Surgical strategy for the treatment of aorto-esophageal fistula

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ABSTRACT

Objective: To present a surgical strategy for aorto-esophageal fistula (AEF).

Methods: From October 1999 to May 2017, 27 patients with AEF were treated at Kobe University Hospital. After 9 patients with malignancies or fish bone penetration were excluded, 18 patients who had AEF secondary to aortic lesions were investigated. The mean age was 67.2 ± 10.4 years, and the male/female ratio was 16:2. Twelve patients had a nondissection thoracic aneurysm, and 6 patients had a chronic aortic dissection. Six patients were in shock. Seven patients had a previous thoracic endovascular aortic repair (TEVAR) in the descending aorta, 2 patients had descending aorta replacement, 1 had hemiarch replacement, and 2 had total arch replacement. As the first treatment for AEF, 3 patients underwent TEVAR as destination therapy, 3 patients had a bridge TEVAR to open surgery, 1 patient had an extra-anatomical bypass from the ascending aorta to the abdominal aorta, and 11 patients had an in situ reconstruction of the descending aorta. The esophagus was resected in 16 patients, and an omental flap was installed in 16 patients. Additional procedures were extra-anatomical bypass in 2 patients and in situ reconstruction of the aorta in 3 patients.

Results: Hospital mortality was noted in 4 patients (22.2% persistent sepsis, $n = 3$: pneumonia, $n = 1$). However, since 2007, only 1 of 13 patients has died (pneumonia). Late death occurred in 5 patients, due to pneumonia, cerebral bleeding, diarrhea, sudden death, and persistent infection. Actuarial survival was $42.4 \pm 12.8\%$ at 5 years and freedom from aorta-related death was $59.4 \pm 13.5\%$ at 5 years. Nine patients achieved completed reconstruction of the esophagus 172 ± 57 days after initial surgery.

Conclusions: Although a comparative study was not performed, 1-stage surgery consisting of resection of an aneurysm and esophagus, in situ reconstruction of the descending aorta, and omental flap installation provided a better outcome in the treatment for AEF. Bridging TEVAR to the open surgery is a useful adjunct in patients with AEF with hemorrhagic shock. Later reconstruction of the esophagus can be performed in the survivors. (*J Thorac Cardiovasc Surg* 2018;155:32-40)

Esophagoscopy demonstrated an aorto-esophageal fistula covered by thrombi.

Central Message

Aggressive strategies for aorto-esophageal fistula and a hybrid approach, including bridge thoracic endovascular aortic repair to open surgery, can provide better patient outcomes.

Perspective

A 1-stage surgery consisting of the resection of the aneurysm and esophagus, in situ reconstruction of the descending aorta, and omental flap installation provided a better outcome in the treatment for aorto-esophageal fistula. Later reconstruction of the esophagus can be performed in the survivors.

See Editorial Commentary page 41.

A fistula between the thoracic aorta and the esophagus, or an aorto-esophageal fistula (AEF), is a rare and highly fatal condition. AEFs usually arise in patients with thoracic

aortic diseases, such as aneurysms, or after previous thoracic aortic surgery.¹ Surgical management also carries a high risk of morbidity and mortality. Although several alternative strategies have been reported in the literature, including extra-anatomical bypass (EAB) and in situ repair,

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Received for publication May 31, 2017; revisions received Aug 24, 2017; accepted for publication Sept 9, 2017; available ahead of print Nov 9, 2017.

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0022-5223/\$36.00

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<https://doi.org/10.1016/j.jtcvs.2017.09.047>

Scanning this QR code will take you to a supplemental video for the article.

Abbreviations and Acronyms

ABF	= aortobronchial fistula
AEF	= aorto-esophageal fistula
CPB	= cardiopulmonary bypass
EAB	= extra-anatomical bypass
TEVAR	= thoracic endovascular aortic repair

there is a lack of consensus concerning the optimal treatment for AEF.

