Rare complication of esophageal necrosis and perforation after fenestrated endovascular aneurysm repair

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ABSTRACT

Fenestrated endovascular aneurysm repair (FEVAR) is a minimally invasive technique used to treat complex abdominal aortic aneurysms. We present the case of a 69-year-old man with a juxtarenal abdominal aortic aneurysm treated with FEVAR. The patient experienced postoperative dysphagia and sepsis. Investigations revealed a perforated esophagus due to esophageal ischemia and necrosis, leading to complete esophagectomy and subsequent esophageal reconstruction. This case highlights esophageal necrosis and perforation as a potential complication of FEVAR and serves as a reminder to have a low threshold for investigating and emergently managing this condition, which otherwise has a high mortality rate. (J Vasc Surg Cases and Innovative Techniques 2020;6:181-4.)

Keywords: EVAR; FEVAR; Complications; Esophageal; Ischemia; Necrosis

Abdominal aortic aneurysm rupture can lead to fatal hemorrhage if it is left untreated. Historically, open surgical repair had been the sole treatment option for patients. However, as minimally invasive techniques have evolved, complex thoracoabdominal and juxtarenal aortic aneurysms can be treated in an endovascular approach with fenestrated endovascular aneurysm repair (FEVAR).

FEVAR involves the placement of a stent graft intraluminally under fluoroscopy guidance to exclude the aneurysmal sac while maintaining blood flow to the visceral arteries. In comparison with open surgery, the treatment of complex abdominal aneurysms with FEVAR has been found to result in less intraoperative blood loss, shorter length of hospital stay, and fewer cardiopulmonary and neurologic complications after the procedure.[1,2] Nevertheless, FEVAR is associated with its own complications, including but not limited to acute renal impairment, endoleak, device migration, visceral stent occlusion, and vascular access injury.[3,4]

We present a rare case of FEVAR complicated by esophageal necrosis in the postoperative period, requiring emergent esophagectomy and esophagostomy.

Written informed consent was obtained from the patient for publication of this case.

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Author conflict of interest: none.

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CASE REPORT

A 69-year-old man was assessed for treatment of a complex juxtarenal aortic aneurysm. The aneurysm was imaged using arterial-phase computed tomography (CT) and measured 5.9 cm in maximal diameter (Fig 1). CT otherwise demonstrated conventional vascular anatomy. The patient's past surgical history included endovascular repair of a previously ruptured left common iliac artery aneurysm as well as thromboembolectomy of the popliteal artery after a delayed postoperative embolism from thrombus lining the iliac stent graft. Subsequently, the patient underwent an urgent right carotid endarterectomy for a transient ischemic attack before elective FEVAR.

Access for FEVAR was through surgical cutdown of the common femoral arteries. During operative exposure of the relatively short left common femoral artery, the external iliac artery tore circumferentially while it was being encircled with a Silastic vessel loop for proximal control. The patient became temporarily hypotensive with a blood pressure of 70/40 mm Hg, which stabilized with fluid resuscitation. The cell saver was introduced at this point. Retroperitoneal exposure of the external iliac artery for repair was challenging secondary to scarring from the patient's previous iliac aneurysm rupture. An iliac occlusion balloon from the contralateral femoral access provided control, allowing arterial repair and placement of a Dacron conduit (left external iliac to superficial femoral artery) for access to the aortic endograft.

Digital subtraction angiography of the aorta was performed through a pigtail catheter, and the body of the fenestrated stent graft (Cook Medical, Bloomington, Ind) was appropriately positioned and partially deployed. Both renal arteries were cannulated through the appropriate fenestrations, 6F Destination sheaths (Terumo, Tokyo, Japan) were inserted, and a 6- × 22-mm Atrium stent (Getinge, Gothenburg, Sweden) was positioned across each ostium. The superior mesenteric artery (SMA) was then cannulated through its fenestration, and the 5F short sheath was exchanged for a 7F Destination sheath, which was advanced into the SMA and used to position a 7- × 38-mm Atrium stent. The fenestrated body was released

