Conservative endoscopic closure with fibrin glue of an aorto-esophageal fistula secondary to endovascular repair of a contained penetrating atherosclerotic ulcer rupture

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ABSTRACT

An aorto-esophageal fistula (AEF) is a rare complication of aortic surgery but can cause potentially lethal upper gastrointestinal tract bleeding. A patient presented with an AEF secondary to emergency endovascular repair of a contained penetrating atherosclerotic ulcer rupture of the thoracic aorta and was successfully treated with endoscopic closure using fibrin glue. As endovascular repair becomes increasingly common, a greater incidence of AEFs should be anticipated and the treatment options better described. (J Vasc Surg Cases Innov Tech 2023;9:101354.)

Keywords: Aorta; Aortic ulcer; Endoscopy; Endovascular procedures; Esophageal fistula; Esophagus vacuum therapy; TEVAR; Vasculitis

An aorto-esophageal fistula (AEF) is an anomalous connection between the thoracic aorta and the esophagus. An AEF can cause serious hematemesis, which is distinguished from other gastrointestinal tract bleeding by being bright red in color representing arterial bleeding.[1] The first report of an AEF was caused by ingestion of a foreign body and >500 other causes have been described, including esophageal malignancy, surgical complication, tuberculosis, corrosive esophagitis, and congenital anomalies. However, >70% are associated with thoracic aorta aneurysms, specifically as a complication of open and, perhaps especially, endovascular repair.[2,3] As thoracic endovascular aortic repair (TEVAR) becomes increasingly common, an increased AEF incidence should be anticipated.[4] Possible AEF etiologies should be considered carefully and confirmed by a range of clinical, laboratory, and imaging assessments because the diagnosis will determine which treatment options are available and increase the chance of a successful outcome.

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An AEF developing after TEVAR could be associated with ischemic necrosis and erosion caused by compression during procedural manipulation or stent graft damage to the aortic wall and infection by microorganisms. A typical treatment strategy combines control of bleeding, radical debridement, and aortic or esophageal reconstruction.[5]

The participant provided written informed consent for the report of her case details and imaging studies. Data from this case report are available from the patient's medical records archived at University Medical Center Mainz, Johannes Gutenberg University, Mainz, Germany on request.

CASE REPORT

A 72-year-old patient presented to an external primary care hospital with joint pain, an elevated temperature, nausea, and vomiting that had lasted for several days. Her symptoms worsened with acute onset of thoracic and back pain, shortness of breath, and a feeling of being trapped, accompanied by sweating. The patient also reported loss of appetite and unwanted weight loss. She did not have hypertension, coronary heart disease, or diabetes. She had no history of alcohol or tobacco consumption. However, her physical examination revealed no abnormalities. Her body temperature was 36.6°C; blood pressure, 110/60 mm Hg; and heart rate, 82 beats/min. The laboratory tests revealed an elevated white blood cell count (11.40 tsd/μL; normal range, 4-11 tsd/μL), low hemoglobin (11.9 g/dL; normal range, 12-16 g/dL), and elevated D-dimer (1361.0 ng/mL; normal range, 0-500 ng/mL), with a normal procalcitonin value. Radiography initially ruled out a pulmonary infection. Computed tomography (CT) revealed a contained penetrating atherosclerotic ulcer (PAU) rupture in the thoracic aorta measuring ~1 × 2 cm just distal to the origin of the left subclavian artery (LSA) on the lesser curvature side with a paratracheal

