Case Report

Aortoesophageal Fistula: Alternatives of Treatment Case Report and Literature Review

Jorge Flores, MD, Norihiko Shiiya, MD, PhD, Takashi Kunihara, MD, PhD, Kimihiro Yoshimoto, MD, PhD, and Keishu Yasuda, MD, PhD

Aortoesophageal fistula (AEF) remains as a life-threatening condition with a high rate of morbidity and mortality. It is usually related to aortic or esophageal disease, and less commonly foreign body ingestion. In spite of several strategies for treatment, there is little consensus regarding the optimal management of this entity. In this paper, we present our experience in successfully managing one patient with AEF by performing open surgical repair. We also include a discussion on criteria for selecting the most appropriate alternative of treatment: open or endovascular repair, based on a review of the literature currently available in MEDLINE. (Ann Thorac Cardiovasc Surg 2004; 10: 241–6)

Key words: aorta, esophagus, fistula, surgical operation, endovascular stent

Introduction

Aortoesophageal fistula (AEF) is an uncommon condition that presents a problem in therapy because of the high rate of morbidity and mortality associated with its surgical management and the uniformly fatal outcome of medical treatment. Thoracic aortic aneurysms account for about two thirds of all AEFs.[1-3] Management of a patient with this disorder demands prompt diagnosis and repair of the lesion considering three basic problems: 1) an aortic lesion that could lead to death from hemorrhaging, 2) an esophageal lesion that can develop sepsis and result in delayed death if not correctly treated, and 3) the potential involvement of the surrounding tissues caused by infection.

Several types of treatment have been described including open surgery, temporary control measures such as percutaneous embolization, and the use of a Sengstaken-Blakemore tube, and more recently endovascular treatment.[4,5] In this work we present our experience in the successful management of one patient with AEF by open surgery. We also include a discussion of the data currently available in MEDLINE regarding both open surgical repair and endovascular stent grafting for the treatment of AEFs.

From Department of Cardiovascular Surgery, Hokkaido University School of Medicine, Sapporo, Japan

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Address reprint requests to Takashi Kunihara, MD, PhD: Kita-ku, Nishi 5, Sapporo 060-8648, Japan.

Case Report

A 67-year-old female patient underwent orthopedic surgery on her knee in a local hospital after she fractured her left proximal tibia several days before. On the second postoperative day, the patient complained of chest pain and experienced shock for hematemesis. Blood transfusion was performed until her hemodynamic status was stabilized. At the same time, an endoscopy was performed which revealed an esophageal ulcer over a pulsating tumor, discovered 25 cm beyond the dental arch (Fig. 1). A subsequent enhanced computed tomography (CT) showed a ruptured distal aortic arch aneurysm, with thrombus and air bubbles inside, which suggested an AEF (Fig. 2). Clinical analysis reported a white blood cell (WBC) count of 21,600/mm^3 and C-reactive proteins (CRP) at 13.2 mg/dl. The patient was referred to our department for emergency surgery. A femoral vein-femoral artery partial cardiopulmonary bypass was performed at first. The aortic aneurysm was approached through a left thoracotomy at the fifth intercostal space. Pleural effusion and adhesion of the left lung to the aorta were found. Since the origin of the left subclavian artery was involved in the aortic aneurysm, it was clamped, transected, and anastomosed to a Dacron graft for selective perfusion during the aortic

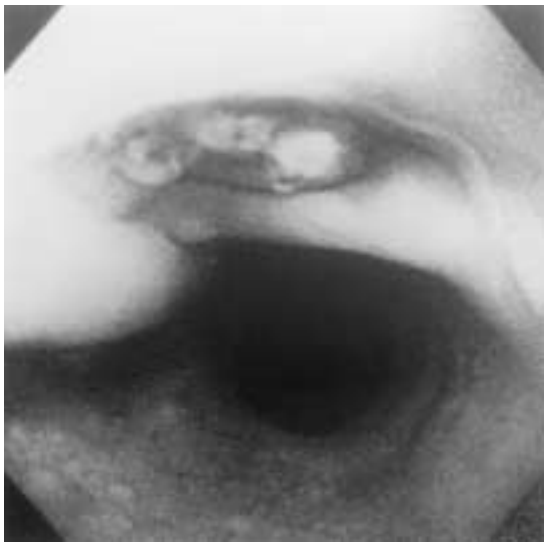

Fig. 1. Endoscopy showing an ulcer that developed over a pulsating tumor on the esophageal wall, discovered 25 cm beyond the dental arch.

