Successful Endovascular Repair of an Aortoesophageal Fistula

An alternative treatment for a patient who is not a candidate for open repair.

BY MOJTABA GASHTI, DO, FACOS; GIANCARLO McEVENUE, BSc, MSIII; AND JASON M. RADECKE, MD

Primary aortoesophageal fistula (AEF) is a rare cause of massive upper gastrointestinal (GI) bleeding. The reported incidence of AEF is .01% to .07% of the general population.[1-3] Chiari first described the syndrome of AEF in 1914 as a triad of midthoracic pain and/or dysphagia, followed by a "herald" hemorrhage, and then massive fatal hematemesis.[4] Early diagnosis and surgical intervention are mandatory for survival, because the probability of fatality is 100% without intervention.[5] Nevertheless, despite various surgical interventions for AEF, few survivals have been reported.[6] We report a case of AEF due to a descending thoracic aortic pseudoaneurysm (TAP) that was successfully treated with a stent graft after initial control of a hemorrhage with a Sengstaken-Blakemore tube (SBT).

CASE REPORT

An 80-year-old man originally presented for elective revision of a right total knee arthroplasty. On postoperative day 8, he complained of epigastric pain with minor hematemesis and melena. An esophagogastroduodenoscopy (EGD) revealed gastritis and duodenal ulcers. This was not treated during the EGD due to coagulopathy (international normalized ratio, 2.9), and warfarin was discontinued.

Three days later, the patient's hematocrit dropped from 29 to 18 due to hematemesis. A second EGD was unsuccessful in controlling the bleeding, and an emergent laparotomy with oversew of the duodenal ulcer was performed. Twelve days later, he again had massive hematemesis. Another EGD revealed an active arterial bleed from the midesophagus. He then developed a cardiopulmonary collapse and was intubated. We began resuscitation and inserted an SBT. A CT scan revealed a 7-cm TAP with evidence of fistulization into the esophagus (Figure 1). The patient was deemed to be at prohibitive risk for open surgical repair, and therefore, the option of endovascular repair was offered to the family.

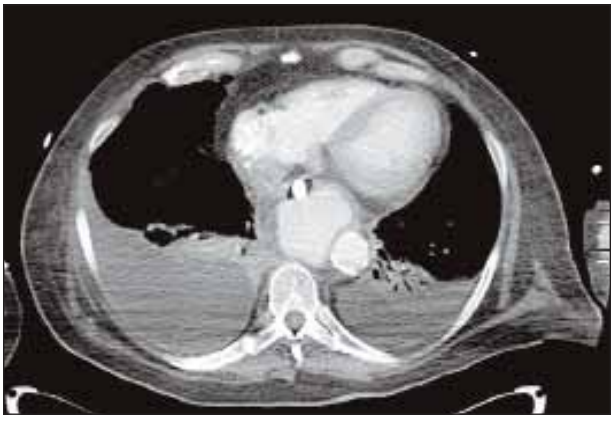

Figure 1. CT image revealing a large pseudoaneurysm extending from the midesophagus.

A preoperative CT scan indicated small iliofemoral vessels not suitable for insertion of the required 22-F sheath. Therefore, we planned a right retroperitoneal approach to create a conduit. The common iliac arteries were circumferentially calcified, and the distal aorta was exposed where 1.5 cm of soft aortic wall was discovered. A sheath was then inserted through a purse-string suture in the anterior aortic wall. An angiogram confirmed the presence of a large pseudoaneurysm extending from the medial aspect of the middescending thoracic aorta (Figure 2A). Next, a 31- X 150-mm TAG endoprosthesis (W. L. Gore & Associates, Flagstaff, AZ) was deployed under fluoroscopy. A completion angiogram showed complete exclusion of the pseudoaneurysm (Figure 2B). A repeat CT scan 4 days later revealed thrombosis of the pseudoaneurysm without evidence of perigraft infection (Figure 3). Although the patient remained hemodynamically stable, definitive treatment of the esophagus had to be postponed. An infectious disease consultation was obtained, and the patient was kept