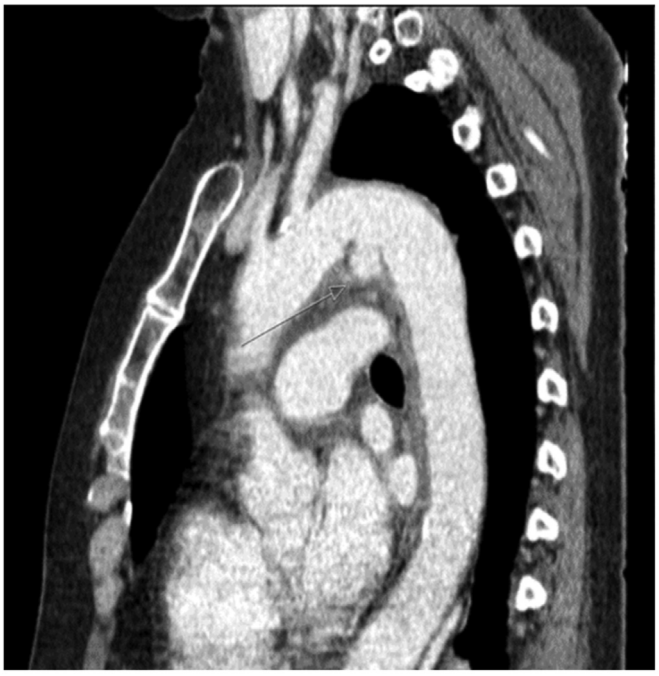

Fig 1. Computed tomography (CT) scan of the chest (sagittal view) showing a contained penetrating atherosclerotic ulcer (PAU) rupture (*arrow*) distal to the origin of the left subclavian artery (LSA).

hematoma (Fig 1). Degenerative changes in the spinal column were also noted.

She was transferred to our department for urgent TEVAR (32 × 164 × 32 mm; RelayPro NBS; Terumo Aortic) in zone 2. TEVAR was performed percutaneously under general anesthesia. The LSA was revascularized via a carotid–subclavian bypass using a 7-mm Gelsoft Plus graft (Terumo Aortic), and the proximal part of the LSA was occluded using a vascular 12-mm Amplatzer plug (Abbott Cardiovascular; Fig 2, *A*). We do not routinely use cerebrospinal fluid drainage prophylactically if <20 cm of aorta is covered and perfusion of the left carotid artery is maintained. The postoperative course was unremarkable, except for an asymptomatic C-reactive protein (CRP) elevation, which regressed with administration of an antibiotic (cefuroxime, 500 mg intravenously, twice daily). A CT scan on postoperative day 3 revealed a well-positioned aortic stent with no signs of endoleaks or extravasation, with otherwise normal findings (Fig 2, *B*). She was discharged on postoperative day 5 without antibiotics.

One month later, the patient presented with chest pain and dyspnea on exertion. CT showed no changes, and the laboratory test results continued to show elevated CRP levels. Blood cultures were positive for *Streptococcus anginosus,* and antibiogram-based treatment was initiated. Subsequent blood cultures showed no microbes; however, her CRP values remained elevated. Thus, vasculitis was suspected and diagnosed. Cortisone shock therapy led to a clear improvement in her general condition, with a reduction in CRP values. Magnetic resonance imaging of the entire aorta was performed and showed wall thickening with increased contrast enhancement of the descending thoracic aorta, compatible with inflammatory changes in the vessel wall, indicative of large-vessel vasculitis, most likely giant cell arteritis (Fig 3, *A*).

Cortisone therapy was continued orally, with additional administration of tocilizumab. Three weeks later (2 months after TEVAR), a follow-up CT scan revealed some air bubbles and fluid accumulation in the upper mediastinum and along the descending thoracic aorta, representative of mediastinitis with suspected esophageal or bronchial perforation (Fig 3, *B*). Bronchoscopy excluded a bronchial fistula; however, a 2-mm esophageal fistula was detected endoscopically 24 cm from the dental line (Fig 4, *A*).

A multidisciplinary discussion recommended open surgical repair, including radical debridement of the infected tissue and replacement of the aorta and esophagus. The patient refused the operation and preferred conservative therapy.

We proceeded with cortisone therapy (prednisolone; initially 3 × 250 mg intravenously, followed by 60 mg orally, and, currently, 4 mg daily) for large-vessel vasculitis, in addition to antibiotic therapy (amoxiclav, 875 mg/12 mg twice daily). After serial endoscopic vacuum treatment of the esophagus with four Endo-SPONGE (B. Braun) changes over 15 days. The sponges were cut to size to cover the fistula and esophagus and held in place by applying an appropriate vacuum pressure. A tube was passed through the nose to provide continuous drainage and prevent accumulation of secretions and maintain the vacuum. The treatment was performed with enteral food restriction. With this parenteral regimen, the fistula was successfully sealed endoscopically through the esophagus by application of 1 mL of fibrin glue (Beriplast; CSL Behring; Fig 4, *B*).