However, relatively little is known regarding the results of surgical management of AEF because of the rarity of the condition and the lack of large surveys. Recently, thoracic endovascular aortic repair (TEVAR) has shown to be successful in excluding thoracic aortic aneurysms and other thoracic aortic pathologies.² With the growing number of interventional thoracic aortic procedures and increases in the length of follow-up periods, late complications of TEVAR increasingly have become evident over time, including postprocedural AEF.^{3,4} Conservative treatment results in no late survival even if conventional surgical repair after endovascular stent-graft insertion or as a primary intervention is attempted. Since 2007, we have adopted an aggressive management strategy for AEF. This report reviews our experiences and discusses our surgical strategy for AEF.

PATIENTS AND METHODS

This study was approved by the institutional review board of Kobe University Hospital, and the need for individual consent was waived. From October 1999 to May 2017, 27 patients with AEF were treated at Kobe University Hospital. After we excluded 6 patients with esophageal cancer, 1 with lung cancer, 1 with bladder cancer, and 1 with a fish bone penetration, clinical data of 18 patients who had AEF secondary to aortic lesions were analyzed. The age was 67.2 ± 10.4 years, and the male/female ratio was 16:2. Twelve patients had a nondissection thoracic aneurysm, and 6 patients had a chronic aortic dissection. Seven patients had previous TEVAR in the descending aorta, 2 patients had descending aorta replacement, 2 had total arch replacement, and 1 had hemiarch replacement. The interval from the TEVAR to the treatment of the AEF was 50.7 ± 48.3 months, ranging from 1 month to 11 years and 8 months.

The diagnosis initially was made by the noting of a history of hematemesis or melena and by the detection of underlying thoracic aortic disease. Every patient underwent contrast-enhanced computed tomography of the chest and esophagoscopy. On computed tomography, a new mediastinal mass was present in all patients, suggesting contained rupture. However, the leading radiographic sign was air surrounding the prosthesis. The diagnosis was confirmed by additional endoscopy in the majority of patients. Lesions were located between 20 and 30 cm from the superior dental arch. Sometimes, there was a pulsatile mass with adherent blood clots in the wall of the esophagus or a simple mucosal fistula or indentation. Six patients had some episodes of hemorrhagic shock. Comorbidities were end-stage kidney disease requiring hemodialysis in 2 patients, chronic obstructive pulmonary disease in 1 patient, and suppurative spondylitis in 1 patient. The diameter of the native aneurysms was quite large, 65.3 ± 21.5 mm, ranging from 55 to 78 mm.

At the first treatment for AEF (Table 1), 3 patients underwent TEVAR as the destination therapy, 3 patients had a bridge TEVAR because of hemodynamic instability, 1 patient had EAB from the ascending aorta to

the abdominal aorta with an 18-mm Dacron graft (Gelweave; Vascutek, Glasgow, Scotland), and 11 patients had in situ reconstruction of the descending aorta. The additional second procedure, such as EAB or in situ replacement of the descending aorta, after destination TEVAR was required in 2 patients. In 3 patients who underwent a bridge TEVAR, staged replacement of the descending aorta was performed in 2 patients and EAB in 1 patient at 7.3 ± 2.5 days after TEVAR. Esophagectomy was done in 14 patients simultaneously with aortic procedures, and 2 patients required a staged esophagectomy from the right thoracotomy (Table 2). An omental flap was installed in 16 patients (Table 3).

Surgery

Our basic surgical strategy for AEF since 2007 is as follows. Simultaneous resection of the aorta and esophagus, in situ reconstruction of the descending aorta with a rifampicin (Rifujin; Daiichi Sankyo, Tokyo, Japan)-soaked Dacron graft, and omental flap installation are performed. The patient is positioned in a right semirecumbent position and a posterolateral skin incision is made from the anterior axillary line to the subscapular area. The left pleural cavity is entered through the fourth or fifth intercostal space and the chest is opened widely with the rib-cross technique. After heparinization, cannulation of the common femoral vessels is achieved by the Seldinger method. Partial bypass is used when clamping of the aorta at the aortic arch is feasible. When an aortic arch replacement is required, total cardiopulmonary bypass under moderate hypothermia is adopted at the tympanic temperature of 28°C .

Aortic control is achieved proximally at the arch and the lower descending aorta. The aorta is incised and back bleedings from the patent intercostal arteries are controlled by transfixing sutures. The fistula is identified, and the aneurysm sac is resected as much as possible. The surrounding necrotic tissue is debrided extensively. Esophagectomy is performed by gastrointestinal surgeons while maintaining lower body perfusion. Both stumps of the esophagus are closed temporarily. The pleural cavity is extensively debrided and copiously irrigated with several liters of 0.2% gentian violet (methylosaniline chloride) solution (Figure 1).

