Clinical Case Study

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PRIMARY AORTOESOPHAGEAL FISTULA DUE TO ESOPHAGEAL FOREIGN BODY: A CASE REPORT

LeAnne Vitito, MS, RN, CGRN, APRN
Department Editor

Jin-Wen Liao, BS
Wen-Xiu Long, BS
Wen-Li Shen, BS

Background

Aortoesophageal fistula (AEF) is a rare but life-threatening condition that causes massive gastrointestinal hemorrhage (Lee, Jang, Kim, Cha, & Choi, 2019). It is imperative to recognize these bleeds as early as possible because the reported mortality approaches 77% with intervention and is 100% fatal without treatment (Parikh, Ali, & Wong, 2015; Yang, Hu, & Peng, 2018). Here, we report a case of a 75-year-old man who was diagnosed with a primary AEF due to hematemesis for 4 hours and was successfully treated with intervention.

Case Presentation

A 75-year-old man was admitted to our hospital because of hematemesis for 4 hours. The patient vomited about 400 ml of bright red blood with clots, accompanied by dizziness and fatigue. He did not have hypertension, coronary heart disease, or diabetes. He was a smoker for 30+ years, smoking five

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About the authors: *Jin-Wen Liao, BS, is Attending Doctor, Department of Gastroenterology, The People's Hospital of Jianyang City, Jianyang, Sichuan Province, China.*

Wen-Xiu Long, BS, is Associate Professor of Nursing, Department of Gastroenterology, The People's Hospital of Jianyang City, Jianyang, Sichuan Province, China.

Wen-Li Shen, BS, is Nurse-in-Charge, Department of Gastroenterology, The People's Hospital of Jianyang City, Jianyang, Sichuan Province, China.

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The authors declare that they have no competing interests.

Correspondence to: *Wen-Xiu Long, BS, Department of Gastroenterology, The People's Hospital of Jianyang City, No. 196, Hospital Rd, Jianyang 641400, Sichuan Province, China (531646739@qq.com).*

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cigarettes per day, but he had since quit smoking. He had no history of drinking or abdominal pain, and he denied chest pain. On admission, his body temperature was 36.7 °C, blood pressure 110/60 mmHg, and heart rate 78 beats/min. Other physical examination parameters were unremarkable.

Laboratory data detected the following abnormalities: white blood cell (WBC) count, 13.51 × 10^9/L (85.4% neutrophils); and hemoglobin, 109 g/L (normal range: 120–160 g/L). Esophagogastroduodenoscopy (EGD) revealed an esophageal submucosal tumor-like protrusion with an ulcerative lesion 26 cm below the incisors (Figures 1A and 1B). These findings raised the suspicion of an AEF, which was confirmed by chest computed tomography angiography (CTA), which revealed the pseudoaneurysm of the descending aortic arch, swelling of the wall of the descending aorta, and a blurred surrounding space (Figures 1C and 1D). Asked again about his medical history, he revealed that he had accidentally swallowed a fishbone 10 days earlier and forced it down with food. He had retrosternal discomfort and no dysphagia following the incident, but he did not seek medical advice until the onset of bleeding. Therefore, primary AEF was diagnosed, which was caused by fishbone injury to the esophagus.

After multidisciplinary discussion, the patient was referred to vascular surgery and underwent urgent thoracic endovascular aortic repair (TEVAR). He was administered general anesthesia to undergo aortic angiography through the right femoral artery using digital subtraction angiography. All branches of the aortic arch were found to be intact. The descending aorta issued the left subclavian artery about 2 cm away and found a cystic protuberance about 2.0 × 4.0 cm in size. Furthermore, the patient was successfully implanted with a MicroPort HT 26-22-160 covered stent through the right femoral artery. The operation went well, and the incision found no contrast agent.

After surgery, the patient underwent fasting, received antibiotic treatment (intravenous imipenem and cilastatin sodium [0.5 g] once per 8 hours for 2 weeks), and reduced vascular shear tension (metoprolol [25 mg] once per 12 hours for 2 months, 12.5 mg for long-term maintenance).

