

Case Report

Primary aortoduodenal fistula with a history of distal gastrectomy

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Case: A 69-year-old man was transferred to our hospital because of an aortoduodenal fistula with hematemesis and pre-shock vital signs. He had a history of alcoholism, malnutrition, and distal gastrectomy and Billroth I reconstruction. Endovascular aneurysm repair was successfully carried out; however, the presence of comorbidities affected further radical treatment.

Outcome: The patient survived for 2 months postoperatively.

Conclusion: Endovascular aneurysm repair is a useful first-line treatment for high-risk aortoduodenal fistula patients; however, it requires improvement for long-term outcomes in complicated high-risk cases.

Key words: Acute abdomen, endovascular aneurysm repair, gastrectomy, primary aortoduodenal fistula

INTRODUCTION

AORTODUODENAL FISTULA (ADF) is defined as a communication between the native aorta and duodenum and is an extremely rare cause of gastrointestinal bleeding. Aortoduodenal fistula should be studied because the mortality rate of untreated ADF with upper gastrointestinal bleeding is almost 100%.¹ Recently, endovascular aneurysm repair (EVAR) has been attempted as a first-line treatment of choice for ADF.^{2,3} With the aging of the population, the number of elderly patients and patients with a greater number of comorbidities and surgical histories has also increased.⁴

Here, we present a very rare case of primary ADF that was complicated by a history of distal gastrectomy and Billroth I reconstruction in a patient with poor preoperative status.

CASE

A 69-YEAR-OLD JAPANESE man with hematemesis and severe anemia was transferred to the emergency unit of our hospital. He had no previous history of hematemesis, and he arrived at our hospital 6 h after this

first episode. He had presented at a hospital 6 months previously because of alcoholism and malnutrition, causing large sores. He was bedridden and emaciated (body mass index, 14.08 kg/m²), and he received nutritional therapy with oral nutrition and i.v. hyperalimentation. The patient also had a history of distal gastrectomy for gastric cancer 8 years previously, with no history of abdominal aortic aneurysm (AAA) treatment.

Physical examination revealed the following findings: pulse, 110 b.p.m.; blood pressure, 110/76 mmHg; body temperature, 35.9°C; and respiratory rate, 16 breaths/min. His abdomen was flat, hard, and painful. A palpable pulsatile mass was detected in the umbilical region. He also had peripheral coldness. Laboratory investigation revealed severe anemia (hemoglobin, 6.5 g/dL), inflammation (white blood cell count, 13,300/μL; C-reactive protein [CRP], 7.25 mg/dL), malnutrition (albumin, 2.0 g/dL), and a coagulation disorder (prothrombin time – international normalized ratio, 1.66; activated partial thromboplastin time, 52.6 s; and fibrin degradation products, 27.0 μg/dL). There were no data that were suggestive of renal or liver dysfunction. Enhanced computed tomography (CT) showed a 50 × 52 × 53-mm infrarenal AAA that was saccular and connected to the duodenum by a hematoma (Fig. 1A, B). The hematoma was also present in the stomach and duodenum (Fig. 1A, white arrows). Based on the clinical course and results of the investigations, the patient was diagnosed with ADF because of AAA rupture.

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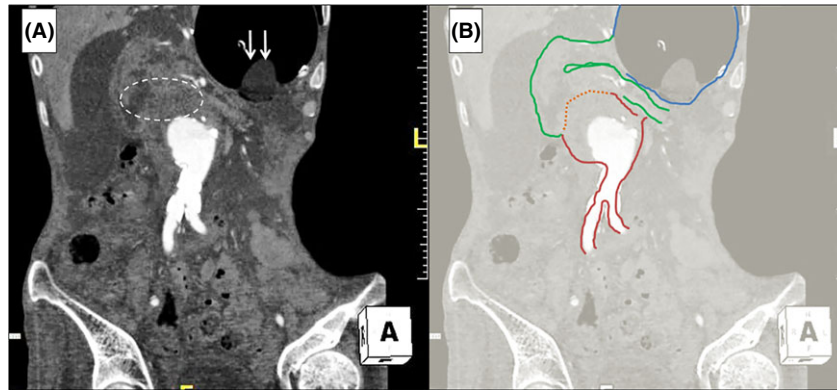


Fig. 1. Computed tomography images of a 69-year-old man with aortoduodenal fistula, taken on arrival at our hospital. A, A large hematoma was found in the stomach and duodenum (white arrows). A sacular aneurysm was connected to the duodenum by the hematoma (white circle). B, Anatomical computed tomography findings. Blue, green, and red lines represent the outlines of the stomach, duodenum, and aneurysm, respectively. Dotted line indicates the unclear boundary between the duodenum and aneurysm.

