In situ aortic allograft insertion to repair a primary aortoesophageal fistula due to thoracic aortic aneurysm

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Aortoesophageal fistula due to thoracic aortic aneurysm is an uncommon cause of gastrointestinal bleeding and has an extremely poor prognosis. In the English literature, we found only 27 successfully managed cases of primary aortoesophageal fistula due to thoracic aortic aneurysm. We present a case of 74-year-old man who experienced the erosion of a thoracoabdominal aortic aneurysm into the esophagus. We successfully performed resection and replacement of the thoracoabdominal aorta with a cryopreserved allograft and total thoracic esophagectomy. A few months later, the esophagus was reconstructed with orthotopic colonic interposition. The patient recovered well and resumed a normal life (12 months' follow-up). (J Vasc Surg 2005;42:1213–7.)

Aortoesophageal fistula (AOF) is a rare (0.01%-0.08%) but life-threatening cause of upper gastrointestinal hemorrhage. According to a comprehensive review by Hollander and Quick[1] (500 cases of AOF gathered from the literature), the most common cause is erosion of a thoracic aortic aneurysm into the esophagus (54%). Other causes are foreign body ingestion (19%; eg, fish bone impaction),[2] advanced esophageal carcinoma (17%),[3] endoscopic esophageal procedures (esophageal stent), trauma, radiotherapy, and tuberculous aortitis.[4] Secondary AOFs after operative procedures are rare (5%), with half of these secondary AOFs occurring after aortic surgery and half occurring after esophageal surgery.[1] Recently, two cases have been reported after endoluminal stenting.[5,6]

Extensive literature review revealed 27 cases of successfully managed primary AOFs secondary to thoracic aortic aneurysms (Table I).[7-32] Early diagnosis and surgical intervention are mandatory for survival. The classical Chiari triad of aortoesophageal syndrome[33] consists of midthoracic pain or dysphagia and a sentinel episode of minor hematemesis followed by fatal exsanguination after a symptom-free period. The symptom-free interval ranges from hours to several days, which leaves the opportunity for early diagnosis and definitive surgical intervention. The surgical intervention should ideally repair both the esophageal erosion and the aortic aneurysm to avoid terminal exsanguination and uncontrollable mediastinitis.[34] We report a successful surgical repair of a primary AOF induced by a thoracoabdominal aortic aneurysm, with special emphasis on the value of in situ aortic replacement with a cryopreserved allograft.

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CASE REPORT

A 74-year-old man presented with dysphagia and a minor episode of hematemesis of bright red blood. He also complained of hoarseness and severe loss of weight. At admission in the outpatient clinic of gastroenterology, the patient was stable. Upper gastrointestinal endoscopy showed an extrinsic mass compressing the mid esophagus 25 to 30 cm beyond the dental arch and an ulcerative lesion covered by adherent fresh blood clots in the midthoracic esophagus. There was no active bleeding.

A subsequent contrast-enhanced chest computed tomographic scan revealed an enormous thoracoabdominal aneurysm measuring 10 cm in diameter in the chest and 16 cm in diameter under the diaphragm. Air bubbles inside the thrombus were suggestive of esophageal erosion (Fig 1).

Urgent operation was performed based on a diagnosis of an AOF secondary to a thoracoabdominal aortic aneurysm. At left thoracophrenolaparotomy, an enormous thoracoabdominal aortic aneurysm was noted with a neck just distal to the left subclavian artery and extending to below the celiac trunk. A small 4-cm-diameter infrarenal aortic aneurysm was noted, and there was a short intermedial segment between the infrarenal aortic aneurysm and the distal extent of the thoracoabdominal aneurysm. Under protection of partial femorofemoral extracorporeal circulation, the thoracic aorta was clamped just distal to the left subclavian artery and below the celiac trunk, and the aneurysm was opened longitudinally. An aortic fistula was apparent in the mid esophagus. The thoracoabdominal aorta was replaced with a cryopreserved arterial allograft (acquired at the European Homograft Bank in Brussels; 14 mm in diameter; 18 cm in length) from the upper thoracic aorta to the celiac aorta, including direct reattachment of the celiac trunk and one intercostal artery. Inspection of the esophagus revealed a 3-cm defect in a necrotic segment, densely adherent to the aneurysm. Subsequently, we performed subtotal esophagectomy, which was completed by lateral cervical esophagostomy. Devitalized and infected tissues, including most of the aneurysm wall, were carefully debrided. The distal esophageal stump was closed with a stapling device, and a jejunostomy tube was placed for enteral feeding. A pyloroplasty was performed to avoid gastric

