CASE REPORT

Secondary aortoesophageal fistula after thoracic endovascular aortic repair for a huge aneurysm

Akhmadu Muradi, Masato Yamaguchi, Atsushi Kitagawa, Yoshikatsu Nomura, Takuya Okada, Yutaka Okita, Koji Sugimoto

ABSTRACT

Thoracic endovascular aortic repair for a descending thoracic aortic aneurysm is an excellent alternative to open surgery, especially in patients with a number of comorbidities. It may cause fatal complications, including aortoesophageal fistula, but these are very rare. Here, we report the case of secondary aortoesophageal fistula four months after the procedure for a huge descending thoracic aortic aneurysm, which presented with new-onset high-grade fever accompanied by elevated inflammatory markers.

From the Department of Radiology and Center for Endovascular Therapy (A.M. 🖂 *akhmadumuradi@gmail.com*, M.Y., T.O., K.S.), Department of Surgery and Division of Cardiovascular Surgery (A.K., Y.N., Y.O.), Kobe University Graduate School of Medicine, Kobe, Japan.

Received 13 April 2012; revision requested 9 May 2012; revision received 6 June 2012; accepted 11 June 2012.

Published online 9 August 2012 DOI 10.4261/1305-3825.DIR.5912-12.1 horacic endovascular aortic repair (TEVAR) is a minimally invasive and generally excellent modality to treat descending thoracic aortic aneurysms. Several complications, however, may occur, including paraplegia, stroke, and occasionally, aortoesophageal fistula (AEF) (1, 2). AEF often has a short clinical course, and many physicians are generally unaware of this fatal complication (2). Here we report a rare case of secondary AEF that occurred four months after TEVAR, and review of the recent literature. These findings emphasize the importance of careful treatment for a huge descending thoracic aortic aneurysm.

Case report

A 65-year-old man with advanced bladder cancer and acute renal failure due to urinary tract obstruction was referred to our hospital for chemotherapy. Computed tomography (CT) revealed a huge descending thoracic aortic aneurysm (90 mm in diameter) shifting the left ventricle and esophagus (Fig. 1a). The patient first underwent percutaneous nephrostomy drainage due to hydronephrosis. TEVAR was then performed using two components of a 28×150 mm TAG thoracic endoprosthesis (WL Gore & Associates, Flagstaff, Arizona, USA) with no endoleak (Fig. 1b). The procedure and postoperative course proceeded uneventfully, and the patient was discharged 10 days later. He received chemotherapy for the advanced bladder cancer without removal of the nephrostomy tube. During the treatment, urinary tract infection occasionally occurred and was treated with oral and intravenous antibiotics.

Four months after the operation, the patient suffered from a high-grade fever of over 38 °C, which persisted for more than two weeks despite antibiotic administration. A blood test revealed an elevated leukocyte count (13 100 cells/µL) and highly elevated C-reactive protein levels (16 mg/ dL). No organism was detected in the blood culture. CT revealed a large amount of air around the stent grafts in the aneurysm sac (Fig. 2a). We initially suspected an infection of the previous stent grafts; however, endoscopy revealed a fistula located at the posterior surface of the middle section of esophagus (Fig. 2b). The patient was diagnosed as a secondary AEF. After being informed about the risks, the end stage cancer, and life expectancy; the patient and his family decided to refuse the aggressive treatment by percutaneous CT-guided drainage of the infected aneurysm sac before definitive open surgery composed of esophagectomy and replacement of the descending thoracic aortic aneurysm with removal of the infected stent grafts. Two months later, he died from hemorrhagic shock due to massive hematemesis under conservative treatment.

Discussion

Secondary AEF is a fatal complication of TEVAR, with an incidence of 1.6% to 1.9% (1, 2). TEVAR does not appear to provide any advantage



Figure 1. a, b. CT image of the huge descending thoracic aortic aneurysm before **(a)** and maximum intensity projection image after **(b)** thoracic endovascular aortic repair (TEVAR). The preoperative diameter of the aneurysm was 90 mm. The descending thoracic aortic aneurysm was treated by TEVAR without any endoleak.



