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Case report

A case of aortoduodenal fistula presenting with postoperative lymphatic leakage

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ABSTRACT

Introduction and importance: Secondary aortoduodenal fistula (sADF) is a complication of prosthetic graft replacement of the abdominal aorta which often follows a fatal course. This report details our experience with a case of lymphatic fistula that developed after sADF repair. The fistula was refractory to conservative treatment but ultimately responded to lipiodol lymphangiography.

Case presentation: A 75-year-old man had undergone prosthetic graft replacement to treat an abdominal aortic aneurysm in 2012 and a thoracic aortic aneurysm in 2015. Upper gastrointestinal endoscopy was performed in 2020, and examination for worsening anemia revealed that the abdominal aortic graft had eroded into the horizontal duodenum. The patient was treated with prosthetic graft replacement and duodenectomy. A refractory lymphatic fistula was noted after surgery, which made ascites accumulation difficult to control, but the patient's condition rapidly improved following therapeutic lymphangiography.

Clinical discussion: Surgery is the first-line therapy for sADF, but clinicians must stay vigilant for infection recurrence and aortoenteric fistulae after a repair, and this requires patient-specific postoperative management. Our modifications, intended to minimize contamination of the operative field in the present case, also facilitated our ability to subsequently treat a refractory lymphatic fistula, which is a rare postoperative complication of the procedure.

Conclusion: Procedural modifications to sADF repair aimed at minimizing perioperative contamination are crucial for preventing infection recurrence. Given the extent of invasion, the surgery can cause various postoperative complications, requiring individualized strategies for management and treatment. Therapeutic lymphangiography is one such approach, which holds promise as a first-line treatment for refractory lymphatic fistula.

1. Introduction and importance

Secondary aortoduodenal fistula (sADF) is a rare postoperative complication of prosthetic graft replacement of the abdominal aorta, occurring in 0.4–4% of cases. Many sADF cases are fatal, being exacerbated by gastrointestinal (GI) bleeding and/or graft infection associated with the defect [1]. Similarly, lymphatic fistula is a relatively rare postoperative complication of abdominal surgery, with poor outcomes if a major lymphatic vessel damaged, leading to impairment in spontaneous closure. Such treatment-resistant fistulae are prone to major chyle loss, causing circulatory insufficiency, malnutrition, and hypoproteinemia [2]. This report details our experience with a case of lymphatic fistula that was refractory to conservative treatment, which developed after sADF repair and ultimately responded to lipiodol lymphangiography (LAG).

This work has been reported in line with the SCARE criteria [3].

2. Case presentation

Our patient was a 75-year-old man (performance status 0) with

Abbreviations: ADF, aortoduodenal fistula; CT, computed tomography; GI, gastrointestinal; LAG, lymphangiography; NBCA, *n*-butyl-2-cyanoacrylate; sADF, secondary aortoduodenal fistula.

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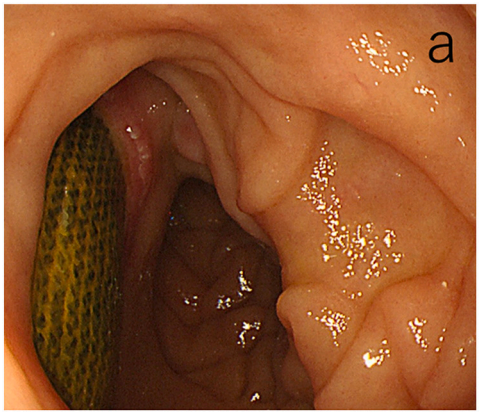

Fig. 1. a. Upper GI endoscopy showed graft wall erosion at the posterior wall of the horizontal duodenum.
b, c. CT showed slight air entrapment and enhanced soft tissue density in the graft vicinity.

anemia. He had a history of rheumatoid arthritis and lower extremity arteriosclerosis obliterans. The patient had undergone prosthetic graft replacement for abdominal and thoracic aortic aneurysm in 2012 and 2015, respectively. In 2017, he underwent laparoscopic right hemicolectomy for ascending colon cancer. In 2018, a soft tissue shadow was detected near the abdominal aortic graft on follow-up computed tomography (CT), which we conservatively monitored. In 2020, prompted by worsening anemia, we performed upper GI endoscopy and found that the graft had eroded into the horizontal duodenum, necessitating surgical repair.