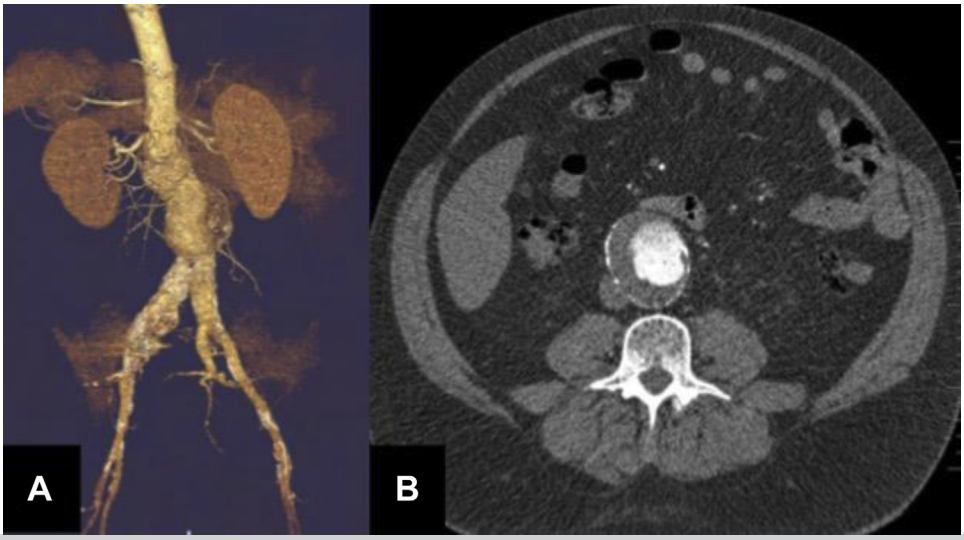

Fig 1. A, Posteroanterior three-dimensional reconstruction of complex juxtarenal abdominal aortic aneurysm. **B,** Axial computed tomography (CT) image of same juxtarenal abdominal aortic aneurysm with intramural thrombus within the sac.

proximally and distally, and the Atrium stents were deployed. Finally, the celiac artery was cannulated, a 7F Destination sheath was advanced into the celiac trunk lumen, and an 8- × 38-mm Atrium stent was deployed across the ostium, with coverage of the left gastric artery (Fig 2, *A*). A Cook bifurcated graft was then inserted from the right groin and the contralateral limb through the left groin. Retrospectively, it was also noted that the splenic artery was not opacified on the post-celiac stent angiogram (Fig 2, *B*), which correlated with coverage of the splenic artery origin on postoperative CT scan by the celiac Atrium stent.

Proximal and distal seal and graft overlaps were dilated with a Coda balloon (Cook Medical). Final digital subtraction angiography confirmed patency of all four visceral stents and both iliac limbs, with no evidence of endoleak. Total blood loss was 4 L, with 3 L returned through the cell saver.

Two days after the procedure, the patient complained of dysphagia. He subsequently developed profound septic shock, followed by respiratory failure requiring intubation and ventilation, and was commenced on broad-spectrum antibiotics. CT on postoperative day 5 demonstrated pneumomediastinum and extraluminal oral contrast material, indicating perforation of the distal portion of the esophagus (Fig 3). Esophagogastroscopy was promptly performed, which demonstrated the middle and distal portions of the esophagus to be ischemic with black mucosa. At the level the gastroesophageal junction, a sizable perforation on the right side of the esophagus was identified. The stomach and proximal duodenum were of normal appearance. Multiple splenic infarcts were also noted on follow-up imaging.

The patient underwent emergent total esophagectomy and cervical esophagostomy and subsequently received long-term nutrition through a jejunostomy tube. Histologic evaluation of this specimen demonstrated transmural infarction with serosal fibrinopurulent exudate and a gross 2- × 3-cm perforation, findings pathologically consistent with ischemic changes. One year later, the patient underwent a second-stage procedure that involved reconstruction of the esophagus with a jejunal interposition. At 1 year after esophageal reconstruction, the patient was clinically well and able to swallow with satisfactory appearances on esophagogastroscopy. There was no evidence of graft infection.

