Bronchoesophageal fistula after endovascular repair of ruptured aneurysm of the descending thoracic aorta

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Aortoesophageal fistula secondary to thoracic aneurysm is rare and is usually fatal without prompt surgical intervention. A 79-year-old man with significant comorbidities and previous cancer surgery was admitted on an emergency basis because of the suspicion of a ruptured thoracic aortic aneurysm. Computed tomographic scan followed by angiography demonstrated a ruptured thoracic aneurysm with aortoesophageal fistula. An endovascular stent graft repair was performed with successful exclusion of both aneurysm and fistula. On postoperative day 6, dyspnea and an isolated episode of hemoptysis occurred. Endoscopy revealed the presence of a bronchoesophageal fistula, which necessitated double exclusion of the esophagus and feeding jejunostomy. At 6 months, clinical, bronchoscopic, and computed tomographic scan follow-up showed complete sealing of the aneurysm and resolution of the bronchoesophageal fistula. At 9 months, the patient was still alive but refused to undergo substernal gastric bypass in an attempt to restore oral feeding. Endovascular repair seems promising as an emergent and palliative treatment of aortoesophageal fistula. To the best of our knowledge, this is the first case in which a bronchoesophageal fistula developed after successful endovascular repair of aortoesophageal fistula. The pathogenesis of this complications remains unclear. (J Vasc Surg 2005;41:712-4.)

Primary aortoesophageal fistula (AEF) is a rare and frequently fatal disorder. Thoracic aortic aneurysm rupture into the esophagus is the most common cause.[1] Surgical repair of AEF carries high mortality, and only a few successful cases are reported in the literature. Recently, endovascular aortic repair of ruptured thoracic aorta has been introduced in an attempt at repairing both the aneurysm and the fistulous tract. Although only a few cases have been reported, this innovative option is increasingly gaining acceptance and is in some institutions the first option for the treatment of AEF. Nevertheless, procedural complications such as endoleak and graft migration may occur, and these often necessitate repeat endovascular intervention or surgical management.[2]

CASE REPORT

A 79-year-old man with systolic hypertension, chronic obstructive pulmonary disease, type 2 diabetes, and a history of myocardial infarction was transferred to our institution on an emergency basis from a community hospital because of the suspicion of ruptured thoracic aortic aneurysm. Three months earlier, the patient had undergone radical cystectomy and bilateral urocutaneostomy. He had been complaining of chest pain and dysphagia for solid food for the past 2 weeks.

Upon hospital admission, the patient complained of acute interscapular chest pain and shortness of breath; peripheral pulses were present, and blood pressure was 100/65 mm Hg. Electrocardiogram and cardiac enzymes were normal. Laboratory tests showed Po_2 96%, hemoglobin 7.6 mg/100 mL, hematocrit 26, and base excess −12. Three units of blood were promptly administered. An emergent computed tomographic (CT) scan showed a ruptured aneurysm of the descending thoracic aortic and blood in the esophageal lumen suggestive of AEF (Fig 1, *A* and *B*). The patient was immediately transferred to the cardiovascular radiology unit.

Diagnostic angiography was performed by a left transbrachial marker pigtail catheter, which clearly indicated the site of AEF. After surgical cutdown of the right femoral artery, a stiff guidewire (Lunderquist; Cook Inc, Bloomington, Ind) was advanced up to the ascending aorta, and a 38 × 160-mm Endofit self-expanding thoracic stent graft (Endomed, Phoenix, Ariz) was implanted with successful exclusion of both aneurysm and fistula. At completion angiogram, adequate proximal and distal fixation of the stent graft were documented, with no evidence of leakage or blood extravasation. Nevertheless, at the end of the procedure, a life-threatening episode of hematemesis and melena occurred that necessitated further transfusion of three units of blood.

On postoperative day 4, the patient was stable and underwent esophagoscopy, which showed submucosal bulging and a mucosal ulcer 25 cm from the incisors, with blood clots in the stomach. On postoperative day 6, dyspnea and an isolated episode of hemoptysis occurred. Flexible bronchoscopy was then performed, and a 10-mm tear of the posterior wall of the left main bronchus was visualized. It was noted that through this tear, the esophagus could be easily entered with the bronchoscope, and this led to the diagnosis of bronchoesophageal fistula (Fig 1, *C*).