The patient was discharged in good condition with oral antibiotics (amoxiclav, 875 mg/12 mg twice daily during follow-up) and continued well when followed up at 1 year. At 2 years postoperatively, she is symptom-free with normal inflammatory parameters. Her most recent CT scan showed a reduction in the mediastinitis and air bubbles, with no residual findings. The entire aorta and supra-aortic branches were visualized, and the carotid–subclavian bypass was patent. Also, the PAU was completely sealed and smaller. On endoscopic evaluation, the fistula had healed with normal scar tissue and focal complete intestinal metaplasia, histologically consistent with type C gastritis, with no evidence of malignancy (Fig 5). The patient developed osteoporosis and is currently receiving pharmaceutical treatment.

DISCUSSION

An AEF secondary to vasculitis is a rare complication but with typically severe consequences; thus, a rapid diagnosis and an appropriate treatment strategy are critical.[6,7] The incidence after TEVAR is reported to be 1.7%. Both open surgical and endovascular management are reported to be associated with high mortality (64%); however, conservative management is usually fatal.[8,9] Its management,

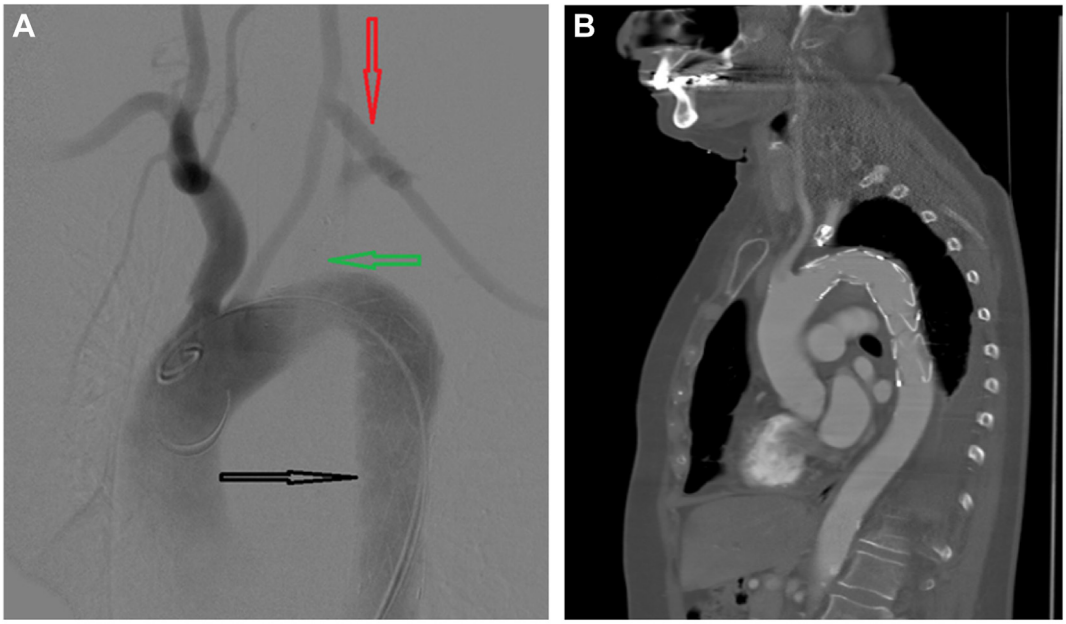

Fig 2. A, Intraoperative angiogram of the thoracic aorta showing the implanted stent graft (*black arrow*), carotid–subclavian bypass (*red arrow*), and occluded origin of the left subclavian artery (LSA; *green arrow*). **B,** Computed tomography (CT) scan (sagittal view) of the chest showing the thoracic aorta after implantation of the stent graft excluding the contained penetrating atherosclerotic ulcer (PAU) rupture.