The patient has signs of anemia (hemoglobin, 6.8 g/dL), leukopenia

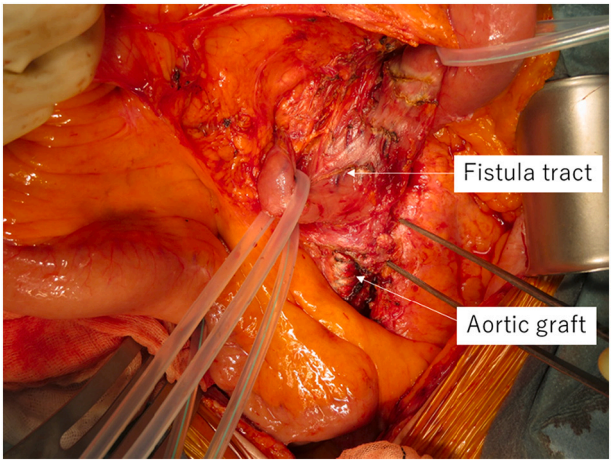

Fig. 2. The surgery findings showed the fistula tract was identified along a segment of the duodenum.

(white blood cell, 2670/μg), and C-reactive protein elevation (11.09 mg/dL). Procalcitonin elevation was also noted but was mild (0.71 ng/dL). Upper GI endoscopy showed graft wall erosion at the posterior wall of the horizontal duodenum. No active bleeding from the fistula tract was observed. CT showed slight air entrapment and enhanced soft tissue density in the graft vicinity (Fig. 1).

Subsequently, prosthetic graft replacement, duodenal segmental resection, and omental flap transposition were performed. The fistula tract was identified along a segment of the duodenum, spanning its third to fourth parts, which firmly adhered to the aorta. To minimize contamination of the operative field, we partially resected the duodenum without opening the fistula tract. First, the Kocher maneuver was performed, and the third part of the duodenum was resected about midway along its length using an automated suturing device. Next, the jejunum was resected using an automated anastomotic device. Duodenal dissection (mobilization) was continued in the oral direction and completed with the resected intestine still attached to the aorta. The duodenum was reconstructed by elevating the jejunum to the right of the transverse mesocolon and joining it to the remaining duodenum by side-to-side anastomosis. After reconstruction, en bloc resection was performed to remove the original aortic wall, prosthetic vessel, stent graft, and adherent duodenum. The defect was repaired by prosthetic graft replacement followed by omental plombage (Figs. 2-4).

On the sixth postoperative day (D6), the patient started a liquid diet and his peritoneal drain was removed. On D8, the fluid oozing from the drainage site had become cloudy and was diagnosed as chyle leakage based on biochemical findings (drain fluid triglycerides: 388 mg/dL). Thus, the patient was switched to a fat-restricted diet. The drainage site eventually closed naturally, and the patient gained weight. Large-volume ascites was noted on CT on D16. Despite attempts to remedy it via peritoneal drainage, diuretic administration, and diet therapy, it was difficult to control, necessitating therapeutic LAG on D26 (Fig. 5a, b).

During the therapeutic LAG, lipiodol was injected into the left inguinal lymph nodes, but the flow was stagnated. When injected into the right inguinal lymph nodes, lipiodol perfused the lower circulation within a relatively short time; lipiodol leakage was observed anterior to the L2/3 vertebrae (Fig. 5c).

Seven days post-LAG, a significant reduction in ascitic volume was confirmed on CT. On D42, the patient was transferred to another hospital for rehabilitation. His subsequent course was normal, without

