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Case Report

Recurrence of Aortoenteric Fistula after Endovascular Aortic Repair

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Aortoenteric fistula (AEF) after endovascular aortic repair (EVAR) is a rare complication, with only 32 cases reported previously. A 71-year-old man who presented with severe duodenal bleeding due to primary AEF (PAEF) underwent successful EVAR. Four years later, the AEF recurred because of dilatation of the aneurysm sac. He underwent emergent surgery, removal of the stent graft, and replacement of an artificial bifurcated graft with placement of a greater omental flap. EVAR for PAEF was an effective option for acute treatment, but it caused refistulization in the long term. EVAR should be considered as a bridge therapy to definitive surgery.

Keywords: type 2 endoleak, endovascular aortic repair, open surgery

Introduction

A primary aortoenteric fistula (PAEF) is exceedingly rare but can cause life-threatening gastrointestinal tract bleeding and infection. Although endovascular aortic repair (EVAR) is a good choice for acute treatment of aortoenteric fistula (AEF), with other such cases having been reported, the long-term outcome is unclear. Here we report a recurrent case of AEF 4 years after successful EVAR for PAEF.

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Case Report

A 71-year-old man was hospitalized with massive hematemesis and loss of consciousness after a traffic accident. Laboratory data showed Hb 8.6 g/dl, Ht 24.1%, WBC 11780/μl, BUN 27.7 mg/dl, and Cre 0.70 mg/dl. His diagnosis was PAEF, duodenal ulcer, and abdominal aortic aneurysm (AAA, ϕ45 mm) detected by enhanced computed tomography (CT) (**Fig. 1A**) and upper gastrointestinal endoscopy (**Fig. 1B**). First, we offered him an open repair, but he declined the option and accepted the second plan, EVAR, instead. His blood cultures were taken and he underwent emergency EVAR (Zenith Flex, Cook Medical Inc., Bloomington, IN, USA). The postoperative course was uneventful, his blood culture tests were negative, and an oral antibiotic therapy was continued for more than 1 year. In the outpatient clinic, signs of inflammation, aggravation of anemia (Hb 13.0 g/dl, Ht 37.3%), or shrinkage of AAA (ϕ35 mm, **Fig. 1C**) were not observed. Four years later, the patient complained of hematemesis and melena without abdominal pain. CT examination showed a dilatation of the remaining portion of the AAA (ϕ45 mm, **Fig. 1D**) due to type 2 endoleak from the inferior mesenteric artery (IMA) and a protrusion of the aortic wall facing the duodenum. His anemia worsened, Hb 10.0 g/dl and Ht 29.5% and diagnosis of recurrent AEF was suspected. Emergent angiography for aneurysm sac embolization was performed. Only a small retroperitoneal branch of the IMA was identified on Riolan's arcade angiogram via the superior mesenteric artery, but the aneurysm sac was not detected. After administering red cell concentrates (RCC) and fresh-frozen plasma (FFP), the vital signs became stable, but the anemia did not improve. We therefore decided to perform open-abdomen surgery.

At laparotomy, the intra-abdominal organs were not adhesive and the aortic sac was not inflamed. After obtaining proximal (infrarenal aorta) and distal (common iliac arteries) control, the aortic aneurysm sac was opened (**Fig. 2A**). It was filled with a relatively fresh blood clot and backflow bleeding from the IMA, which caused the type 2 endoleak. First, the IMA was ligated, and the duodenal wall around the fistula was normal (**Fig. 2B**), so the

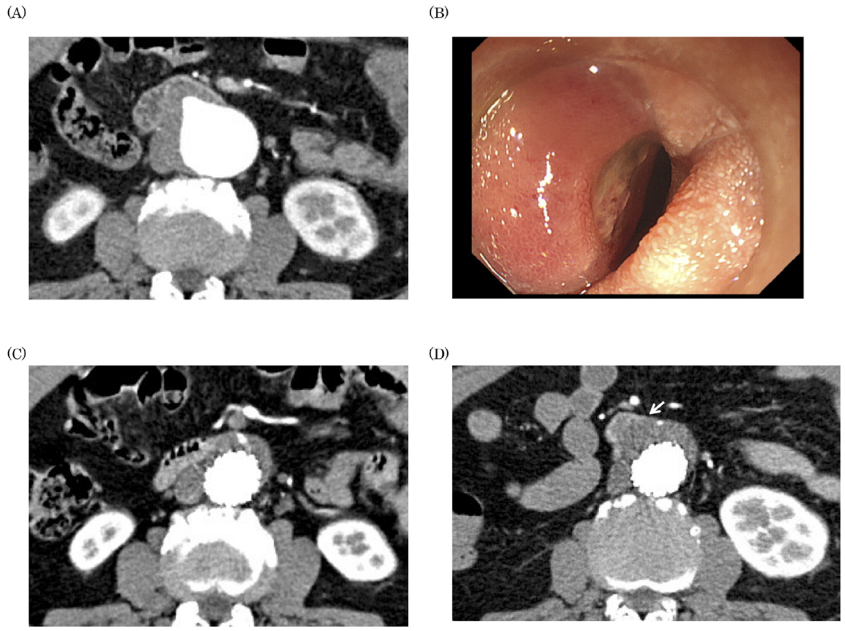

Fig. 1 (**A**) Computed tomography (CT) image showing dilatation of the abdominal aortic aneurysm and a protrusion of the aortic wall facing the duodenum. (**B**) Upper gastrointestinal endoscopy showing a pulsatile mass and an ulcer in the third portion of the duodenum. (**C**) CT imagery from 2 years postoperatively, (**D**) CT imagery from 4 years postoperative showing a lack of continuity in the duodenal wall facing the aortic aneurysm (arrow).