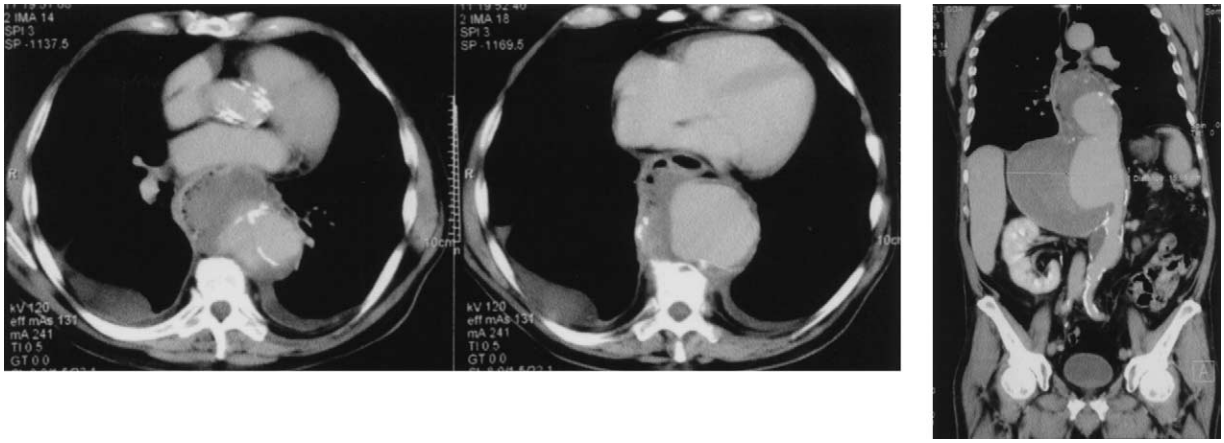

Fig 1. Computed tomography showing air bubbles within the mural thrombus of a thoracoabdominal aortic aneurysm ruptured into the esophagus.

Table I. Cases of successful surgical treatment of primary aortoesophageal fistula secondary to thoracic aortic aneurysm