We initially decided to treat the patient with EVAR to control the bleeding. A laparotomy was also planned to repair the duodenal perforation. The operation was started 90 min after the patient's arrival at the hospital. At this point, hemodynamic status was maintained by fluid infusion (pulse, 119 b.p.m.; blood pressure, 105/77 mmHg). First, a stent graft (Excluder; W.L. Gore, Flagstaff, AZ, USA) was successfully deployed in the infrarenal aorta. Angiography showed no extravasation or endoleak, and the bleeding was controlled (Fig. 2A, B). The operation time for EVAR was 56 min. Consequently, laparotomy was carried out through a median incision. Bloody ascites was found in the abdominal cavity. The stomach and duodenum were anastomosed using the Billroth I method. The transverse mesocolon and duodenum were densely and broadly adhered to

the AAA with fibrous tissue (Fig. 2C). Therefore, we could not separate the aneurysm and duodenum. This prevented direct identification and repair of the ADF. Intraoperative gastrointestinal endoscopy was carried out; however, we could not locate the duodenal perforation. There was no active bleeding. Accordingly, we decided to undertake intestinal tubing for decompression and indirect duodenal lesion isolation. Two tubes, 12- and 10-Fr, were inserted into the oral and anal sides of the jejunum, respectively (Fig. 3A). Additional procedures were deemed to be too invasive for this patient, and the operation was ended. The total surgery time was 4 h 50 min. The total blood loss was 850 mL, and 1120 mL of suspended red cells were transfused. We planned a second elective operation for retrieval of duodenal perforation closure or surgical isolation of the

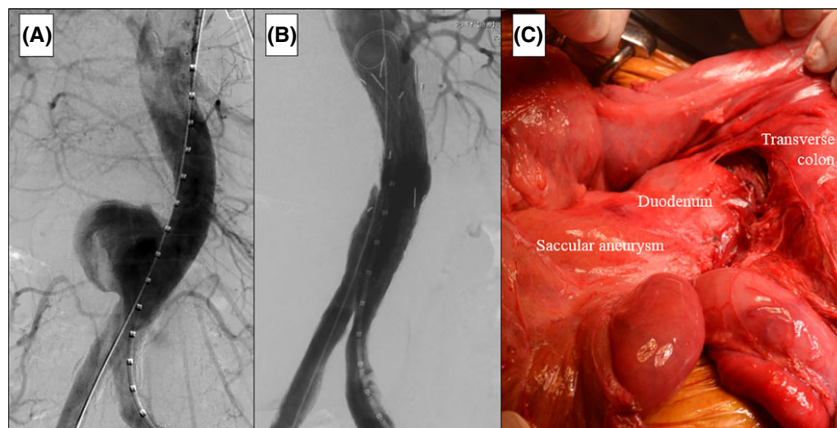


Fig. 2. Intraoperative findings in a 69-year-old man with aortoduodenal fistula and a history of alcoholism, malnutrition, and distal gastrectomy and Billroth I reconstruction. A, Angiography showed a sacular aneurysm. B, After endovascular aneurysm repair, the aneurysm disappeared and no extravasation or endoleaks remained. C, The transverse mesocolon and duodenum densely adhered to the abdominal aortic aneurysm by fibrous tissue. The aneurysm was covered by the mesocolon.

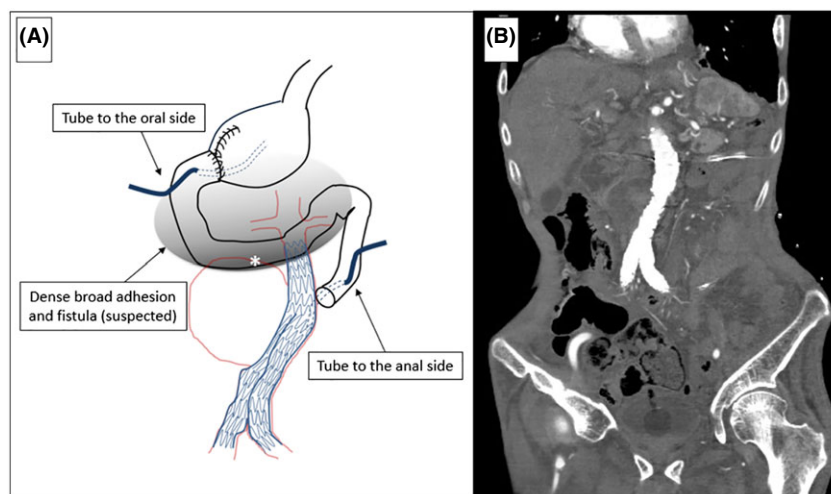


Fig. 3. Schematic representation of the operative findings and our procedures in a 69-year-old man with aortoduodenal fistula who underwent endovascular aneurysm repair. A, Abdominal aortic aneurysm and the duodenum were densely and broadly adhered to each other. The fistula was suspected to exist on the third portion of the duodenum (*). Following endovascular aneurysm repair, two tubes were inserted in the oral and anal sides of the jejunum for intestinal tract decompression. B, Postoperative computed tomography image at 7 days showed no endoleak or free air around the stent graft.