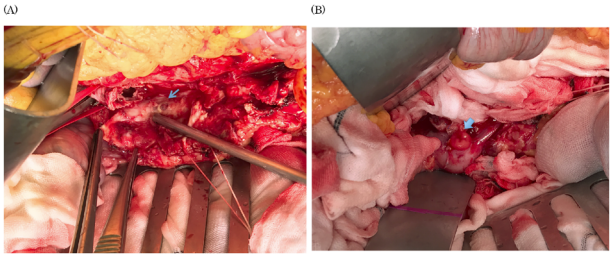

Fig. 2 Operative view.
(**A**) The aortic aneurysmal sac was opened. The narrow arrow points to the fistula. (**B**) The duodenal fistula (wide arrow).

duodenal portion of the fistula was repaired simply in two layers. Complete removal of the stent graft was difficult because of the suprarenal barb fixation at the level of the renal arteries. Almost all of the stent graft was removed, but a part of the bare metal stent and top stent graft was left in place at the level of the right renal artery. A Dacron bifurcated artificial graft (INTERGARD 22 mm × 11 mm, MAQUET Cardiovascular LLC, Wayne, NJ, USA) was placed, and the greater omentum was interposed between the duodenum and the aortic wall. Finally, a temporary feeding jejunostomy was created.

The patient was transferred to the intensive care unit in stable condition, and with transfusion of RCC and FFP, his postoperative course was uneventful. Bacterial cultures of the removed stent graft and resected aortic wall were negative. On postoperative day 10, oral ingestion was restarted, and the jejunostomy was removed. The patient was discharged with oral antibiotic therapy. One year after the open repair, he showed no sign of inflammation or anemia.

Discussion

AEF is a rare but well-known complication after open AAA surgery, in which case it is termed as secondary AEF. PAEF has an incidence rate of 0.04%–0.07%, and the incidence of AEF after EVAR is poorly defined, with only a few reported cases.[1–4] According to these reports, persistent endoleak, graft migration, kinking, graft material breakdown, and inflammatory AAA, led to the risk of fistula formation.[2] In our case, the AEF recurred as a consequence of an increase in the size of the aortic aneurysm sac due to type 2 endoleak after EVAR for PAEF. Although the EVAR was effective for sealing the bleeding point in the acute phase of treating our patient, primary fistula repair was not performed during the first operation. The patient had no symptoms of infection and had been on antibiotic therapy for one year after discharge.

Surgical treatment must be associated with antibiotic therapy, but there are no guidelines on the exact duration of treatment. In the short term, EVAR with antibiotic therapy for AEF seems to be associated with decreased

perioperative morbidity and mortality and a shorter inhospital stay.[5] However, in the long term, EVAR for AEF (both primary and secondary) is reported to be associated with a high incidence of persistent/recurrent/new infection or recurrent bleeding.[6] In the previous 32 cases, the time between EVAR and diagnosis of AEF was 4 to 60 months.[4] In our case, AEF recurred 48 months after EVAR. Without fistula repair, the vulnerabilities of the aortic wall and duodenum wall may result in a higher rate of refistulization within several months. Therefore, EVAR for AEF should be considered a "bridge" option until a more definitive repair is performed at a later time. Close examination and CT follow-up are necessary, and when the aneurysm sac is dilated, open repair intervention should be considered without hesitation.

Conclusion

EVAR for PAEF is an effective option for acute treatment, but in the long term, it may contribute to a refistulization due to endoleak. We should consider a stepwise treatment as a "bridge" to further surgery at a later time.

Disclosure Statement

None declared.

Author Contributions

Study conception: DA, YS
Data collection: DA
Analysis: DA
Investigation: DA
Writing: DA
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References

1) Chenu C, Marcheix B, Barcelo C, et al. Aorto-enteric fistula after endovascular abdominal aortic aneurysm repair: case report and review. Eur J Vasc Endovasc Surg 2009; **37**: 401-6.
2) Ruby BJ, Cogbill TH. Aortoduodenal fistula 5 years after endovascular abdominal aortic aneurysm repair with the Ancure stent graft. J Vasc Surg 2007; **45**: 834-6.
3) Arworn S, Orrapin S, Chakrabandhu B, et al. Aorto-enteric fistula after endovascular abdominal aortic aneurysm repair for Behcet's disease patient: a case report. EJVES Short Reports. 2018; **39**: 54-7.
4) Koda Y, Murakami H, Yoshida M, et al. Secondary aorto-enteric fistula and type II endoleak five years after endovascular abdominal aortic aneurysm repair. EJVES Short Reports. 2019; **43**: 12-7.
5) Baril DT, Carroccio A, Ellozy SH, et al. Evolving strategies for the treatment of aortoenteric fistulas. J Vasc Surg 2006; **44**: 250-7.
6) Antoniou GA, Koutsias S, Antoniou SA, et al. Outcome after endovascular stent graft repair of aortoenteric fistula: a systematic review. J Vasc Surg 2009; **49**: 782-9.