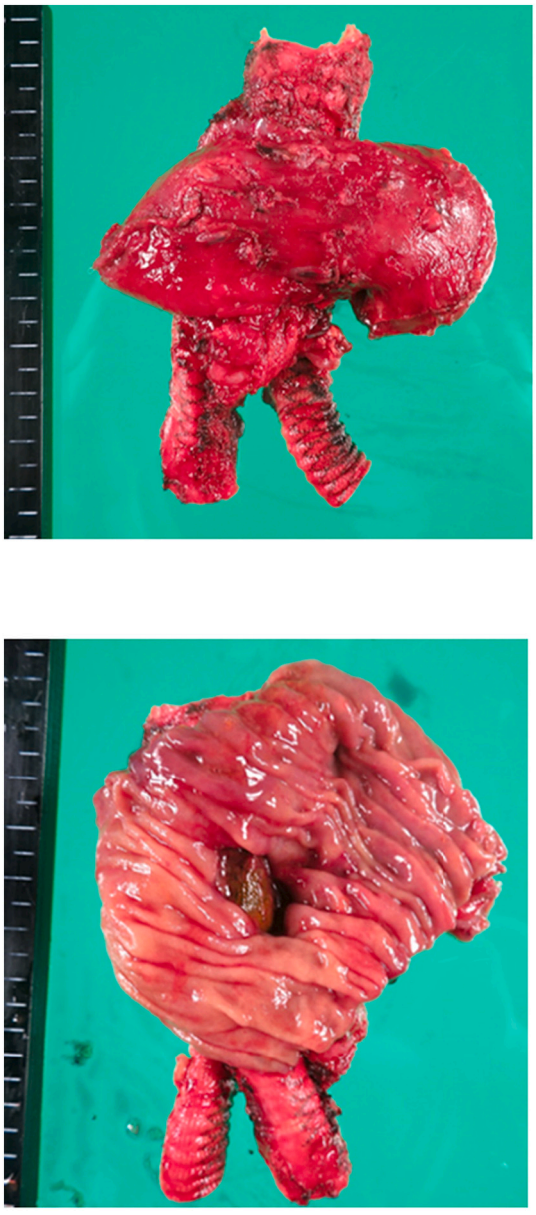

Fig. 3. a, b. The aortic wall, artificial blood vessel, stent graft, and duodenum are stuck together in a group and resected.

infection recurrence. The patient died of another illness approximately 1 year later (Fig. 5d, e).

3. Discussion

sADF, an abnormal connection between the aorta and duodenum that occurs after prosthetic graft replacement, typically develops several years postoperatively. Although GI bleeding is a core symptom of primary ADF, it only appears in 46.2% of sADF. sADF more frequently presents with fever or chills (61.5%) and abdominal pain (53.8%). With fistula formation, infection usually occurs because intestinal fluids leak and severely contaminate the perigraft environment. This is one reason prosthetic graft infection and other complications reduce patients' survival after aortic reconstruction surgery [4].

Bleeding control, infection control, and revascularization are key goals of sADF repair; considering the aorta and duodenum separately is crucial when planning surgery [5,6]. Non-anatomical revascularization followed by total explantation of the prosthetic graft has conventionally been the recommended protocol for the aorta [7]. However, postoperative complications such as aortic rupture and graft occlusion result in non-negligible mortality rates (23–44%) in the early postoperative period, with rarely satisfactory outcomes. Recently, a few reports showed that mortality and complication rates can be improved by selecting anatomical revascularization [8,9]. However, Batt et al. compared sADF treated with non-anatomical versus anatomical revascularization and reported no superiority with either technique [10], although in Japan, satisfactory outcomes for anatomical revascularization were reported by Hashimoto et al. [8]

Chronic infection and inflammation are suspected to have caused fistula formation in the present case. Our patient developed sADF approximately 8 years after prosthetic graft replacement for an abdominal aortic aneurysm, but soft tissue shadows were seen near the graft as early as 3 years after the procedure. Removing the infection focus was thus central to our decision to perform partial duodenal resection; similarly, our reconstruction protocol—raising the jejunum to the right transverse mesocolon and anastomosing side-to-side with the remaining duodenum—was selected to avoid perigraft contamination if suture failure occurred. Knowing that opening the fistula tract could contaminate the perigraft environment, we instead resected the fistulous segment at the oral and anal sides using an automated suturing device and excised it en bloc during aortic replacement. We believe our protocol is well-suited for sADF repair, as resection by automated suturing devices limits intestinal fluid leakage and obviates the need to cut open the fistula tract, minimizing contamination [11].

Lymphatic fistula is another relatively rare postoperative complication of abdominal surgeries, with an estimated incidence of 0.9% in colon cancer surgery, 0.3–0.4% for D2 lymphadenectomy in gastric cancer, and 0.8–3.4% in hepato-pancreato-biliary surgery [2,12]. The lymphatic system reabsorbs fluids and proteins from the interstitial space and excretes excess tissue fluids. From the intraabdominal organs, lymph travels through the intestinal lymph trunk to the cisterna chyli, located posteromedial to the aorta at the level of the L1/2 vertebrae, before passing into the thoracic duct via the aortic hiatus. When a major lymphatic vessel is damaged during abdominal surgery, it may fail to naturally close and cause poor treatment response. Lymphatic fistulae are typically diagnosed based on triglyceride levels of $\geq$200 mg/dL in ascites [13]. Possibly, our patient's lymphatic system was damaged in the surgery before prosthetic graft replacement during attempts to remove contaminated tissue. Alternatively, a previous accident may be to blame; he had undergone prosthetic aortic replacement and a procedure for ascending colon cancer which involved lymphadenectomy. Although the exact relationship is unclear, such frequent retroperitoneal operations could have adversely affected the lymphatic system.