Figure 2. a, b. Esophagogastroduodenoscopy (**a**) and CT (**b**) images of the aortoesophageal fistula detected air bubbles around the stent graft in the aneurysm sac of the descending thoracic aorta (**b**, *white arrow*). The precise fistula site was not visible, but no contact between the stent graft and the fistula was observed. A fistula without any bleeding was seen at the inner surface of the mid-esophagus (**a**, *black arrow*).

over conventional open surgery for this complication, and anastomotic pseudoaneurysm as an indication for TEVAR, as well as emergent TEVAR, increases the possibility of secondary AEF (1). Approximately 24 cases of secondary AEF have been previously reported in the literature (2–20), with

most (54.2%) occurring within four months after TEVAR with various types of endograft, and only six patients (25%) surviving, with a mean followup period of 11.6 months (Table).

A pathophysiologic mechanism of secondary AEF after TEVAR has been hypothesized by Eggebrecht et al. (2) as follows: 1) direct erosion of the relatively rigid stent graft through the aorta into the esophagus as the most common cause of AEF after TEVAR; 2) pressure necrosis of the esophageal wall due to the continuing forces of the selfexpanding endoprosthesis; 3) ischemic esophageal necrosis due to stent graft coverage of aortic side branches that feed the esophagus; and 4) infection of the stent graft prosthesis that eventually extends to the esophagus, eroding its wall. Pressure necrosis mechanisms also occur in the presence of endoleak that enlarge the aneurysm sac (2–4, 6, 10), or in an already large aneurysm sac prior to TEVAR (4, 5, 7, 9, 11, 19), with an average diameter of 7.5 cm (range, 6–10 cm).

The possible pathological mechanism in our case consisted of potential systemic infections from tube nephrostomy, leading to stent graft infection, although the stent graft itself showed no invasion to the esophageal wall. Furthermore, continuous compression of the huge aneurysm sac might have contributed to the erosion of the esophagus wall. Thus, we postulate that the combination of these two mechanisms might be the cause of AEF. Some reports described how these mechanisms induce an inflammatory process, adhesion, and tissue necrosis, leading to erosion and finally to fistulation, with subsequent massive hematemesis when the fistula reached the proximal or distal aspect of the endograft (1, 17).

Our patient presented with a newonset of high-grade fever with elevated markers of inflammation, thus we suspected AEF and performed CT and endoscopy for an accurate diagnosis. This symptom does not match with the general clinical presentation of AEF, which involves Chiari's triad of dysphagia or mid-thoracic pain and a short symptom-free interval, followed by a herald hemorrhage and fatal hematemesis (19). Therefore, physicians should alert and closely monitor patients who present with this symptom.