DISCUSSION

The pathogenesis of esophageal necrosis in this case was multifactorial, with a series of insults contributing to esophageal ischemia. The esophageal blood supply is segmentally derived: the cervical area, from the inferior thyroid arteries; the thoracic segment, from the tracheobronchial artery branches as well as esophageal arteries originating directly from the thoracic aorta; and the abdominal esophagus, predominantly by branches of the left gastric artery and to a lesser extent branches of the left phrenic artery and splenic artery.

Hypotension as a cause of reduced perfusion pressure is a well-documented cause of esophageal ischemia and ultimately necrosis.[5] Hemorrhagic shock after variceal bleeding, rupture of aortic aneurysms, and iatrogenic perisurgical blood loss have been reported as causes of esophageal necrosis in the literature.[6-8] The hypotensive effect of the left external iliac artery injury could therefore certainly have caused an ischemic burden on the esophagus. Other hypotensive causes of hemodynamic compromise resulting in esophageal necrosis include septic shock, cardiogenic shock, and hypovolemic shock.[5]

We hypothesize that ischemic esophageal necrosis occurred in our case in part because of coverage of the

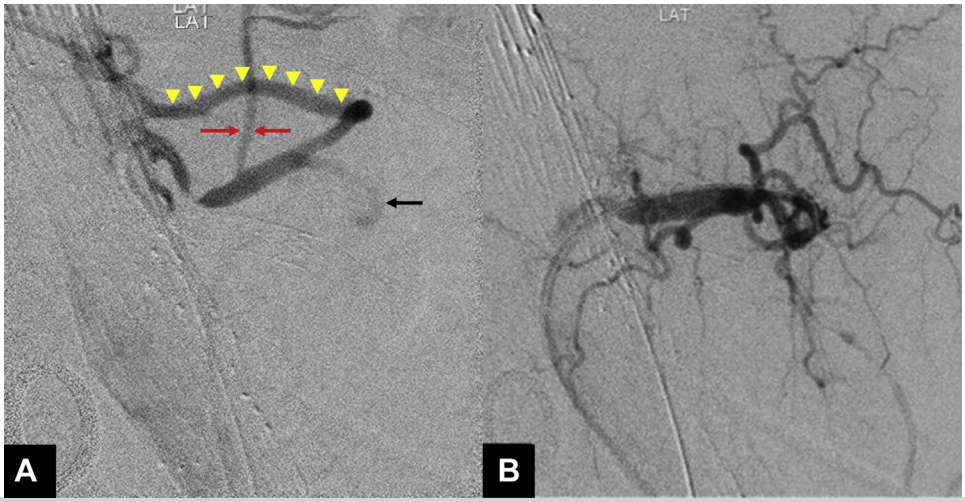

Fig 2. A, Lateral view of the celiac trunk with left gastric artery originating proximally (*red arrows*) and coursing in a cranial direction. Splenic artery is demarcated with *yellow arrowheads*. Common hepatic artery is identified by *black arrow*. **B,** Deployment of celiac trunk Atrium stent with complete coverage of the left gastric artery. The previously opacified splenic artery is no longer visualized, indicating coverage of the origin by the covered stent.

