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Staged Surgical Treatment of Primary Aortoesophageal Fistula

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Aortoesophageal fistula (AEF) is a rare and potentially fatal disease that causes massive gastrointestinal bleeding. Therefore, early diagnosis and treatment are essential to prevent mortality. Controlling the massive bleeding is the most important aspect of treating AEF. The traditional surgical treatment was emergent thoracotomy, but intraoperative or perioperative mortality was high. We report a case of a patient presenting with hematemesis who was successfully treated by a staged treatment, in which bridging thoracic endovascular aortic repair was followed by delayed surgical repair of the esophagus and aorta.

Key words: 1. Fistula
2. Aorta
3. Esophagus
4. Endovascular stent

Case report

A 74-year-old man presented with hematemesis that had lasted for 1 day. He complained of intermittent epigastric pain of 2 weeks' duration. Upon arrival, he had a blood pressure of 133/78 mm Hg, a heart rate of 59 bpm, and a body temperature of 36°C. Laboratory tests showed mild leukocytosis of 14,300/㎕, a hemoglobin level of 11.4 g/dL, a platelet count of 248,000/㎕, and otherwise non-specific blood chemistry. Chest computed tomography (CT) showed a thoracic aortic aneurysm with intramural hematoma compressing the mid-thoracic esophagus (Fig. 1A). Emergent thoracic endovascular aortic repair (TEVAR) was performed to prevent further massive bleeding. A S&G seal thoracic stent graft (diameter 36 mm, length 200 mm; S&G Biotech Inc., Yongin, Korea) was deployed under fluoroscopy. The endovascular procedure was completed uneventfully and intraoperative angiography after deployment showed no evidence of contrast leakage. The patient's post-procedural course was uneventful. After 2 days, esophagoscopy was performed to identify the esophageal lesion, and a 1.5-cm necrotic area of the esophageal wall with blood clots was found at the middle third of the esophagus (31.2 cm from the incisor) (Fig. 2).

After maintaining adequate parenteral nutrition and intravenous antibiotics for 5 days, we planned surgical repair of the lesion. Left thoracotomy was performed through the fifth intercostal space. An area of fibrous adhesion and necrotic tissue was noted between the aortic aneurysm and the esophageal wall (Fig. 3). However, there was no evidence of fulminant mediastinitis, and an extensive hematoma was observed after dividing the fistulous tract. After evacuating the hematoma, the necrotic tissue was debrided thoroughly, which resulted in exposure of the

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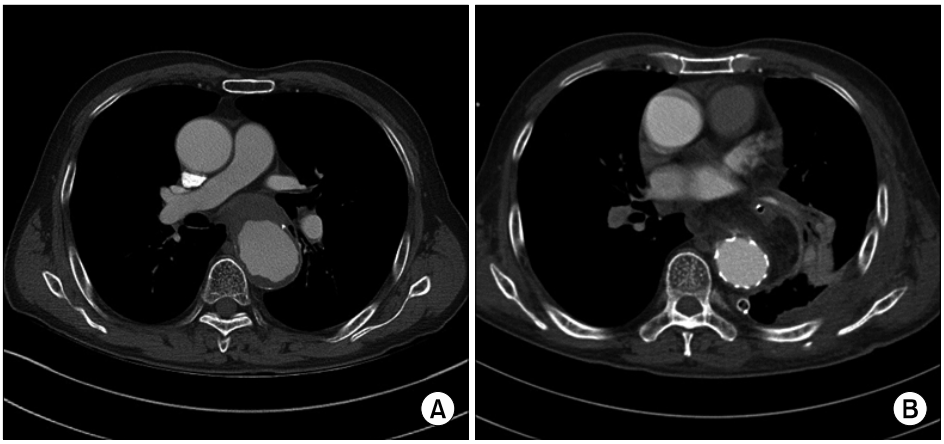

Fig. 1. (A) Preoperative computed tomography shows focal dilatation of the mid-thoracic aorta with an eccentric thrombus at the subcarinal level, compressing the esophagus; the wall and fat between the esophagus and aorta had disappeared. (B) Postoperative computed tomography shows no specific complications.

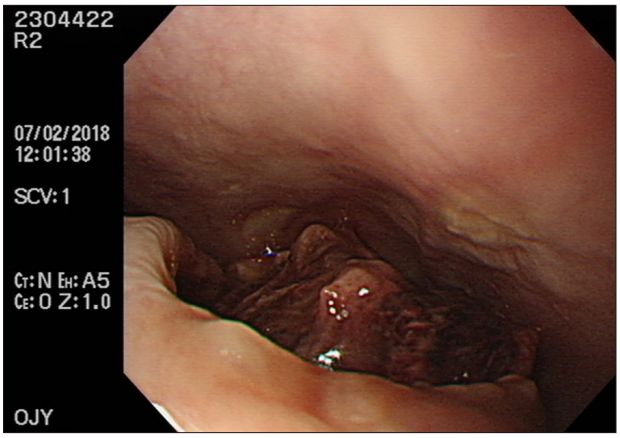

Fig. 2. Endoscopy shows the presence of a necrotic area of the esophageal wall covered with blood clots at the middle third of the esophagus (31.2 cm from the incisor).