Three days later, a bipolar exclusion of the esophagus was performed. The procedure consisted of transection of the cervical esophagus followed by terminal esophagostomy, double-stapling of the cardia with vagal preservation, and feeding jejunostomy. No

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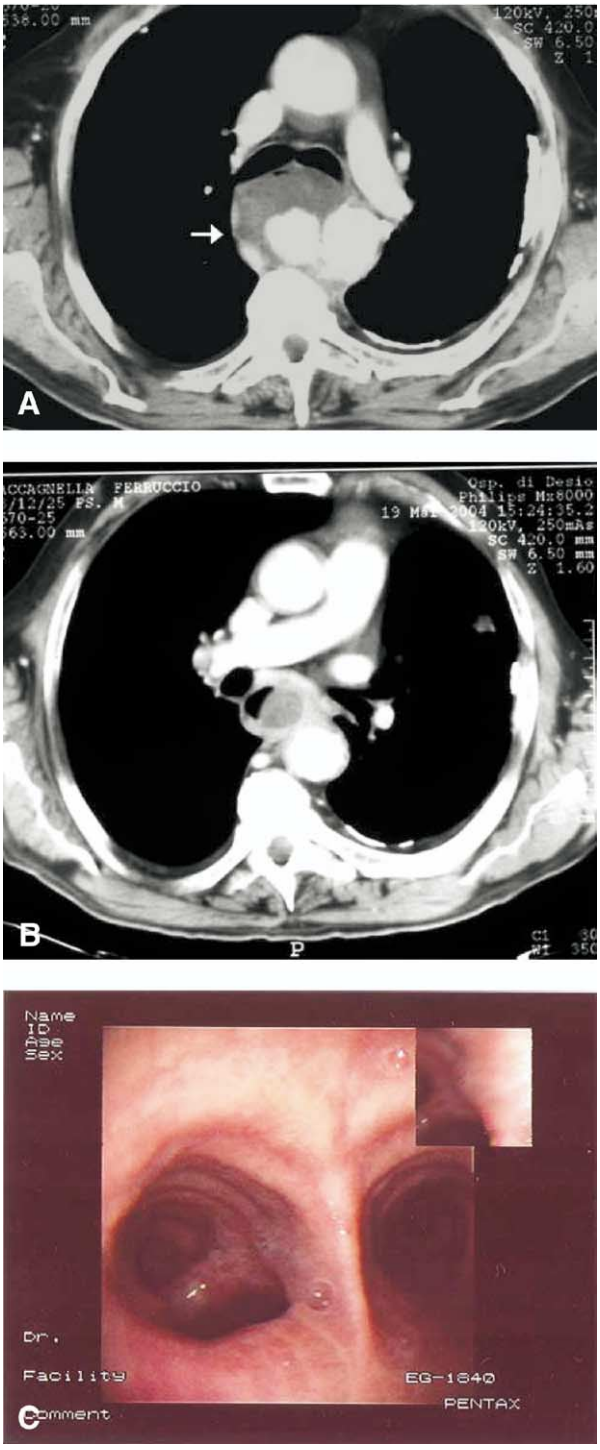

Fig 1. A, Enhanced chest computed tomographic scan demonstrates rupture of a descending thoracic aortic aneurysm with mediastinal hematoma. **B,** Caudal view showing blood clots in the esophagus due to an aortoesophageal fistula. **C,** Bronchoscopic image showing a defect in the posterior aspect of the left main bronchus.

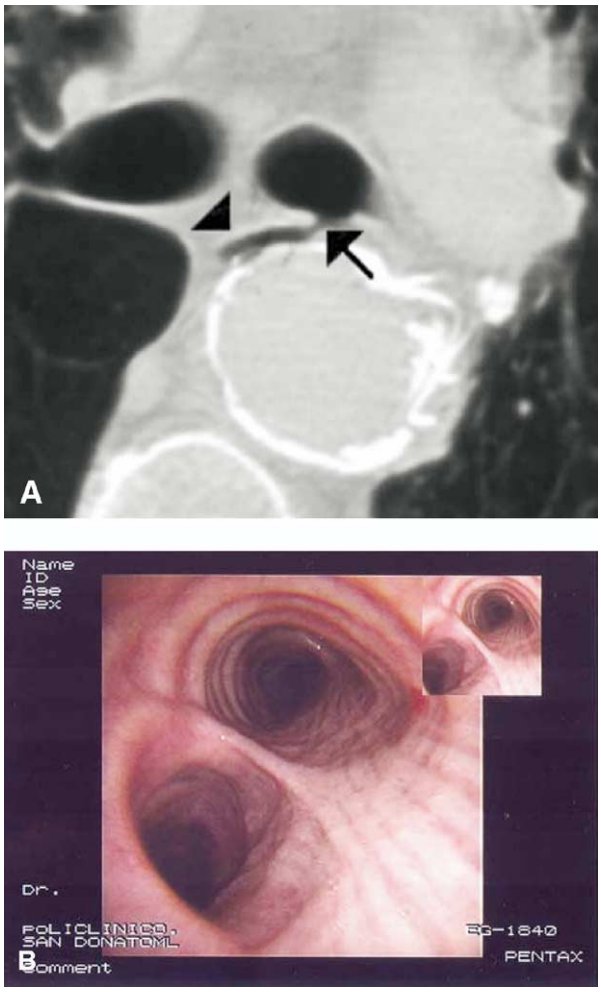

Fig 2. Bronchoesophageal fistula after endovascular repair. **A,** After successful deployment of a thoracic stent graft, chest computed tomographic scan demonstrates residual communication *(arrow)* between the left main bronchus and esophagus *(arrowhead)*. **B,** At 6-month follow-up, complete resolution of bronchoesophageal fistula was documented at bronchoscopy.