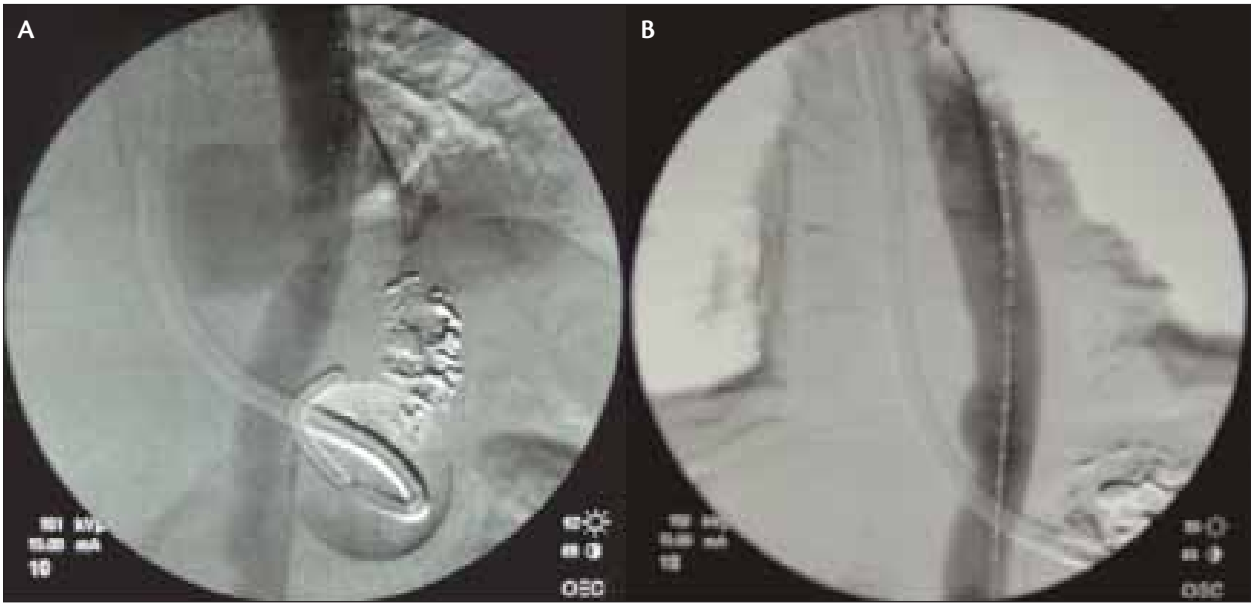

Figure 2. Intraoperative angiogram showing the thoracic aortic pseudoaneurysm (A). Completion angiogram indicating exclusion of the pseudoaneurysm (B).

on broad-spectrum antibiotics. A percutaneous gastrostomy tube was placed for nutritional support. The patient was considered to be at extremely high risk for definitive treatment of the esophagus, and therefore it was never treated. The patient subsequently had a tracheostomy, and approximately 3 weeks later, he was transferred to a rehabilitation facility. Unfortunately, the patient died approximately 4 months later. No autopsy was performed, but there never was an indication of systemic infection, although that remained a potential concern.

DISCUSSION

AEF is a rare cause of massive GI bleeding, but the consequences are often catastrophic. AEF constitutes approximately 10% of all primary aortoenteric fistulae (PAEF). Fewer than 400 cases of PAEF have been reported, with the majority of patients dying before a definitive diagnosis is made, indicating that the true incidence may be even higher.

The leading cause of AEF is erosion of a thoracic aortic aneurysm (TAA) into the esophagus.[6] Other causes of AEF include esophageal and bronchial neoplasm, complications of thoracic aortic surgery, and ingestion of foreign objects.[7] Before the 1970s, almost 25% of all PAEF were caused by tuberculosis and syphilis.[8] With effective treatment of these infections, only 4% of all PAEF were caused by infectious processes between 1994 and 2004.[1]

Our patient had an episode of hematemesis, midthoracic pain, and dysphagia followed by a near-fatal exsanguination episode, thus exhibiting all of Chiari's triad. Thirty percent of patients experience a massive bleed within 6 hours of the initial episode. Spontaneous cessation of the herald bleed may be due to temporary occlusion of the fistula from arterial wall spasm, intravascular hypotension, or occlusion by the periaortic hematoma. Therefore, aggressive resuscitation should be avoided, but controlled hypotension with a mean arterial pressure of 60 mm Hg could prove lifesaving. The tamponade effect of SBTs in the esophagus has previously been reported to control bleeding in AEF.[9,10] In our case, it allowed time for an accurate diagnosis and placement of a stent graft to occlude the fistulous defect.

Esophagoscopy is both sensitive and specific for AEF diagnosis; however, there have been reports of the endoscope dislodging existing clots, causing fatal bleeding.[11] The endoscopic findings that are consistent with AEF include a pulsating submucosal mass, bluish-gray mucosa due to submucosal blood/hematoma, foreign body, ulcer, or esophagitis, and rarely a fistula opening.[3] Endoscopy is nevertheless still necessary because it can exclude more common causes of upper GI bleeding including peptic ulcer disease, esophageal varices, arteriovenous malformations, Mallory-Weiss tears, and tumors.