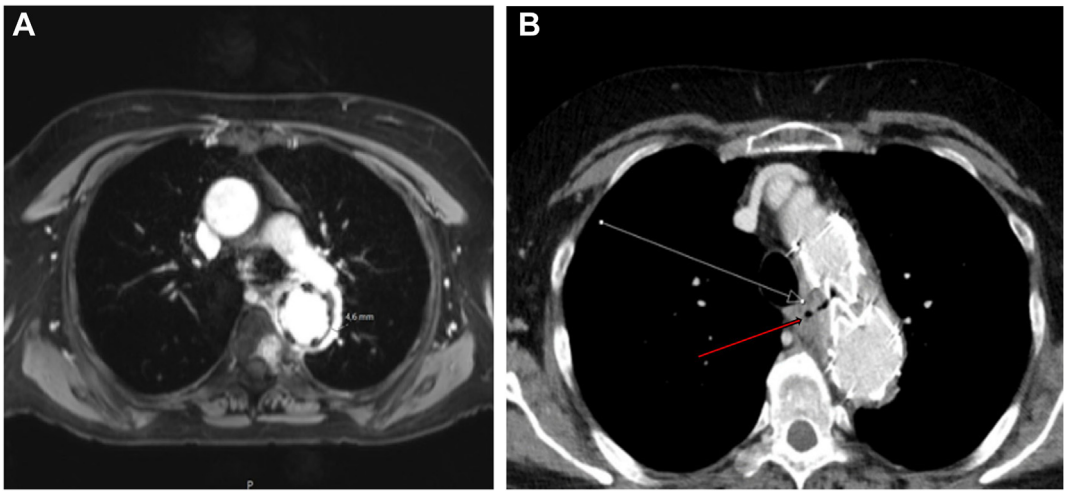

Fig 3. A, Magnetic resonance image showing wall thickening compatible with inflammatory changes and suggesting the presence of large-vessel vasculitis. **B,** Computed tomography (CT) scan of the chest showing some air bubbles (*red arrow*) and fluid accumulation (*black arrow*) in the mediastinum along the descending aorta.

especially after previous endovascular repair, is associated with high morbidity and mortality. Surgical debridement and replacement of an infected segment could be necessary. A wide spectrum of graft materials for extra-anatomic or in situ reconstruction is available, including homograft, biologic or biosynthetic material, and antibiotic-impregnated grafts. However, none of these is universally applicable due to the associated risks.

A pseudoaneurysm as an indication for TEVAR has been reported as a significant risk factor for fistulization. Our patient showed no signs of an endoleak or stent graft-induced aortic wall injury. Thus, it might have been that the PAU caused a similar compression and local inflammatory response.[9]

The most important goal of treating an AEF is to prevent massive hemorrhage. Although in our case, the

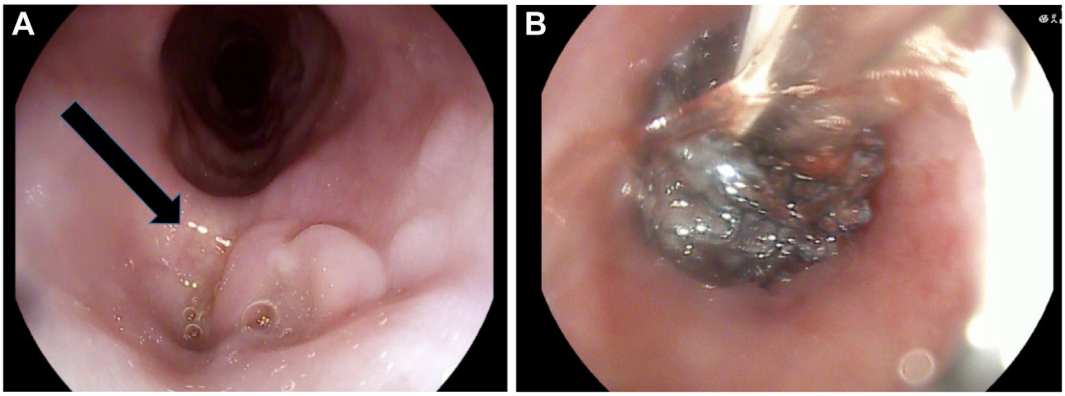

Fig 4. A, Endoscopy showing the origin of the aorto-esophageal fistula (AEF). **B,** A vacuum sponge is placed endoscopically in the esophageal lumen in front of the fistula opening.