Study	*Sex*	*Age (y)*	*Surgical procedure on aorta + esophagus*
Fuentes et al[7] 1979	M	75	In situ Dacron* prosthesis + primary repair
Snyder et al[8] 1983	M	66	In situ Dacron graft + primary repair; leak followed by esophagectomy
Snyder et al[8] 1983	F	56	In situ Dacron graft + esophagectomy, immediate intrathoracic esophagogastrostomy
Coselli and Crawford[9] 1990	F	65	In situ Dacron graft + primary repair, omental wrap
Von Oppell et al[10] 1991	M	67	In situ Dacron graft + esophagectomy, immediate cervical esophagogastrostomy, omental wrap
Bogey et al[11] 1992	F	60	In situ Dacron graft + primary repair, wrap with aneurysm wall
Peck and Eidemiller[12] 1992	NA	NA	In situ Dacron graft + primary repair; leak with esophagectomy, intrathoracic esophagogastrostomy, omental wrap
Takasaky et al[13] 1994	NA	NA	In situ Dacron graft + esophagectomy, immediate intrathoracic esophagogastrostomy
Tkebuchava et al[14] 1997	M	73	In situ Dacron graft + primary repair
Hariya et al[15] 1998	F	81	In situ Dacron graft + esophagectomy, immediate intrathoracic esophagogastrostomy
Amin et al[16] 1998	M	76	In situ Dacron graft + esophagectomy, wrap with aneurysm wall, cervical esophagostomy
Goto et al[17] 1998	F	79	In situ Dacron graft + esophagectomy, omental wrap
Nakayama et al[18] 1998	M	57	In situ Dacron graft; postoperative graft infection because of residual esophageal fistula followed by esophagectomy + immediate intrathoracic esophagogastric anastomosis
Da Silva at al[19] 1999	M	72	In situ Dacron graft + esophagectomy, cervical esophagostomy
Mehta et al[20] 2000	F	58	In situ Dacron graft + esophagectomy, omental wrap
Reardon et al[21] 2000	F	48	In situ Dacron graft + primary repair, wrap with aneurysm wall, cervical esophagostomy
Patel et al[22] 2001	M	66	In situ Dacron graft + esophagectomy
Uchida et al[23] 2001	F	78	In situ Dacron graft + esophagectomy, immediate intrathoracic esophagogastrostomy
Van Doorn et al[24] 2002	F	66	Stent-grafting + transhiatal esophagectomy
Pellizzari et al[25] 2002	M	68	Stent grafting
Taniguchi et al[26] 2002	F	68	Extra-anatomic bypass grafting + primary repair
Shiraishi et al[27] 2002	M	76	In situ Dacron graft + omental wrap, covered esophageal stent
Unosawa et al[28] 2003	M	68	In situ Dacron graft + esophagectomy, cervical esophagostomy
Kieffer et al[29] 2003	M	76	In situ allograft + subtotal esophagectomy, cervical esophagostomy
Cho et al[30] 2004	F	71	In situ Dacron graft; without manipulating esophagus
Flores et al[32] 2004	F	67	In situ Dacron graft + esophagectomy, omental wrap
This study	M	74	In situ arterial cryopreserved allograft + esophagectomy, omental wrap, cervical esophagostomy
Tokuda et al[31] 2004	M	67	In situ Dacron graft + esophagectomy, omental wrap

NA, Not available.
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distention. The chest and the mediastinum were copiously irrigated with saline solution. The omentum was mobilized and brought through the diaphragm to fill the mediastinal cavity and to wrap the allograft. Thoracic tubular drainage was deployed.

Intravenous antibiotic coverage was started to control infection. Even though the aneurysmal thrombus was very ill smelling, bacteriological culture was negative. Enteral nutrition was started at the seventh postoperative day. The patient recovered well with-

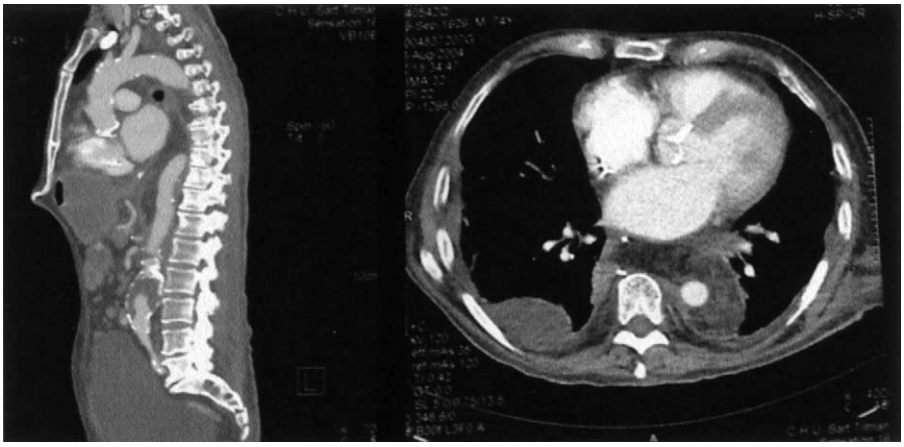

Fig 2. Postoperative magnetic resonance image of the thoracic allograft and omentoplasty around the allograft.

out any complication and was discharged at day 30. A postoperative magnetic resonance image shows the aortic replacement with a tubular allograft and a omentoplasty in the posterior mediastinum around the graft (Fig 2).