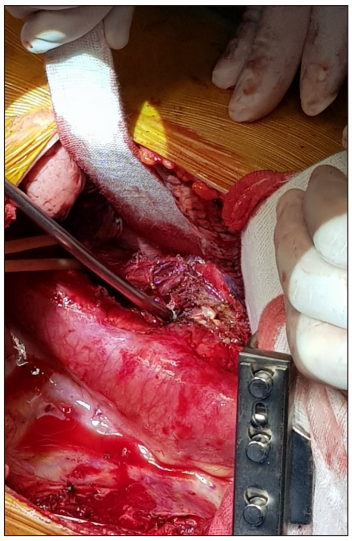

Fig. 3. Intraoperative photography showing a fistulous tract between the aorta and esophagus. There was an area of fibrous adhesion and necrotic tissue, but no evidence of fulminant contamination.

esophageal lumen and bare aortic stent graft. After debridement, a 3-×5-cm opening at the aortic wall and a 3-cm-long longitudinal opening of the esophageal wall developed, but no exsanguination was noted at the aortic opening. For separation and reinforcement, the aortic opening was closed with a bovine pericardium patch (Vascu-Guard; Bio-Vascular Inc., Saint Paul, MN, USA) with a few interrupted sutures of pledgetted 3-0 Prolene. Vancomycin slurry was applied between the patch and stent graft for further sterilization. The esophageal opening was closed primarily in 2 layers after tissue debridement and trimming of the mucosal and muscular layer of the esophagus. The entire esophageal layer was closed with a few interrupted 4-0 polydioxanone sutures and the outer muscular wall with a few interrupted 4-0 black silk sutures.

Through an upper midline abdominal incision, an omental flap based on the right gastroepiploic artery was prepared and passed through a small hole made at the anterior margin of the left diaphragm to the left pleural cavity. The aorta and esophagus were separated by placement of an omental flap in between. The flap was fixed to the esophagus and aorta with a few 4-0 black silk sutures. A feeding jejunostomy tube was placed for optimal nutrition support. After massive irrigation of the mediastinum, a 28F thoracic drain was placed. The chest and abdomen were closed in the usual manner.

The patient was transferred to the intensive care

unit and extubated 1 day after the operation. He was placed on jejunostomy feeding with supplementary parenteral nutrition and intravenous antibiotic therapy with ceftriaxone, metronidazole, and isepamicin for a week. On postoperative day 6, a CT scan (Fig. 1B) showed complete improvement of the aortic lesion and no evidence of infection. The patient started oral intake on postoperative day 7 and showed no fever or elevation of inflammatory markers. Intravenous antibiotic therapy was applied for 1 month after the operation and he was discharged on the 40th day of hospitalization with oral antibiotics.

Written informed consent was obtained from patient.

Discussion

Aortoesophageal fistula (AEF) is a rare and life-threatening disease with a high rate of morbidity and mortality. The most common cause of AEF is erosion of the esophageal wall, followed by rupture of an aortic aneurysm, esophageal cancer, ingestion of foreign bodies, esophageal ulcer, and bronchial cancer [1]. In some cases, secondary AEF can occur as a complication after surgical repair or endovascular repair of an aneurysm [2].

Chiari [3] first reported the clinical triad of mid-thoracic pain, sentinel hemorrhage, and exsanguination after a few hours. Our patient presented with mid-thoracic pain and hematemesis.

Esophagoscopy is useful for the diagnosis of AEF; however, it should be noted that endoscopies can cause severe re-hemorrhage by irritating blood clots. Chest CT also can help in the early diagnosis [4].

The most important goal of treating AEF is to prevent massive bleeding. Therefore, once the diagnosis is confirmed, treatment should be performed without delay. However, early diagnosis can be difficult because AEF is not a common cause of upper GI bleeding [4].

The traditional surgical treatment of AEF was emergent open surgery, including repair of the aortic lesion (replacement with an extra-anatomic bypass or in situ reconstruction using homografts or rifampicin-soaked grafts) and immediate esophageal repair or delayed repair. However, intraoperative and perioperative mortality was high with this method [5].

Marone et al. [5] reported that endovascular stent insertion was a viable option for preventing massive bleeding and thereby helping to stabilize the patient's hemodynamic status. Although there is no definitive indication for TEVAR rather than open repair, it is likely in our opinion that TEVAR can control early massive bleeding more rapidly than open repair, thereby decreasing perioperative mortality. Furthermore, TEVAR can be a good choice for patients who cannot endure open repair, especially elderly patients.

However, TEVAR alone cannot be a definitive treatment, because esophageal wall fibrosis and necrotic tissue debris remain present, increasing the risk of mediastinitis and potentially resulting in contamination of the stent graft. Vallabhajosyula et al. [6] stated that 2-stage treatment (bridging TEVAR to open surgical repair) showed better results. Yamazato et al. [7] reported that although the optimal timing of open surgical repair after TEVAR is not clear, less than 1 week is recommended because contamination of the stent graft may occur if a longer interval is used.

Esophagectomy is better for infection control than primary closure of a perforated esophagus, because primary suturing can be unraveled [7]. In our case, primary closure of the esophagus was performed because we thought that if there was little grossly visible contamination and the rim of the esophageal opening was fibrotic and hard, primary closure could be a good option. For better infection control, a muscular or omental flap is recommended [7].

We suggest that TEVAR is an effective procedure to prevent early exsanguination in patients with AEF. However, it must be followed by open surgical repair or replacement of esophageal and aortic debris to decrease the risk of mediastinitis and stent graft infection, which can result in graft failure. Our patient remained well in the early follow-up period. However, multiple cases with long-term follow-up are required to clarify the outcomes and safety of this procedure. The long-term prognosis of our patient should be investigated because aortic patch closure was done, instead of graft replacement or in situ reconstruction, based on the conviction that there was a low probability of infection in the operative field.

Conflict of interest

No potential conflict of interest relevant to this ar-

ticle was reported.

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