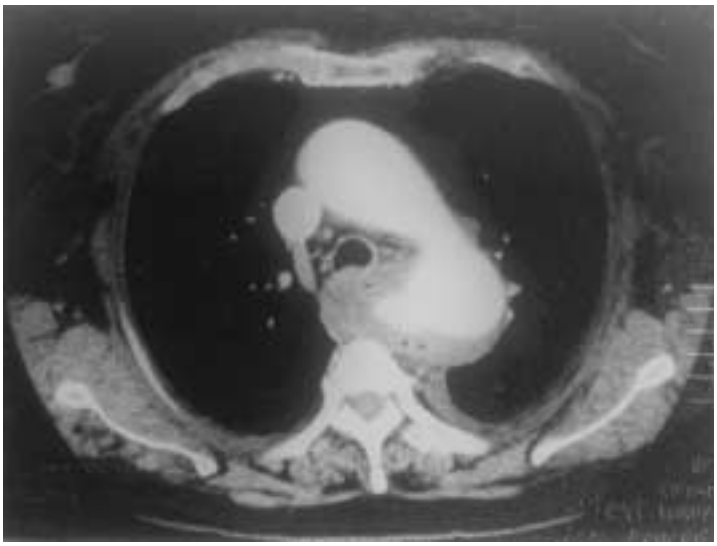

Fig. 2. Enhanced CT showing a ruptured distal aortic arch aneurysm with thrombus and air bubbles inside.

Fig. 3. Picture of a resected esophageal piece showing orifice of the aortoesophageal fistula.

reconstruction. The femoro-femoral cardiopulmonary bypass was established, and the left subclavian artery was also perfused. The aorta was clamped proximal to the left subclavian artery and distal to the origin of the fifth intercostal artery. The aneurysm was opened and an AEF of 5 mm in diameter, covered by thrombus, was noted in the right posterolateral aneurysmal wall. The distal aortic arch, including the origin of the left subclavian artery, and the proximal descending thoracic aorta were replaced by a Dacron graft. The subclavian artery was reattached to the aortic graft through the graft used for its selective perfusion. Subsequently, subtotal esophagectomy (Fig. 3) with cervical esophagostomy was performed. Nonviable tissues were carefully debrided. The abdominal cavity was opened through a medial laparotomy, followed by suture of the distal esophageal stump at the cardiac segment of the stomach, and placement of a gastrostomy tube for enteral feeding. The thoracic cavity was copiously irrigated with saline solution and the aortic graft was wrapped with an omental flap. Thoracic tubular drainage was deployed through the seventh intercostal space. Incisions at the thorax and abdomen were closed in a standard fashion. A pathologic examination revealed atherosclerosis of the aortic wall. On the 10th postoperative day, the patient developed fever in response to a wound infection at the thorax. After debridement of the infected tissues and

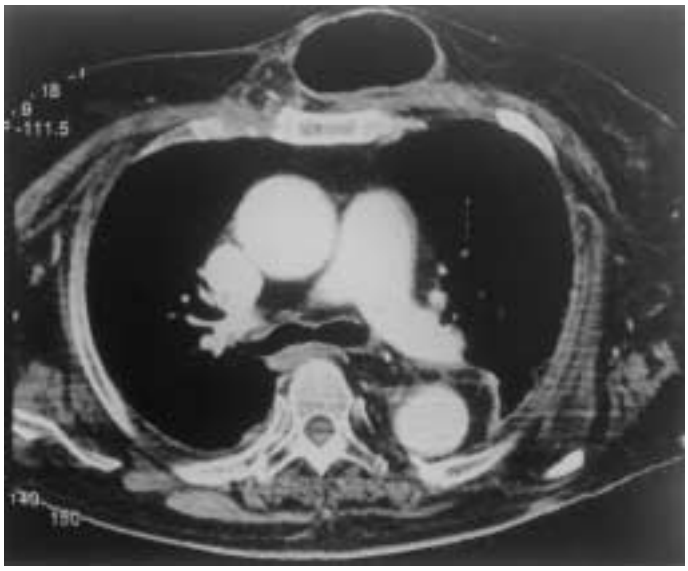

Fig. 4. Enhanced CT showing the pre-sternal reconstruction of the esophageal truck.

