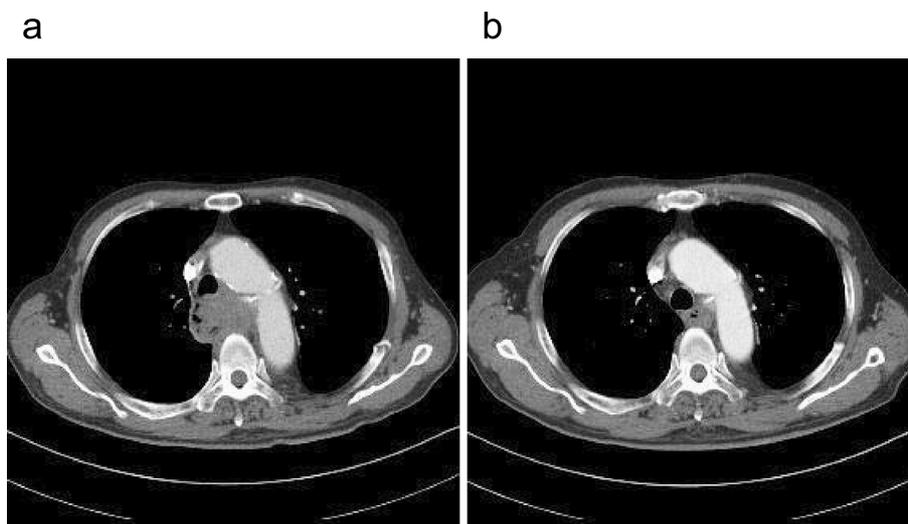


Figure 5. CT performed at five months after surgery showed that the thoracic aortic aneurysm had reduced in size (a). CT performed 10 months after surgery showed that the thoracic aneurysm had almost completely disappeared (b).

and fatal hemorrhage (5). The most common symptom of AEFs is hematemesis (6), and the presentation of only dysphagia in the absence of the triad, as seen in our patient, is rare.

There have been several reports that involved the examination of AEFs using endoscopy (4, 7-10). The most common form of AEF is a submucosal tumor-like protrusion; other forms include extrinsic compression, ulcerative lesions, bleeding only, pulsating protrusions with central fistulation and exposure of the aortic stent-graft. The endoscopic findings observed in our case consisted of a protrusion similar to a submucosal tumor, exposing the coagula and necrotic tissue. In most previous reports, the AEFs were located between the posterior to the left lateral wall of the esophagus. The location of the lesion may be useful for making a diagnosis of AEF. There is no literature reporting endoscopic findings after surgery for AEF. In the present case, omental implantation was performed to avoid esophagectomy; thus, the defect of the esophageal wall with coagula and necrotic tissue persisted for more than five months. However, the dysphagia disappeared and did not recur after surgery.

In this case, we performed small caliber endoscopy due to the presence of esophageal stenosis. However, the load of the endoscopic examinations may constitute a risk for rupture of the aortic aneurysm. Transnasal small caliber endoscopy is a safer procedure than transoral conventional endoscopy particularly in terms of cardiopulmonary effects (11, 12). Therefore, for the management of patients suspected of having AEFs, transnasal endoscopy is considered after CT.

In conclusion, the development of AEFs after TEVAR is a rare but typically fatal complication. We were able to diagnose the patient with an AEF and successfully treat the condition, although the patient presented with dysphagia only, without hematemesis. Physicians should therefore consider a

diagnosis of AEF and perform CT first followed by endoscopic examinations in any patients who has undergone TEVAR and complains of dysphagia.

The authors state that they have no Conflict of Interest (COI).

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