The postoperative CTA after 2 weeks showed complete sealing of the primary entry tear with a thoracic aortic stent and no contrast extravasation from the aortic arch into the mediastinum and esophagus (Figures 2A and 2B). Thereafter, he reported no rebleeding episodes and was in good condition.

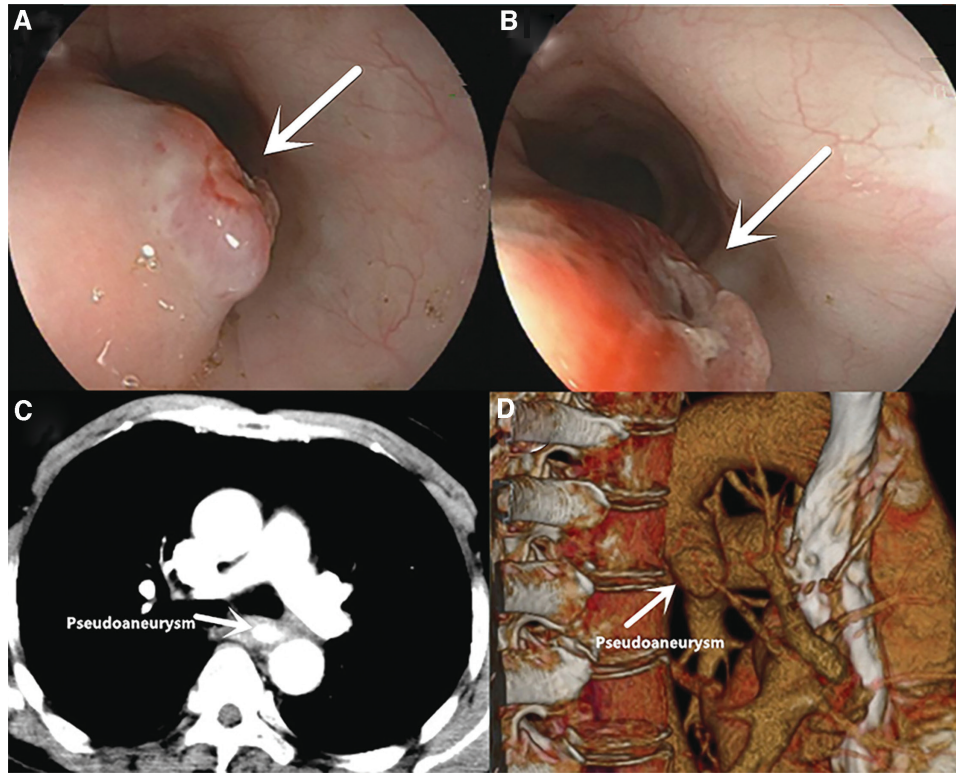

FIGURE 1. The presurgical examination. (A, B) Esophagogastroduodenoscopy shows an esophageal submucosal tumor-like protrusion with an ulcerative lesion 26 cm below the incisors. (C) Transverse and (D) three-dimensional volume-rendered computed tomography angiography shows the pseudoaneurysm of the descending aortic arch, swelling of the wall of the descending aorta, and blurred surrounding space.