An aortogram in stable patients may be useful; however, it seldom reveals a fistulous tract due to the absence of active bleeding. A CT scan of the chest is also a reliable and rapid approach for diagnosis. In our case, the CT scan delineated a large pseudoaneurysm displacing the esophagus. These two imaging modalities, together with upper GI bleeding, have been used to describe a diagnostic triad that strongly suggests AEF.[12]

Once AEF is identified or strongly suspected, surgical

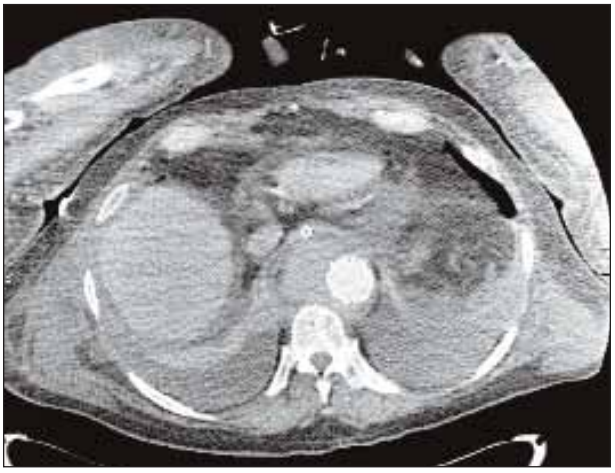

Figure 3. CT image indicating complete thrombosis of the pseudoaneurysm.

repair is mandatory. An extensive literature review by Prokakis et al[6] found only 37 cases of successful surgical or endovascular management of AEF from 1978 to 2008. Traditional AEF treatment consists of aneurysm excision, in situ or extra-anatomical bypass, and reconstruction of the alimentary tract. Open access makes it possible to control the aneurysm and the esophagus, as well as drainage of the potentially infected mediastinum. The open technique involves many risks including damage to the spinal cord, kidney, brain, and other viscera due to aortic cross-clamping and embolization.[13] Mortality rates associated with these procedures remain extremely high.

The feasibility of less invasive endovascular techniques for thoracic aortic pathology, including emergency repair of aortic rupture, has been established and deemed at least as good as conventional open repair.[13] Baril et al[3] reported decreased perioperative morbidity and mortality, shorter hospital stay, and acceptable survival rates in patients treated endovascularly for both primary and secondary AEF. Kato et al[14] were the first to report successful endovascular repair of an AEF in a patient with esophageal cancer. Due to our patient's moribund condition, endovascular management was undertaken. The major drawbacks to this approach include access issues due to the anatomy of the iliofemoral vessels, possible contamination of the graft with possible future hemorrhage from an infected endoprosthesis, and inability to simultaneously repair the esophagus.[3] These drawbacks may be of little importance to the elderly patient, because there is not a prolonged life expectancy due to existing comorbidities. As Patel et al[15] have recently demonstrated, this population of patients benefit from endovascular repair because of significant early benefits, including a trend toward lower mortality rates and a reduced length of hospitalization.

CONCLUSION

A high index of suspicion is necessary for prompt diagnosis and treatment of AEF. An AEF due to a TAP, once recognized or strongly suspected, can be initially controlled with an SBT. Endovascular repair in view of the literature appears to be a feasible alternative to open repair in certain patients. In this group of patients with significant comorbidities, endovascular repair may in fact be the treatment of choice. Although this approach may not be curative, it may allow the patient to recover from the initial insult and undergo an elective and more definitive repair at a later time. An obvious drawback would be the potential risk for endoprosthesis infection, for which long-term antibiotics and surveillance may be necessary. ■

Mojtaba Gashti, DO, FACOS, is from Union Memorial Hospital in Baltimore, MD. He has disclosed that he holds no financial interest in any product or manufacturer mentioned herein. Dr. Gashti may be reached at mojtaba.gashti@medstar.net.

Giancarlo McEvenue, BSc, MSIII, is a fourth-year medical student at SABA University School of Medicine in the Netherlands-Antilles. He has disclosed that he holds no financial interest in any product or manufacturer mentioned herein. Mr. McEvenue may be reached at g.mcevenue@gmail.com.

Jason M. Radecke, MD, is is from Union Memorial Hospital in Baltimore, MD. He has disclosed that he holds no financial interest in any product or manufacturer mentioned herein. Dr. Radecke may be reached at jradecke@gmail.com.

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