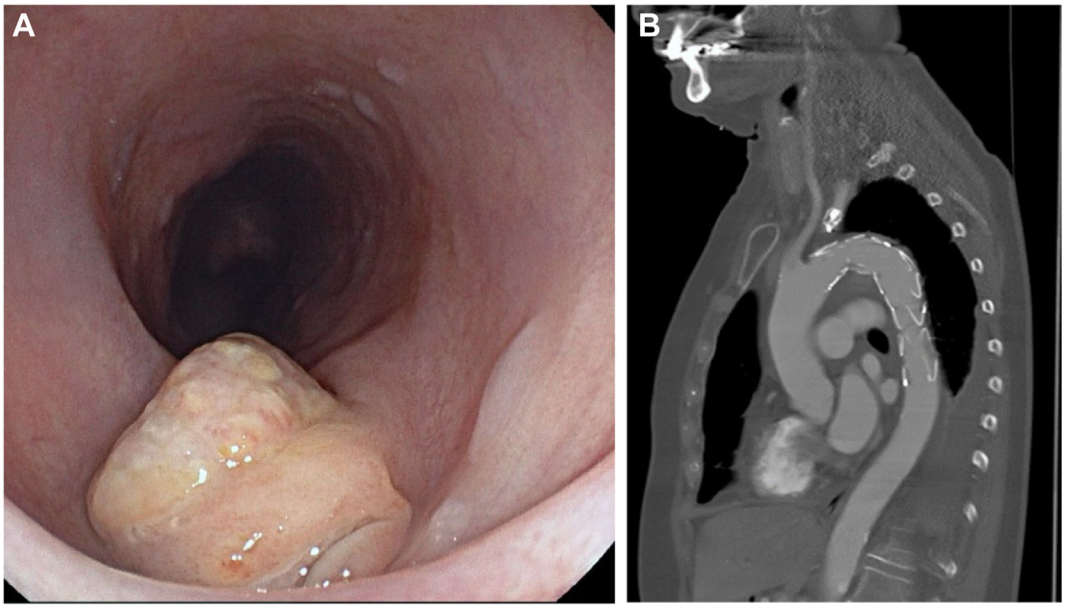

Fig 5. A, Endoscopy imaging showing granulation or glycogenic acanthosis (small raised white plaques) and healed aorto-esophageal fistula (AEF) 2 years after successful closure using fibrin glue. **B,** Computed tomography (CT) scan showing the thoracic aorta 2 years after successful endovascular exclusion of the contained penetrating atherosclerotic ulcer (PAU) rupture and successful sealing of the AEF.

patient had no signs of bleeding, hemorrhage could have occurred at any point. Furthermore, the imminent danger of an infection of the pre-existing aortic stent at the level of the AEF in our patient was also a major concern. In the literature, surgical treatment of AEFs, consisting of thoracic exploration, repair of the aorta, partial esophageal resection, and esophageal reconstruction with stomach and omental flaps, is the gold standard.[10]

The treatment decision-making process should depend on the patient's specific situation. In the present case, treatment was chosen in accordance with the patient's desire, who refused exploratory thoracotomy. However, at 1 year of follow-up, she reported being symptom free, and CT imaging showed a well-positioned thoracic aortic stent graft. In addition, esophagogastroduodenoscopy showed the esophageal fistula had completely sealed.

CONCLUSIONS

The incidence of AEFs is likely to increase with the widespread adoption of TEVAR. It is important, therefore, that awareness of this rare, but serious, complication contributes to a faster diagnosis and that the treatment options are well described. Open surgical repair remains the first option to consider; however, endoscopic vacuum therapy with oral antibiotics is also feasible and could be an appropriate option for some patients.

DISCLOSURES

None.

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