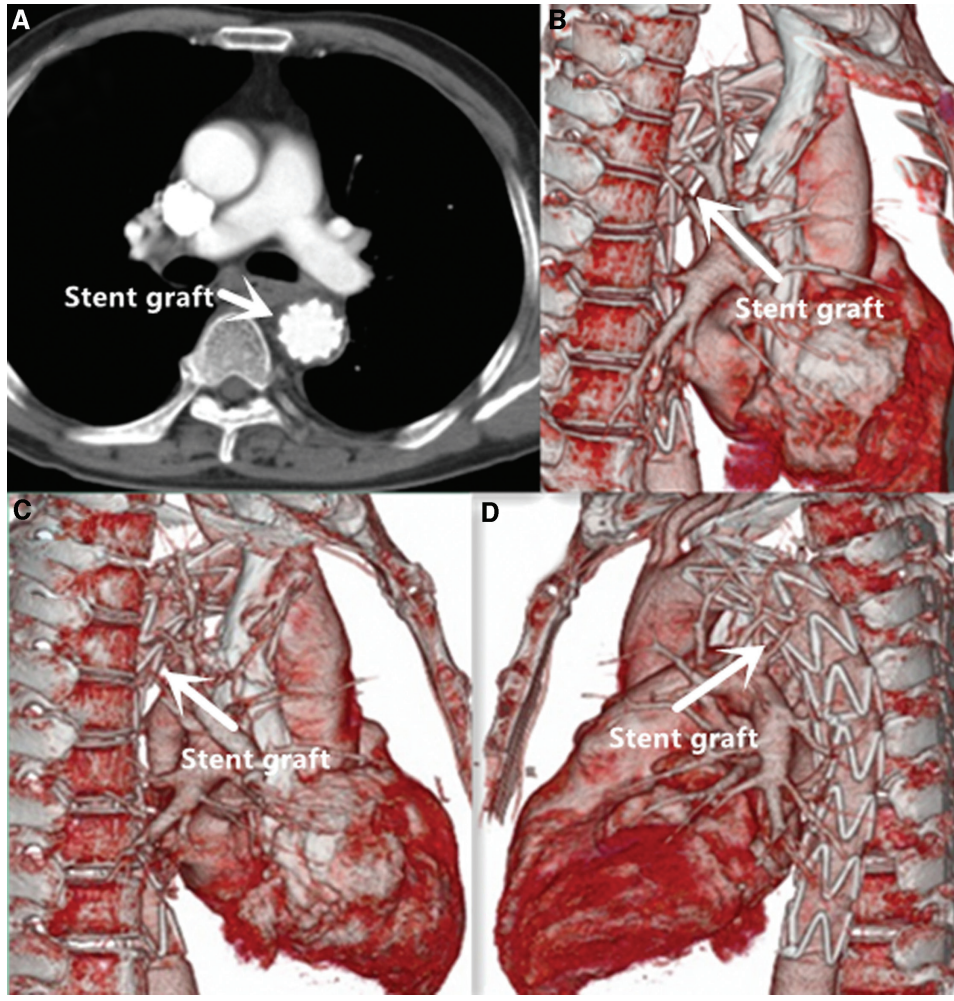

FIGURE 2. The postsurgical examination. (A) Transverse and (B) three-dimensional volume-rendered computed tomography angiography of predischarge shows complete sealing of the primary entry tear with a thoracic aortic stent. No contrast extravasation from the aortic arch into the mediastinum and the esophagus. (C, D) Three-dimensional volume-rendered computed tomography angiography after 3 months shows a well-positioned thoracic aortic stent. No contrast extravasations into the aneurysmal sac.

The postoperative CTA after 3 months showed a well-positioned thoracic aortic stent and no contrast extravasations into the aneurysmal sac (Figures 2C and 2D). Based on the patient's age and financial status, he and his family refused to undergo exploratory thoracotomy. Two years later, the postoperative CTA showed a well-positioned thoracic aortic stent and no contrast extravasations into the aneurysmal sac, and it remained in a similar state as the previous examination without any significant change (Figures 3A and 3B). Esophagogastroduodenoscopy showed an esophageal submucosal tumor-like protrusion with an ulcerative lesion that was completely healed (Figure 3C). Ultrasound gastroscopy showed a strong half-moon echo that was seen between the aortic wall and the esophageal wall (Figure 3D).

This case study was approved by the ethics committee of The People's Hospital of Jianyang City. The written informed consent was obtained from the patient for publication.