In situ reconstruction of the descending aorta is achieved with a rifampicin-soaked, gelatin-impregnated Dacron graft (J graft [Japan Life Line, Tokyo, Japan] or Gelweave graft [Vascutek]). The aortic allograft was used in only one patient in this series because of limited availability.

TABLE 1. Reconstruction of the aorta

Reconstruction	Methods	Secondary procedure	n	HD	LD
First in situ	Total arch + Desc Ao		1		
	Total arch		3	1	1
	Desc Ao		6		2
	TAAA		1		1
First EAB	Asc Ao–Abd Ao bypass		1	1	
First TEVAR		TAAA	1	1	
		EAB	1	1	
		None			1
Bridge TEVAR		Desc Ao	2		
		EAB	1		
Total			18	4	5

HD, Hospital death; LD, late death; Desc Ao, descending aorta; TAAA, thoracoabdominal aortic aneurysm; EAB, extra-anatomical bypass; Asc Ao, ascending aorta; Abd Ao, abdominal aorta; TEVAR, thoracic endovascular repair.

TABLE 2. Esophageal resection

Esophageal resection	n	Median sternotomy	Thoracotomy	Aortic infection–related death	Survival
Simultaneous	14	3	Left, 11	1	9
Staged	2		Right, 2	2	0
None	1			1	0

A simple continuous technique with a 4-0 or 3-0 monofilament suture with a Teflon felt reinforcement is used for anastomosis. The intercostal arteries are never attached to the graft.

After termination of the cardiopulmonary bypass and protamine reversal, the full length of the omental flap is taken and brought through small holes of the diaphragm to the left pleural cavity. The graft is wrapped completely by the omental flap. The chest is closed in the usual manner. The proximal stump of the esophagus is brought up to the left neck and an esophagostomy is made. A gastrostomy and jejunostomy also are made (Figure 2 and Video 1).

In 3 patients in this series with an arch aneurysm with severe chronic obstructive pulmonary disease or after right pneumonectomy, a midsternotomy was chosen. They underwent total arch replacement, esophageal resection, and omentopexy. During the esophagectomy, lower body perfusion was maintained from the femoral arterial perfusion with aortic occlusion balloon in the descending aorta as well as antegrade cerebral perfusion (Figure 3).

Twelve patients received partial cardiopulmonary bypass (CPB), and 6 patients required total CPB with cardiac arrest. Antegrade cerebral perfusion was used in 5 patients for brain protection during aortic arch reconstruction. The CPB time was 167 ± 54 (61-287) minutes, duration of myocardial ischemia was 117 ± 23 (23-206) minutes, and cerebral perfusion time was 80.8 ± 72.6 (18-165) minutes. The lowest tympanic temperature was 27.7 ± 7.0 (18.5-36.0)°C, and the lowest rectal temperature was 32.8 ± 3.8 (26.2-36.0)°C.

RESULTS

There was no mortality at 30 days, and 4 in-hospital deaths were reported. Two patients died of continued infection, 1 died of multiple-organ failure, and 1 died of pneumonia. The first patient had a resection of the aorta and EAB from the ascending aorta to the abdominal aorta. The esophagus was not resected. The second patient underwent esophageal and aortic resection, in situ replacement of the thoracoabdominal aorta (Crawford extension I) via an allograft, and omentopexy. However, detachment of the anastomosis occurred. An expanded polytetrafluoroethylene graft replacement was performed 2 weeks after allograft replacement; however, it was not successful. The third patient had esophagectomy through the right thoracotomy next day of the TEVAR for AEF; however, the infection was not well controlled, and removal of the TEVAR, aortic resection, and EAB from the

ascending aorta to the abdominal aorta was performed 2 weeks afterward. Unfortunately, the EAB graft was infected, and a right axillobifemoral bypass (J graft; Japan Life Line) was performed. Four months later, he died of rupture of the aortic stump. The last patient was 85-year-old woman having hemodialysis. She had total arch replacement and esophagectomy through the midsternal incision, but she died of pneumonia 8 months after surgery. All patients except 1 had surgery before the year 2007. Complications in these 18 patients consisted of respiratory failure, in which tracheostomy was required in 3, acute renal failure requiring dialysis in 4, and multiple organ failure in 3.