Three months later, the patient was readmitted to the hospital for a scheduled second-stage reconstruction of the gastrointestinal continuity by transmediastinal interposition of the right ileocolon with esophagoileal anastomosis in the cervical area and distal cologastric anastomosis. The postoperative course was uneventful, and the patient was discharged in good condition. The patient is currently doing well 12 months after the diagnosis of AOF.

DISCUSSION

Primary AOF is a rare cause of massive and usually fatal gastrointestinal hemorrhage, with few reported survivors.[7-32] The most recent series[29] mentions a mortality rate of 66%. Most patients die before diagnosis secondary to exsanguinating hematemesis.[1,16,19,32,33]

Endoscopy is the procedure of choice in the initial assessment of significant upper gastrointestinal bleeding. Cautious esophagoscopy is the most sensitive and specific method to confirm the diagnosis of AOF. It usually demonstrates a submucosal hematoma and esophageal compression at 25 to 30 cm from the incisors. A pulsating protruding mass in the esophageal wall covered with adherent blood clots is often visualized.[15,19,28,29] A high index of suspicion for AOF is imperative, because biopsy of this ulceration or forcing the endoscope to progress could result in fatal hemorrhage by dislodgement of the occluding blood clot that usually covers the ulcerated lesion. Computed tomographic scanning of the chest is most valuable for surgical planning. Computed tomographic scanning does not demonstrate the fistula, but it delineates secondary signs suggestive of AOF, such as esophageal compression or air bubbles inside the aneurysmal thrombus.[29] Esophageal contrast studies can reveal esophageal perforation by visualizing leakage of contrast medium.[6] Aortography does not add additional information and fails to visualize the fistula. It is considered by most authors as an useless investigation and a loss of time when emergent repair of the AOF is necessary.

Once the diagnosis of AOF is confirmed, surgical correction should be undertaken without delay. Ideally, surgery should be performed on a hemodynamically stable patient before the massive hematemesis. In the past, some authors proposed insertion and inflation of a Blakemore tube inside the esophagus before the thoracotomy to obtain immediate control of the bleeding.[1,32,34] Surgical management consists of a two-stage operation: repairing the thoracic aortic aneurysm and controlling the bleeding is the initial concern, followed by correcting the esophageal defect. Partial extracorporeal cardiopulmonary bypass by femorofemoral cannulation is performed before thoracotomy and clamping of the proximal thoracic aorta. An alternative to venoarterial partial bypass is a left heart bypass. Direct aortic suture or patch angioplasty is conceivable only for small perforations in an otherwise normal aorta, as observed in AOF secondary to fish bone ingestion.[2,32] In a case of atherosclerotic thoracic aneurysm, aortic replacement is required.

An in situ graft replacement of the thoracic aortic aneurysm is the standard procedure. Omentoplasty prevents the accumulation of fluids and clots in the perigraft space and reduces the chance of reinfection. Considering the contamination of the operative field by saprophyte oral flora and esophageal secretions leaking through the erosion, we preferred the in situ insertion of a cryopreserved arterial allograft. Cryopreserved arterial allografts may be more resistant to infection than prosthetic vascular conduits.[29,35,36] However, arterial allografts are not always available "on shelf" in emergency situations; if a cryograft cannot be obtained within a short time, there is no time to waste, and a textile graft soaked in rifampin can be used. In recent years, there has been a revival of interest for in situ aortic allografts for management of both primary and secondary aortic infection.[29,35] Vogt et al[36] reported successful use of an in situ allograft for treatment of seven patients with thoracic aortic infection, including one patient who

presented with a postoperative secondary AOF. Kieffer et al[29] reported on four cases of allograft replacement of the thoracic aorta in the repair of AOF (one secondary and three primary), of which three patients survived.