intravenous antibiotics, her condition improved, and the patient was eventually discharged. Forty-five days later the patient was readmitted to the hospital for a scheduled reconstruction of the esophagus tract. A gastric tube was pulled up subcutaneously to the cervical esophageal stoma (Fig. 4). Five days later, the patient developed cardiopulmonary arrest due to pulmonary embolization caused by deep venous thrombosis. She recovered after treatment with heparin and concomitant percutaneous cardiopulmonary support, which was discontinued three days later. The subsequent postoperative course was uneventful and the patient was discharged in good condition.

Discussion

AEF which usually arises from aortic or esophageal disease, is associated with a high morbidity and a high mortality. Despite several strategies for surgical treatment, there is little consensus about the optimal management of this disease. Classically, dysphagia or chest pain occurs because of compression of the esophagus or the vagus nerve, and these precedes to episodes of hematemesis, as was experienced by our patient. The earliest occurrence of hematemesis is recognized as sentinel hemorrhage. Then, the patient becomes stable until the next hemorrhage, usually a fatal exsanguination. There is an unpredictable symptom-free interval between the herald hemorrhage and subsequent exsanguinations.[3,6,7)] Chest radiographs should reveal an enlargement of the mediastinum. Endoscopic examination may reveal a mass in the esophageal wall, as in our patient (Fig. 1), which is covered by adherent blood clots or with active hemorrhaging. The diagnosis can be confirmed by CT, or by digital subtraction angiography (DSA) when there is active bleeding (≥0.5 ml/min).[3,8)]

On account of the high probability of death from exsanguination or from infection of the surrounding tissues and subsequent sepsis, an aggressive treatment without delay is the only form of effective therapy. At present, two main types of surgical management have been described: open surgery[7,9)] and endovascular repair.[1,10)]

Since most surgical corrections of AEFs are performed in emergencies, no specific maneuvers for avoiding neurologic damages are applied. It is advisable that those patients with clinical signs of infection and wide contamination of the surrounding tissues demonstrated by imagenologic studies, as air bubbles or pleural effusion, or patients with AEF caused by ingestion of a foreign body,[11)] should undergo open surgery immediately. Procedures should include debridement of any devitalized tissue, including the compromised segment of the esophagus, and replacement of the diseased aortic segment with in situ prosthesis,[7,12,13)] cryopreserved aortic homografts,[14)] or extra-anatomical bypass grafting.[15,16)] da Silva et al.,[8)] in a review of 11 successfully repaired AEFs caused by atherosclerotic aortic aneurysms that ruptured into the esophagus, reported that three patients underwent in situ aortic graft replacement and esophagectomy, two with primary and one with delayed esophago-gastroanastomosis. None of the patients developed postoperative complications. However in the other eight patients, in situ aortic graft replacements were performed in seven pa-

Table 1.

Authors/date	Sex	Age	Precedent for AEF	Contraindication for OS
Oliva et al. 1997[10]	M	40	Esophago-gastric partial resection and reanastomosis for esophagus Ca	Poor physical condition after two thoracotomies due to persistant AEF, Hemorrhagic complications
Kato et al. 2000[18]	M	59	Irradiation therapy due to esophagus Ca	Esophagus Ca with high probability of aortic wall invasion
Burks et al. 2001[19]	M	76	Graft replacement of a TAAA	CAD, COPD, CRI
Bond et al. 2001[20]	F	58	Graft replacement of a transected TA	CAD, hemorrhagic complications
van Doorn et al. 2002[21]	F	66	Mycotic TAA	Respiratory failure, GI hemorrhage
Leobon et al. 2002[1]	M	80	TAA	Advanced age, recurrent CVAs, treatment with antivitamin K
D'Ancona et al. 2002[22]	F	78	TA pseudoaneurysm	Severe COPD
Haulon et al. 2002[23]	NA	NA	Esophageal surgery	Poor physical condition due to two recent thoracotomies, unstable hemodynamic status
Nishibe et al.[unpublished]	F	71	TAA	Advanced age, recurrent GI hemorrhage