Follow-up was completed in 100% and duration was 685 ± 644 (23-2384) days. We continued a lifetime oral antibiotics regimen in these patients. Late deaths occurred in 5 patients. One patient who had undergone thoracoabdominal aortic aneurysm grafting died of hematemesis 2 years after surgery. The second patient died of sepsis 1 year after. One died of intracranial bleeding 2 years after, and 1 died of persistent diarrhea 1 year after surgery. One patient refused further surgery after TEVAR and died of hematemesis 1.5 years after TEVAR. There were no long-term survivors among the 5 patients who underwent the destination TEVAR or EAB. On the contrary, all 3 patients who underwent the bridge TEVAR had succeeding definitive treatments and survived. There was only 1 AEF-related late death among the 10 patients who have been managed since 2007. Kaplan-Meier analysis revealed that survival at 5 years was $42.4 \pm 12.8\%$ (Figure 4) and that freedom from aortic infection–related death was $59.4 \pm 13.5\%$ (Figure 5).

Nine patients had reconstruction of the esophagus, with a pedicled jejunum in 1, a pedicled jejunum with internal thoracic artery and vein anastomosis to the jejunal vessels (the supercharge anastomosis) in 7, and an ilio-cecum graft with a supercharging anastomosis in 1. The duration from esophageal resection to reconstruction was 172 ± 57 (79-248). One patient had a dehiscence of the esophagojejunostomy (Table 4).

TABLE 3. Omentopexy

Aorta	n	Aortic infection–related death
In situ reconstruction	13	2
EAB	2	1
No omentopexy, EAB	1	1

EAB, Extra-anatomical bypass.

DISCUSSION

The AEF is a relatively rare and usually life-threatening condition. Described for the first time by Dubrueil in 1818, primary AEF has been known to be very rare but a lethal clinical entity.⁵ Chiari reported the classic, clinical triad of mid-thoracic chest pain,

FIGURE 1. Operative illustration of a patient who underwent simultaneous resection of the esophagus and the aortic arch to the descending aorta and in situ reconstruction of the aorta wrapped by the omental flap through the left thoracotomy. AEF, Aorto-esophageal fistula; AD (B), type B aortic dissection; BCA, brachiocephalic artery; L CCA, left common carotid artery; L SCA, left subclavian artery; Desc Ao, descending aorta; Asc Ao, ascending aorta.

sentinel arterial hemorrhage, and fatal hemorrhage as “aorto-esophageal syndrome,” which is also known as AEF.⁶ The first survival after surgical repair was reported by Snyder and Crawford in 1983, but very few survivors were documented.⁷ There are not many large series available in the literature, and only reports of several cases or collective reviews have been published.^{4,8}

The incidence of AEF was reported to be 5% to 23% of critical gastrointestinal bleeding by meta-analysis.⁹ Secondary AEF after open thoracic aortic surgery occurs with an increased incidence of up to 1.7% of patients. Because TEVAR is now used routinely in aortic emergencies or in elderly, high-risk patients, it seems that there has been an increased number of AEF cases after TEVAR. Chiesa and colleagues³ reported that 19 of

FIGURE 2. Operative illustrations of bridge TEVAR. AEF, Aortoesophageal fistula; TEVAR, thoracic endovascular aortic repair.

VIDEO 1. The patient was a 73-year-old man who had undergone a replacement of the descending aorta for chronic type B aortic dissection 2 years previously. He had a fever and hematemesis. Preoperative computed tomography scan showed free air around the graft, and esophagoscopy showed a fistula in the middle portion of the esophagus. A left posterolateral incision was made, and a partial cardiopulmonary bypass was used. The aortic arch was clamped between the left common carotid artery and left subclavian artery. The distal aorta also was clamped, and the infected graft was removed. The aortic wall was resected as much as possible, and esophagectomy was performed by the gastrointestinal surgeons. Lavage with copious saline was done, and the aorta was reconstructed with a rifampicin-soaked, gelatin-impregnated 22-mm Dacron graft. After the patient was weaned off the cardiopulmonary bypass, a mid-abdominal incision was made. A pedicled omentum was harvested, was brought to the left thorax through the diaphragm, and the graft was wrapped. A cervical esophagostomy and gastrostomy were made. Video available at: [http://www.jtcvsonline.org/article/S0022-5223\(17\)31964-5/fulltext](http://www.jtcvsonline.org/article/S0022-5223(17)31964-5/fulltext).