duodenum with a gastrojejunal anastomosis. Intravenous antibiotics (piperacillin/tazobactam, 13.5 g/day) were given preoperatively. The preoperative blood bacterial culture was negative. Enteral nutrition was started on postoperative day 3. Antibiotics were continued until postoperative day 6. Physical examination on postoperative day 6 did not indicate infection, and the laboratory findings had improved (white blood cell count, 5,190/ μ L; CRP, 4.18 mg/dL). In addition, CT on postoperative day 7 showed no endoleak, extravasation, or abdominal abscesses (Fig. 3B). The patient was transferred to the previous hospital to continue internal therapy on postoperative day 8; however, the patient died suddenly 2 months after the first operation with recurrent inflammation (white blood cell count, 13,000/ μ L; CRP, 6.11 mg/dL) and severe anemia (hemoglobin level, 3.3 g/dL).

DISCUSSION

THE CLINICAL COURSE of ADF is typically characterized by “herald bleeding,” which is spontaneous brief hematemesis followed by massive bleeding from hours to weeks later.⁵ In our patient, the first symptom was hematemesis, and enhanced CT showed an AAA rupture and large hematoma in the stomach and duodenum. In addition, intraoperative endoscopy showed no other cause of the upper gastrointestinal bleeding. Therefore, we were able to diagnose ADF clinically.

Primary ADF is caused mainly by AAA rupture.¹ Proposed theories for the formation of primary ADF are direct wear and inflammatory destruction of the aortic wall.¹ Secondary ADF is known as a long-term complication of abdominal surgical procedures, especially for AAA.^{6,7} Other causes of secondary ADF are variable and include trauma, infection, or malignancy.⁷ However, studies on the relationship between ADF and a history of gastrectomy do not generally recognize gastrectomy as being a risk factor for secondary ADF.^{8,9}

In our case, we only found non-specific inflammation and did not recognize any evidence of recurrent or new malignancy. In addition, the base of the aneurysm existed on the infrarenal aorta, which was far from the surgical field of the previous gastrectomy. Therefore, our case could be a rare case of primary ADF, despite the previous history of laparotomy. With respect to the surgical treatment of ADF, conventional open surgery for aneurysm repair and duodenal perforation closure is radical and highly invasive, with a high 30-day mortality rate of approximately 40%.^{2,6} Therefore, EVAR has been attempted for rapid control of bleeding and less invasiveness.^{2,3} In particular, initial EVAR is expected to be a “bridge therapy,” followed by elective open surgical repair.^{2,10} Also, surgical isolation of the duodenum with duodenojejunal anastomosis is expected to become an adjunctive radical treatment after EVAR.^{11,12} In our case, however, previous gastrectomy and Billroth I reconstruction, which caused broad dense adhesions around the aneurysm

and duodenum, precluded any additional open repairs. Poor preoperative general status because of alcoholism and malnutrition also made our situation difficult. Alternatively, therefore, we undertook intestinal tract tube decompression, which is an effective treatment for duodenal injury.¹³

In the present case, the combination of EVAR and intestinal tract decompression prevented perioperative death within 30 days, and the patient survived for 2 months. The patient acutely died with the recurrence of inflammation and severe anemia and could not be transferred to our hospital in time. The synchronous recurrent inflammation and severe anemia indicated the occurrence of rebleeding of ADF possibly due to stent graft infection. In general, however, a 2-month survival period is adequate for preparation of a subsequent elective surgery. If the patient's infirm status had improved, we would have been able to undertake an additional surgery for duodenal repair or isolation of the duodenal perforated lesion with gastrojejunal anastomosis. Therefore, the combination of EVAR and intestinal tract decompression may contribute to extension of the rebleeding interval and might be an optimal temporary treatment for high-risk patients with ADF. At the same time, for radical treatment of ADF, surgical isolation or repair of duodenal perforated lesions seemed essential. Hence, more investigations are required to clarify the effective and safe treatment for complicated high-risk ADF patients, especially with a history of laparotomy.

CONCLUSION

WE REPORTED A rare case of primary ADF complicated by a history of distal gastrectomy and Billroth I reconstruction. Primary EVAR was successful; however, further studies are needed to improve the long-term outcomes in high-risk ADF patients.

CONFLICT OF INTEREST

NONE.

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