Discussion

Aortoesophageal fistula is an abnormal anatomical communication between the aorta and esophagus. Its annual incidence is estimated to be 0.007 per million (Zhan & Xu, 2019). It could be divided into primary and secondary based on its cause. Primary AEF arises from several underlying diseases such as thoracic aortic aneurysm, foreign bodies, and esophageal malignancy, whereas secondary AEF represents a well-known complication resulting from aortic or esophageal surgery and graft placement (Hollander & Quick, 1991; Yang et al., 2018).

The first case of AEF was reported by Dubrueil in 1818 (Carter, Mulder, Snyder, & Brewer, 1978). In 1914, Chiari first described the AEF symptom triad of

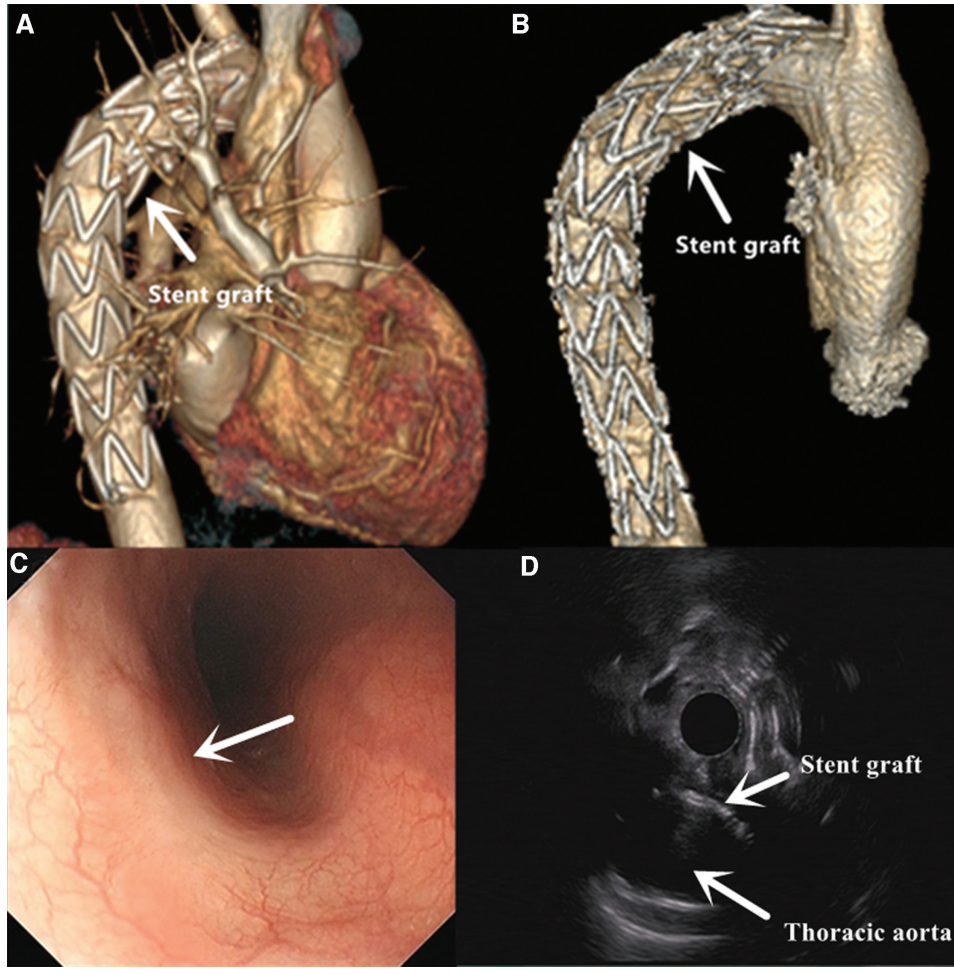

FIGURE 3. Examination 2 years later. (A, B) Three-dimensional volume-rendered computed tomography angiography shows a well-positioned thoracic aortic stent. No contrast extravasations into the aneurysmal sac. (C) Esophagogastroduodenoscopy shows a completely healed esophageal submucosal tumor-like protrusion with an ulcerative lesion. (D) Ultrasound gastroscopy shows a strong half-moon echo between the aortic wall and the esophageal wall.