1113 (1.7%) patients who had TEVAR developed AEF/aortobronchial fistula (ABF) in 39 Italian centers between 1998 and 2008. Eleven patients underwent AEF/ABF surgical treatment, with a perioperative mortality of 64% (7/11). At a mean follow-up of 17.7 ± 12.5 months, overall survival was 16% (3/19). Canaud and colleagues¹⁰ most recently reported on their results after secondary open surgery in a heterogeneous post-TEVAR cohort of 14 of 236 patients and reported an extraordinary low hospital mortality of 14.3% with a 2-year survival rate of 87.7%. However, their surgical series of 14 patients included only 7 patients (50%) with AEF (n = 1) and ABF (n = 6).

The main causes of the AEF are ruptured or nonruptured aortic aneurysm, foreign body ingestion, and advanced esophageal malignancy.¹ In addition, some cases of AEFs occur as a complication after surgical prosthetic repair of an aneurysm or TEVAR.¹¹ The exact mechanism of AEF remains unknown, and many pathophysiologies have been speculated. Investigators suggested that the pathogenesis is related to esophageal ischemia secondary to blockade of the esophageal artery, increased pressure in the posterior mediastinum, inflammation of the resorption hematoma, mechanical compression by a large aneurysm and secondary erosion, or the geometric change in the aortic arch and the descending aorta after TEVAR.⁴ The radial force of the graft against the native aortic wall might be a causative factor. Primary infection or inflammation of the

FIGURE 3. Operative illustrations of simultaneous resection of the esophagus and the aortic arch and in situ reconstruction of the aorta and omentopexy through mid-sternotomy. *RFP*, Rifampicin; *SCP*, antegrade selective cerebral perfusion; *Desc Ao*, descending aorta; *RT*, rectal temperature.

aneurysmal wall is also a potential cause of AEF.⁴ Recently, we adopted preoperative endoluminal echo esophagography to assess the thickness of the esophageal wall. Sometimes, we find a very thin esophageal wall compressed by an aneurysm.

Conservative medical treatment results in no late survival.^{1,4} Control or prevention of fatal bleeding is mandatory, and definitive aortic treatment should be accompanied by methods to stop continuous contamination through the fistula to the aortic prosthesis. Primary closure of the perforated esophagus has been tried and resulted in a certain amount of success; however, radical esophagectomy is more effective to control contamination because there is a significant morbidity associated with a breakdown of primary suturing of the esophageal defect. In a recent multicenter report analyzing patients with AEF after TEVAR, Czerny and colleagues⁴ demonstrated that there were no 1-year survivors without esophagectomy. We prefer simultaneous resection of the esophagus during aortic reconstruction under CPB. The best results were obtained with (1) esophagectomy, (2) debridement and removal of the devitalized tissues followed by abundant irrigation of the mediastinum, (3) in situ restoration of the aorta with a synthetic prosthesis, and (4) protection of the prosthesis with a viable pedicle flap of the greater omentum, pleura, or adventitia.¹²⁻¹⁴ Akashi and colleagues⁸ collected 47 cases of AEF from 14 Japanese centers and

found that mortality rate at 6 and 18 months after diagnosis was significantly correlated with graft replacement ($P = .0188$) and esophagectomy ($P = .0257$), respectively. They concluded that use of thoracic endovascular aortic repair alone for AEF should not be considered a definitive surgery.

In contrast, esophagectomy, open surgery with aortic replacement using prostheses and homograft, and greater omentum wrapping significantly improve the mid-term survival of AEF. Luehr and colleagues¹¹ underscored the importance of a radical approach for AEF. They reported 8 cases of AEF and 2 cases with aortobronchial fistula after TEVAR, and 8 patients were treated surgically with a hospital death of 25%. We previously reported an improved outcome when the treatment strategies for AEF included resection of an aneurysm and esophagus, in situ reconstructions of the descending aorta, and omental flap installation.¹⁵ Since then, 8 cases have been added, and all patients have been discharged from the hospital after this strategy.