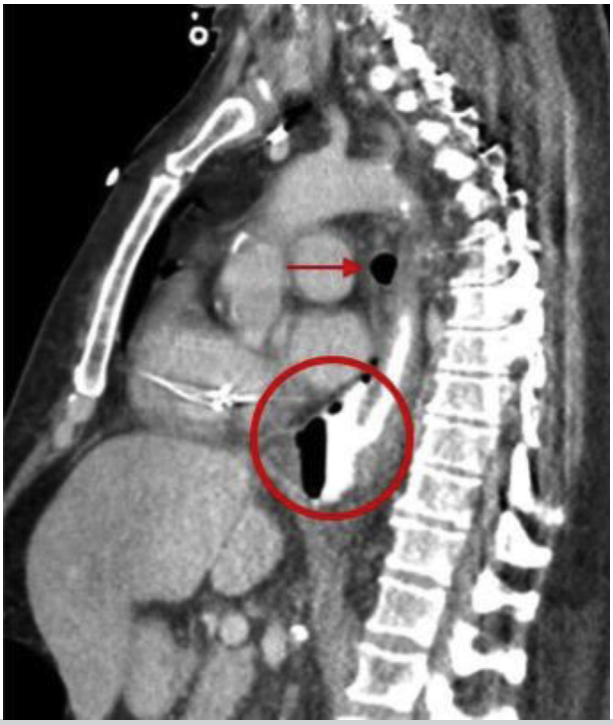

Fig 3. Sagittal computed tomography (CT) views of large perforation involving the distal portion of the esophagus with extraluminal oral contrast material (*circle*). Pneumomediastinum is also present (*arrow*).

left gastric artery during stenting of the celiac trunk, which is common during FEVAR procedures to maintain stent stability. Furthermore, stent coverage of the splenic artery origin would have caused a reduction in collateral flow to the esophagus through the short gastric arteries. The phrenic arteries were covered by the endograft, which would have further impeded collateral blood flow to the esophagus. Although the foregut has a rich collateral blood supply, previous studies have reported upto 16% major ischemic complication rate after embolotherapy for upper gastrointestinal bleeding, indicating that this collateral network is variable, and certain patients may be at higher risk of the complications of impaired blood flow.[9] A study by Song et al[10] described multiple collateral pathways in patients with celiac stenosis, including right gastric to left gastric and left hepatic to left gastric arterial anastomoses; these anastomoses may be absent in those patients who develop esophageal and gastric ischemia after FEVAR. Our patient had a relatively heavy burden of complicated vascular disease and may have been more prone to compromise of the collateral network to the esophagus.

Esophageal necrosis with or without perforation has been described as a rare sequela of thoracic endovascular aortic repair (TEVAR) for both aneurysmal disease and dissection, with a high mortality rate. Yaguchi et al[11] presented a case series of seven patients who developed esophageal perforation between 12 days and 2205 days after TEVAR for thoracic aortic dissection or aneurysm. The group postulated several potential factors that may result in esophageal perforation, including direct erosion of the stent graft into the esophagus, graft infection, pressure necrosis caused by the expanding stent graft, and ischemic esophageal necrosis due to disruption of the blood supply to the esophagus. Further case reports by Koizumi et al[12] and Tobisch et al[13] have also described esophageal necrosis after TEVAR for aortic dissection. Both authors agree that as well as impaired blood flow to the esophagus secondary to the endoprosthesis, mediastinal hematoma reducing intramural arterial blood supply and causing stagnation of venous outflow

is critical in the development of esophageal necrosis after TEVAR. Interestingly, a study by Katahira et al[14] confirmed significant reduction in local esophageal mucosal blood flow using a novel sensor in a swine model after thoracic endovascular graft implantation.

CONCLUSIONS

To our knowledge, esophageal necrosis occurring after FEVAR has not been reported in the literature. On reflection, given the initial complication of hemorrhagic shock, if a similar scenario were to occur in the future, preservation of the splenic artery could be a prudent measure. If there is sufficient collateral supply to the hepatic arteries through pancreaticoduodenal artery anastomoses from the SMA, stenting into the splenic artery to preserve short gastric supply to the esophagus could be considered. Furthermore, in patients in whom esophageal collateralization is thought to be compromised as a result of vasculopathy, a scalloped stent graft may allow avoidance of celiac axis stenting, therefore preventing potential bridging stent coverage of the splenic artery origin.

As the symptoms of esophageal ischemia and necrosis are usually nonspecific, including pain, sepsis, and dysphagia, this is likely to be an under-recognized complication. Given the high mortality rate of esophageal necrosis, clinicians should be vigilant and have a low threshold for investigating early potential clinical signs of this complication.

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