M, male; F, female; AEF, aortoesophageal fistula; OS, open surgery; ATB, antibiotic; PO, postoperative; Ca, cancer; TA, thoracic aorta; TAA, thoracic aortic aneurysm; TAAA, thoracoabdominal aortic aneurysm; CAD, coronary artery disease; COPD, chronic obstructive pulmonary disease; CRI, chronic renal insufficiency; CVA, cerebrovascular accident; GI, gastrointestinal; NA, no data; IV, intravenous; MOSF, multiple organ system failure.

tients and an extra-anatomic bypass was performed on one patient. The esophagus was repaired by primary suture in seven patients, except in one patient in whom the esophagic lesion was not found. Among them, six patients developed recurrences of AEFs or disruption of the esophageal closure. Three died. Since the omentum promotes the remission of infected processes, and prevents reinfections because of its widespread vascular and lymphatic supply and its ability to fill dead spaces,[3,17] we used an omental pedicle graft surrounding the aortic graft and the anastomosis in our patient. This helps prevent the accumulation of fluids and clots in the perigraft space and reduces the chance of reinfection.

Although it can be assumed that contamination of the surrounding tissues by esophageal flora occurs in AEF, several patients show no clinical or imagenologic evidence of infection. Those patients at high risk for open surgical repair and without evidence of infection should be considered for endovascular treatment. Our review of the literature in MEDLINE and one unpublished case reported in our own department (in submission) revealed that to date, nine patients with the diagnosis of AEF have been treated by endovascular surgery (Table 1). Two patients had clinical evidence of infection at the time of the endovascular procedure. One of them died from persistent sepsis. Of the remaining seven patients who showed no evidence of infection, two patients died, one from mediastinitis related to complications of the stent graft deployment, and the other patient who had received radiation treatment for esophageal carcinoma and died from pneumonia due to a tracheoesophageal fistula. The six survivors were doing well at follow-up examinations that ranged from 6 to 36 months. The fact that three of the four patients with postoperative complications died in this group may be related to their poor clinical condition and life expectancy.

Patients with AEFs and clinical signs of infection who are in critical physical condition that makes them at high risk for open surgery should be considered for endovascular surgery as a palliative treatment, or a temporary alternative until they are healthy enough to tolerate open surgery. There have been cases reported of the successful use of endografts as a provisional treatment in patients with aortoduodenal and ilioenteric fistulas that allowed improvement of their general condition until definitive treatment by open surgery was possible.[24,25] Ag-

Sepsis	Anesthesia	PO complications	ATB therapy	Follow-up
No	General	None	3 weeks	13 months
No	General	Tracheo-esophageal fistula	Lifetime regimen	Died of pneumonia after 5 months
Yes	Epidural	Persistent sepsis	IV until dead	Died of MOSF after 26 days
No	NA	None	6 weeks	33 months
Yes	General	Tracheobronchial edema, aneurysm sac hygroma	30 weeks	24 months
No	NA	Endoleak, reopening of the AEF after new stent graft deployment	NA	Died of mediastinitis after 25 months
No	NA	None	1 month	6 months
No	General	None	NA	NA
No	Combined general and spinal	None	NA	36 months

gressive antibiotic therapy is also recommended as an adjunct to control the infectious process in these procedures.

According to our review of the literature in MEDLINE and our own experience, we classified patients diagnosed with AEF into three groups. Group 1: patients who are in relatively good physical condition with clinical signs of infection, or patients who ingested a foreign body, are good candidates for open surgical repair; group 2: patients who are at high risk for open surgical repair and show no clinical signs of infection should undergo endoluminal stent-graft deployment; group 3: patients who are at high risk for open surgery because of poor physical condition, and show clinical signs of infection should undergo endovascular repair as a palliative therapy or temporary treatment until their general condition improves for open surgery.

In conclusion, we assume that a careful selection for open or endovascular repair of an AEF, in accordance with the physical condition of the patients, and the laboratorial and radiological findings will improve the rates of morbidity and mortality for this entity. However, AEFs are uncommon, with only a small number of cases reported in the literature, and there is limited follow-up for any option of treatment. Further evaluations, including a randomized study of patients, are necessary for a better assessment of proper AEF management.

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