Table. Published data of secondary AEF after TEVAR

	Age (year)	Stent graft	Time after TEVAR (month)	Onset symptoms	Treatment	Outcome	AEF etiology
Hance et al., 2003 (3)	24	Z+Dacron	15	Hematemesis	Surgery	Alive at 15 months	Type 1 EL
Eggebrecht et al., 2004 (4)	62	Talent	1	High-grade fever	Conservative	Died	Erosion by stent graft
	74	Talent	9	High-grade fever	Conservative	Died	Type 1 EL
	77	Talent	2	Hematemesis	Esophageal stent	Died	Erosion by stent graft and ischemic necrosis
Czerny et al., 2005 (5)	57	Talent	1	Dysphagia	Surgery	Alive at three months	Pressure erosion
Porcu et al., 2005 (6)	59	Talent	2	High-grade fever	Surgery	Died	Type 1 EL
Martens et al., 2007 (7)	64	Talent	3	High-grade fever	Surgery	Died	Infection
Santo et al., 2007 (8)	31	Jomed	8	Hematemesis	Surgery	Alive at 12 months	Pseudoaneurysm
Riesenman et al., 2007 (9)	52	TAG	4.5	High-grade fever	Surgery	Died	Infection
Girdauskas et al., 2008 (10)	NA	NA	49	Hematemesis	Surgery	Died	Type 1 EL
	NA	NA	3.5	NA	Surgery	Alive, NA	Erosion by stent graft
Christensen and Heyneman, 2009 (11)	79	NA	4	High-grade fever	Conservative	Died	Infection
lsasti et al., 2009 (12)	74	NA	24	High-grade fever	Conservative	Died	Infection
Eggebrecht, 2009 (2)	67	NA	6	Hematemesis	Re-TEVAR	Died	Type 1 EL
	49	NA	2	Hematemesis	Surgery	Died	NA
	52	NA	15	Hematemesis	Esophageal stent	Died	NA
Kim et al., 2010 (13)	75	NA	2	Hematemesis	Surgery	Died	NA
Sager et al., 2011 (14)	70	Valiant	24	Hematemesis	Conservative	Died	Aortic vasa vasorum bleeding
Albors et al., 2011 (15)	66	TAG	1	High-grade fever	Death before treatment	Died	NA
Kasai et al., 2011 (16)	54	Z+Dacron	36	High-grade fever	Conservative	Alive at 14 months	Ischemic necrosis
Numan et al., 2012 (17)	68	NA	4	High-grade fever	Conservative	Alive at 14 months	Infection
Canaud et al., 2011 (18)	86	TAG	18	Dysphagia	Surgery	Died	NA
Gavens et al., 2011 (19)	80	ZTX2	3	Hematemesis	Conservative	Died	Erosion by stent graft and infection
Yavuz et al., 2011 (20)	60	Talent	48	Hematemesis	Re-TEVAR	Died	NA
Current case	65	TAG	4	High-grade fever	Conservative	Died	Infection and pressure erosion

AEF, aortoesophageal fistula; EL, endoleak; NA, not applicable; TEVAR, thoracic endovascular aortic repair; Z+Dacron, Giantruco Z stent with Dacron graft; ZTX2, Zenith TX2.

The mortality rate of secondary AEF after TEVAR is extremely high, both with conservative (88.3%–100%) and surgical treatment (63.6%) (1. 2. 4). Conservative managements with broad-spectrum antibiotics and proton-pump inhibitor treatment, or secondary interventional treatment, such as total enteral feeding via percutaneous gastrostomy, esophageal stents and re-TEVAR, are frequently associated with a fatal outcome due to recurrent hemorrhage or chronic mediastinitis (2, 4, 12, 14-17, 19, 20). Thus, definitive surgical treatment involving effective debridement, esophageal resection and staged reconstruction, and aortic reconstruction using a graft appears to be the only effective method of saving patients with an AEF (3, 5, 8, 10). On the other hand, as most AEF patients have contraindications to open surgery, owing to their poor general condition or comorbidities, surgical therapy is not frequently undertaken, as was the case with our patient. CT-guided percutaneous drainage may have several benefits (17) to release the compression and to identify the exact organism that caused infection for selection of the appropriate antibiotic. In our case, combining conservative management with CT-guided percutaneous drainage of the infected aneurysm sac as a bridging management could have improved the patient outcome.

In conclusion, a secondary AEF after TEVAR is a catastrophic complication that is not easily treated; still, because conservative management has not been effective, it should be attempted with some form of invasive treatment. Close and regular follow-up after TEVAR by endoscopy and CT to detect the early signs of AEF should be performed, especially in the case of a huge aneurysm sac.

Conflict of interest disclosure

The authors declared no conflicts of interest.