Percutaneous endovascular stent grafting of the thoracic aortic aneurysm[24,25,37] and extra-anatomic bypass (so-called ventral aorta from the ascending aorta to the infrarenal aorta)[6,26] have been proposed as alternative operative techniques. Endovascular stent grafting is attractive because it is a minimally invasive technique.[37,38] Nevertheless, in the particular case of AOF, endovascular repair of a thoracic aortic aneurysm has some inherent drawbacks. The esophageal fistula is left as such and could reactivate the infection of periaortic tissues in contact with the stent graft.[39] Furthermore, the contaminated posterior mediastinum is neither debrided nor drained. One author proposed a combined minimally invasive approach of endovascular stent grafting and endoscopic injection of fibrin sealant into the esophageal defect. The aneurysmal sac and periaortic abscess are separately drained by a limited left lateral thoracotomy.[38] That particular patient did well for 14 months' follow-up. Bell et al[40] reported on a successful exclusion of a secondary AOF with a stent graft (16.5 months' follow-up).No details are mentioned concerning the management of the esophageal erosion and mediastinal collection.

Endovascular treatment is an ultimate alternative to conventional surgery for high-risk patients with important comorbidities, for whom surgical treatment is excluded in view of a prohibitively high operative risk.[37,38] Endoaneurysmal stent grafting in case of an AOF could be considered as a measure to stop aortic bleeding and as a bridge to second-stage treatment consisting of aortic replacement and esophagectomy. This is a pure theoretical consideration, with so far no cases reported in literature. In our observation, the extent of the thoracoabdominal aneurysm precluded stent graft insertion.

Regarding the treatment of the esophageal defect, some controversy also exists. Neglect of the esophageal defect invariably leads to mediastinitis and death.[40] The two basic approaches are primary repair and limited esophageal resection. It is possible, in selected cases of esophageal erosion by thoracic aortic aneurysm without gross contamination of the mediastinum, to manage the esophageal lesion with primary closure.[7-9,11-13,21,26] When successful, this will simplify the procedure and avoid a secondary surgical intervention to reestablish gastrointestinal tract continuity. However, necrosis of the mid esophagus is partly a consequence of ischemia (occlusion of esophageal arteries that arise directly from the thoracic aorta) and impedes tissue healing. Dehiscence of the esophageal suture and secondary leaking exposes the patient to the risk of sepsis and death.[8,18,26] To minimize the risk of leakage, it is recommended to perform cervical lateral esophagostomy for proximal diversion, tube gastrostomy for distal drainage, and omental transposition into the chest to cover the esophageal repair. Patients receive parenteral feeding for 2 weeks before resuming oral food intake.[21]

Extensive esophageal wall necrosis, triggered by ischemia and infection, may preclude direct esophageal repair and necessitates subtotal esophagectomy with bipolar exclusion by means of cervical esophagostomy and gastrostomy or jejunostomy. We prefer jejunostomy, by means of which the stomach is preserved for subsequent reconstruction of the digestive tract continuity.[19] In our case, we preferred ileocolic interposition between the cervical esophagus and the stomach, because the stomach was densely adherent to the site of the excluded thoracoabdominal aneurysm and the omentoplasty. Moreover, tubulation of the stomach would not have given sufficient length to replace the entire esophagus. Esophageal reconstruction is considered as a second-stage procedure 4 to 6 months after the first operation. A minimum of 6 weeks of intravenous antibiotics is recommended. Some authors prefer to continue lifelong oral antibiotic therapy.[9,19,21,36]

This case is another instance of successful in situ aortic replacement by using a cryopreserved arterial allograft for the management of a primary AOF secondary to a thoracic aortic aneurysm.

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