mid-thoracic pain, sentinel arterial hemorrhage, and a symptomless period, followed by exsanguination (Wei et al., 2015). According to a literature review by Hollander and Quick (1991), 65% of AEFs have a sentinel arterial hemorrhage and 59% of patients recall a history of mid-thoracic pain. In this case, the patient developed hematemesis and had a history of a foreign body esophageal injury and chest pain 2 weeks before. Therefore, we retrospectively presumed his bleeding to be due to sentinel arterial hemorrhage. Secondary infection due to a foreign body may result in pseudoaneurysms and spreading necrotic tissue. The aortic wall was gradually eroded and ultimately created an AEF.

Because AEF is not a common cause of upper gastrointestinal hemorrhage, early diagnosis may be difficult (Hwang & Cho, 2019). Although EGD is useful for the diagnosis of AEFs in patients with gastrointestinal hemorrhage, its diagnosis rate is low at around 25% (Kayashima et al., 2019). Also, it should be noted that EGD can irritate blood clots, leading to severe re-hemorrhage (Hwang & Cho, 2019). The characteristic finding of AEF on EGD is a submucosal tumor-like protrusion, ulcerative lesion, or pulsating protrusion with central fistula (Yang et al., 2018). In this case, the findings on EGD were highly suggestive for AEF and CTA was necessary for confirmation. The usefulness of CTA in this context was recently highlighted, with a diagnosis rate of 30%–61% (Kayashima et al., 2019). The CTA scan revealed not only the classic AEF signs (i.e., ectopic gas adjacent to the aortic lumen and effacement of the periaortic fat plane), but also more characteristic and rare signs of direct extravasation of vascular contrast into the esophagus (Monteiro, Martins, Martins da Cunha, Moleiro, & Patrício, 2020).

Whether the hemorrhage is sentinel or fatal, in either case there is penetration of the aorta (Zhang et al., 2011). Therefore, once the diagnosis of AEF is confirmed, treatment should be started without delay. The most important goal of treating AEF is to prevent massive hemorrhage. According to some literature reports, TEVAR is a type of endoluminal aortic stent that was

developed recently as minimally invasive therapy for AEF (Shen et al., 2018; Takeno, Ishii, Nanashima, & Nakamura, 2020). Endovascular repair is a less invasive method, which refers to the insertion of a covered stent through the femoral arteries to fix the normal aorta above and below the aneurysm, acting as a cannula through the aneurysm sac (Kent, 2014). In this case, although the patient was at a symptomless interval, massive hemorrhage could have occurred at any time. Therefore, we performed TEVAR without hesitation. According to some literature reports, there is an increased probability of secondary AEF after TEVAR (Sugiyama et al., 2020; Zhu, MacArthur, Lui, & Lee, 2020), but our patient was followed up for 2 years without hemorrhage or secondary AEF.

According to a report that for AEF with pseudoaneurysm, the optimal treatment is initial endovascular stent graft placement to control bleeding followed by thoracic exploration, aortic repair, partial esophageal resection, and esophageal reconstruction with stomach and omental flaps (Wei et al., 2015). In this case, based on the patient's age and financial status, he underwent TEVAR but refused exploratory thoracotomy. However, at 2 years' follow-up, he reported no rebleeding episodes after TEVAR, and CTA showed a well-positioned thoracic aortic stent. Also, EGD showed the esophageal submucosal tumor-like protrusion with an ulcerative lesion was completely healed. There are some reports of patients with AEF who have been successfully treated by interventional therapy alone (Kayashima et al., 2019; Monteiro et al., 2020; Takeno et al., 2020).

Conclusion

Aortoesophageal fistula with pseudoaneurysm induced by an esophageal foreign body is rare but has severe complications. The treatment decision-making process should depend upon the patient's specific situation. Thoracic endovascular aortic repair not only becomes bridge therapy for hemostasis but also may become one of the main therapies for AEF.

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