References

- 1. Chiesa R, Melissano G, Marone EM, Marrocco-Trischitta MM, Kahlberg A. Aorto-oesophageal and aortobronchial fistulae following thoracic endovascular aortic repair: a national survey. Eur J Vasc Endovasc Surg 2010; 39:273–279.
- 2. Eggebrecht H, Mehta RH, Dechene A, et al. Aortoesophageal fistula after thoracic aortic stent-graft placement: a rare but catastrophic complication of a novel emerging technique. JACC Cardiovasc Interv 2009; 2:570–576.
- 3. Hance KA, Hsu J, Eskew T, Hermreck AS. Secondary aortoesophageal fistula after endoluminal exclusion because of thoracic aortic transaction. J Vasc Surg 2003; 37:886–888.
- 4. Eggebrecht H, Baumgart D, Radecke K, et al. Aortoesophageal fistula secondary to stent-graft repair of the thoracic aorta. J Endovasc Ther 2004; 11:161–167.
- 5. Czerny M, Zimpfer D, Fleck T, et al. Successful treatment of an aortoesophageal fistula after emergency endovascular thoracic aortic stent-graft placement. Ann Thorac Surg 2005; 80:1117–1120.
- Porcu P, Shavanon O, Sessa C, et al. Esophageal fistula after endovascular treatment in a type B aortic dissection of the descending thoracic aorta. J Vasc Surg 2005; 41:708–711.
- Martens K, De Mey J, Everaert H, Delvaux G, van Den Brande P. Aortoesophageal fistula following endovascular exclusion of a thoracic aneurysm. Int Angiol 2007; 26:292–296.
- Santo KC, Guest P, McCafferty I, Bonser RS. Aortoesophageal fistula secondary to stentgraft repair of the thoracic aorta after previous surgical coarctation repair. J Thorac Cardiovasc Surg 2007; 134:1585–1586.
- Riesenman PJ, Farber MA, Mauro MA, Selzman CH, Feins RH. Aortoesophageal fistula after thoracic endovascular aortic repair and transthoracic embolization. J Vasc Surg 2007; 46:789–791.

- 10. Girdauskas E, Falk V, Kuntze T, et al. Secondary surgical procedures after endovascular stent grafting of the thoracic aorta: successful approaches to a challenging clinical problem. J Thorac Cardiovasc Surg 2008; 136:1289–1294.
- Christensen JD, Heyneman LE. Aortoesophageal fistula complicating thoracic aortic aneurysm stent graft repair. Semin Roentgenol 2009; 44:4–7.
- Isasti G, Gomez-Doblas JJ, Olalla E. Aortoesophageal fistula: an uncommon complication after stent-graft repair of an aortic thoracic aneurysm. Interact Cardiovasc Thorac Surg 2009; 9:683–684.
- Kim HW, Suh JH, Jo KH, Yoon JS. Concomitant aortoesophageal and aortobronchial fistula after endovascular aortic repair. Ann Thorac Surg 2010; 90:2062.
- 14. Sager HB, Wellhöner P, Wermelt JA, Schunkert H, Kurowski V. Lethal hemorrhage caused by aortoesophageal fistula secondary to stent-graft repair of the thoracic aorta. Cardiovasc Intervent Radiol 2011; 34:60–63.
- Albors J, Bahamonde JA, Sanchis JM, Boix R, Palmero J. Aortoesophageal fistula after thoracic stent grafting. Asian Cardiovasc Thorac Ann 2011; 19:352–356.
- 16. Kasai K, Ushio A, Tamura Y, et al. Conservative treatment of an aortoesophageal fistula after endovascular stent grafting for a thoracic aortic aneurysm. Med Sci Monit 2011; 17:39–42.
- Numan F, Gulsen F, Cantasdemir M, Solak S, Arbatli H. Percutaneous treatment of an infected aneurysm sac secondary to aortoesophageal fistula with a history of stent-graft treatment for thoracic aortic aneurysm. Cardiovasc Intervent Radiol 2012; 35:690–694
- Canaud L, Alric P, Gandet T, et al. Surgical conversion after thoracic endovascular aortic repair. J Thorac Cardiovasc Surg 2011; 142:1027–1031.
- Gavens E, Zaidi Z, Al-Jundi W, Kumar P. Aortoesophageal fistula after endovascular aortic aneurysm repair of a mycotic thoracic aneurysm. Int J Vasc Med 2011; 2011:649592.
- 20. Yavuz S, Kanko M, Ciftci E, et al. Aortoesophageal fistula secondary to thoracic endovascular aortic repair of a descending aortic aneurysm rupture. Heart Surg Forum 2011; 14:249–251.