postoperative complications occurred. Toilet bronchoscopy was performed daily over the next 7 days. On postoperative day 29, a chest CT scan showed significant reduction of the bronchoesophageal fistula and complete exclusion of the thoracic aneurysm, with no evidence of any type of endoleak (Fig 2, *A* and *B*). Two days later, the patient was discharged home under wide-spectrum antibiotic coverage. He was afebrile and in stable condition. At the 6-month clinical, endoscopic, and CT follow-up, the patient was doing well, with complete resolution of both thoracic aneurysm and bronchoesophageal fistula (Fig 2, *C*). At 9 months, the patient was still alive but refused to undergo substernal gastric bypass in an attempt to restore oral feeding.

DISCUSSION

AEF was described almost two centuries ago by Dubrueil,[3] a French naval surgeon who assisted a sailor

with thoracic pain and hematemesis after ingestion of a beef bone fragment. The sailor died 5 days later, and autopsy revealed a fistula between the esophagus and the thoracic aorta. In 1914, Chiari[4] described the "aortoesophageal syndrome," which was characterized by chest pain followed by a relatively asymptomatic latent period, sentinel hemorrhage, and later exsanguination, which in 60% of patients occurs within 6 hours and is usually fatal.

Rupture of a thoracic aortic aneurysm into the esophagus was first reported by Funk[5] in 1918, but it was only in 1983 that the first successful repair was performed.[6] However, the prognosis of AEF after surgical repair remains poor. Awareness of the disease, a high index of suspicion, and an early diagnosis are crucial to successful treatment of AEF. In patients with midthoracic pain and hematemesis, particularly in those whose histories suggest thoracic aortic aneurysm, foreign body ingestion, or esophageal carcinoma, the diagnosis of AEF should be seriously considered. Thoracic aortic aneurysm is regarded as the most common cause of AEF. According to Carter et al,[1] as many as 75% of AEFs are due to thoracic aortic aneurysm. The esophageal fistula occurs as a result of chronic pressure causing erosion and ischemic necrosis of the esophageal wall. Although esophageal resection with restoration of gastrointestinal continuity, in either a primary or staged fashion, combined with prosthetic aortic repair may prevent this complication, some authors prefer the use of stent graft for the treatment of AEF, particularly when patients are severely debilitated or when urgent treatment is needed. Several successful reports of AEFs have recently been described with patients alive at follow-up.[7,8] In our case, we believed that immediate exclusion of the esophagus by endovascular aortic repair might decrease the chance of mediastinal infection (regarded as the main cause of late mortality in such patients) and allow esophageal reconstruction with a substernal bypass at a later date. Also, because of the poor general condition of the patient, the endovascular option was deemed most appropriate.

Endovascular aortic repair is a relatively new technique that still has many issues to be solved; technical failure and early or late vascular complications such as leak and graft migration are reported. In our case, although the stent graft placement was successful, with immediate exclusion of the AEF, the patient developed a fistula between the esophagus and the left main bronchus. Bronchoesophageal fistula is a rare complication that can occur from a variety of causes. The most common etiology is esophageal cancer, but lung cancer, radiotherapy, and esophageal diverticula account for other possible causes.[9]

Fistulas between the tracheobronchial tree and esophagus are very rare after thoracic aorta repair. A case of AEF in which the aneurysm was replaced with a Dacron tube (DuPont, Wilmington, Del) in situ and the esophagus was primarily repaired was reported by Coselli and Crawford[10]; despite the successful result of the procedure, the patient developed a fistula between the esophagus and the tracheobronchial tree. To our knowledge, this life-threatening complication has never been reported after endovascular aortic repair of AEF.

No evident causative factors of the bronchoesophageal fistula were found in our patient. Although on CT scan the mid portion of the device was shown to be adjacent to the site of bronchoesophageal fistula, no direct responsibility of the stent and the endovascular procedure itself can be envisaged. Rather, the almost-simultaneous occurrence of AEF and bronchoesophageal fistula may have been the result of the natural history of the ruptured aneurysm and the mediastinal hematoma. Indeed, because the periaortic hematoma may persist for several days after stent graft repair,[11] the esophagus and tracheobronchial tree may have been compressed after the procedure, and this may have led to esophageal and bronchial ischemia and subsequent bronchoesophageal fistula development.

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