Since it was first described in 1994, increasing reports have focused on the role of endovascular management to stabilize patients' conditions.¹⁶ Those reports have confirmed that endovascular insertion of a stent graft plays a pivotal role in the cessation of hematemesis and in the stabilization of hemodynamic status. The open repair therapies had relatively better outcomes than TEVAR alone

FIGURE 4. Actuarial survival. Dotted line indicates 95% confidence limit.

in terms of early mortality, but TEVAR followed by early open repair (bridging) resulted in better outcomes.¹⁷ Kubota and colleagues¹⁸ reported a successful case of a patient who underwent bridging TEVAR, concomitant esophageal resection and conventional aortic graft replacement 2 days later, and simultaneous gastric tube reconstruction performed with an intact whole omental flap covering the aortic prosthesis. In contrast, 3 patients with TEVAR alone did not survive more than 1 year. In addition, our 3 patients who underwent destination TEVAR did not survive. As for the time interval from TEVAR to open repair, we think that optimal timing is less than 1 week; otherwise, the stent graft will have succumbed too deeply to the infection process.

As for aortic reconstruction, orthotopic in situ aortic reconstruction should be chosen.^{12,15} The most common surgical approach for AEF repair involves a left posterolateral thoracotomy with the use of CPB. For lesions involving the distal thoracic aorta, partial CPB is an appropriate option; however, cardiac arrest or brain perfusion is sometimes necessary for extensive debridement. An alternative to reconstruction with a prosthesis in situ could be AEB

grafting (ascending aorta to abdominal aorta or axillary to bilateral femoral artery). However, these procedures carry significant morbidity, including late rupture of the aortic stump, and risks for inadequate perfusion for the lower body.¹⁹

There is no consensus regarding the optimal aortic substitute, and surgical outcomes associated with these respective grafts vary in the literature.²⁰ The availability of allograft might be limited and usually, the allograft tends to be too short and of a small diameter to allow aortic replacement in cases of AEF.²¹ Therefore, we advocate the use of a rifampicin-soaked, gelatin-impregnated Dacron graft²² as an aortic substitute in the presence of infection because extensive debridement of the infection and sterilization is critical to improving outcomes.

There have been not many reports regarding reconstruction of the esophagus after AEF surgery. Optimal timing for esophageal reconstruction is unknown, but in fact, at least several months were required for our patients to recover from the initial surgical insults. In the present series, 8 patients have undergone esophageal reconstruction, with a pedicled jejunum graft in 7 and ileocecum in 1 patient 3 to 8 months

FIGURE 5. Freedom from aorta-related death. Dotted line indicates 95% confidence limit.

after the initial surgery. Kawamoto and colleagues²³ reported that 5 of 10 patients who underwent radical treatment for AEF successfully had esophageal reconstruction after 3 to 6 months. As for the surgical procedure, we exclusively used an antesternal route and a pedicled jejunum graft with a “supercharge” technique (anastomosing the internal thoracic artery or vein to the pedicled jejunal artery or vein, respectively).

TABLE 4. Reconstruction of esophagus

Conduit	n	Interval from AEF surgery
Pedicled jejunum	1	172 ± 57 (59-248) days
Pedicled jejunum with supercharging anastomosis	7	
Pedicled ilio-cecum with supercharging anastomosis	1	

AEF, Aortoesophageal fistula.

Limitations

This study consisted of a small number of cases within a single institution, which can be considered a limitation.

CONCLUSIONS

TEVAR as an initial bridging intervention to stabilize hemodynamics before subsequent open surgery is useful in the treatment of AEF. Although a comparative study was not performed, an aggressive approach, including concomitant esophageal, aorta, and stent graft resection; aortic reconstruction; and omentopexy may provide a better outcome. Later reconstruction of the esophagus can be performed in the survivors.

Conflicts of Interest Disclosure

Authors have nothing to disclose with regard to commercial support.

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Key Words: aortoesophageal fistula, TEVAR, infected aneurysm