Low-fat, high-protein dietary management is the first-line treatment for lymphatic fistula. Other conservative therapies include octreotide administration, fasting and total parenteral nutrition, minocycline/OK-432 administration, and fibrin sealant application. Surgical therapies include the ligation of damaged lymph vessels and peritoneovenous shunt placement [14]. Cell-free concentrated ascites reinfusion therapy has also been reported to be useful [15]. In a recent paper, LAG using lipiodol was effective in closing refractory lymphatic fistulae that had

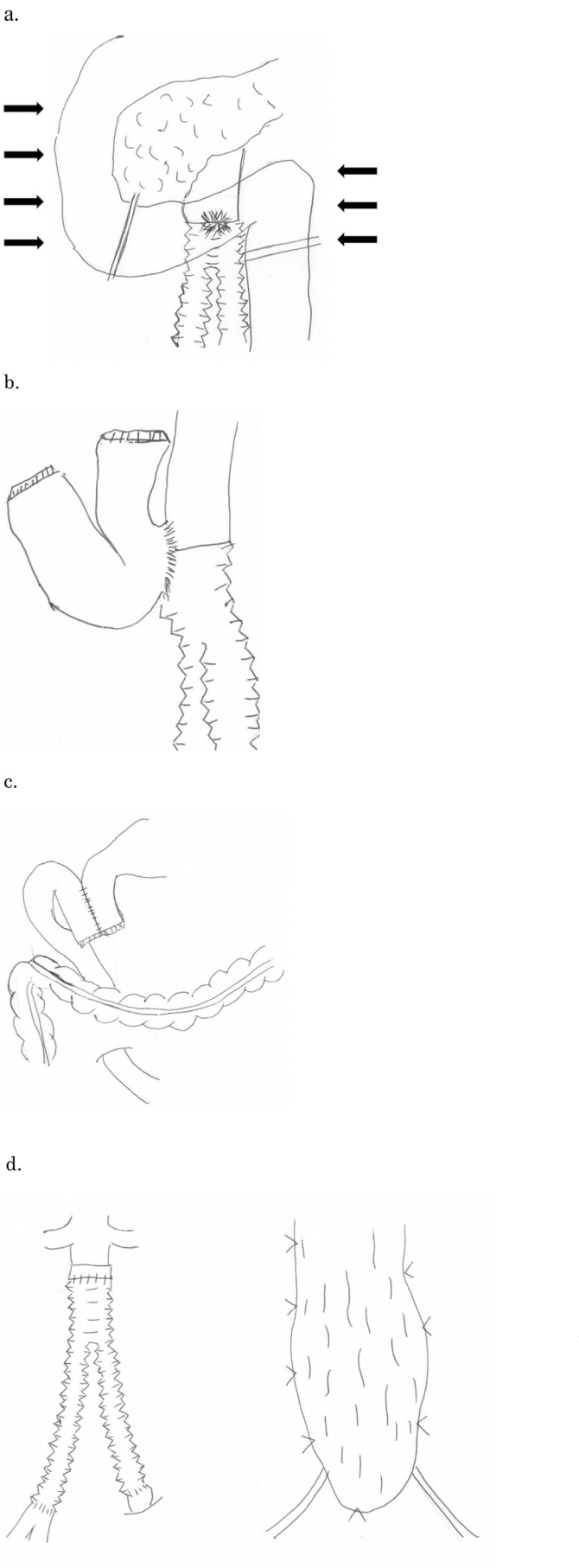

Fig. 4. Surgical schema.
a. Abrasion, mobilization of the duodenum, and separation of the actinal side and anal side are performed with automatic suture instruments.
b. The syringeal part remains attached to the duodenum from the aortic wall without leaving it open.
c. The side-to-side anastomosis of the duodenal descending limb and jejunum are performed via a posterior colon route.
d. Artificial blood vessel reimplantation and omental flap transposition are performed.

developed after surgery for esophageal cancer [16]. Despite its high viscosity, lipiodol is notable for its ability to reach even the lymphatic capillaries of injured regions; it is hypothesized to heal lymphatic fistulae by triggering inflammation and promoting granulation at the leakage site. Moreover, performing LAG using a combination of lipiodol and the vascular embolic agent *n*-butyl-2-cyanoacrylate (NBCA) was reported to effectively treat cases not markedly improved by lipiodol alone (although this use of NBCA is off-label) [17]. Therapeutic LAG can be performed relatively safely, even in older adults and convalescent surgery patients. Although its side effects include pulmonary embolism, fever, and injection site edema, they occur in less than 5% of cases [18]. We performed therapeutic LAG in the present case hoping it would enable us identify and effectively treat the leakage site of a refractory lymphatic fistula, which had not responded to conservative treatments such as dietary management, pharmacotherapy, or ascites drainage. The significant reduction in ascitic volume observed on CT after 7 days suggests that lipiodol LAG enabled the lymphatic fistula to heal. Based on the point of contrast extravasation, we suspect the damage occurred near the cisterna chyli, a structure from which chylous leakage would naturally be considerable. However, even in such a challenging case, therapeutic LAG proved effective.

4. Conclusion

Procedural modifications to sADF repair aimed at minimizing perioperative contamination are crucial for preventing infection recurrence. Given the invasiveness, various postoperative complications require individualized management and treatment. Therapeutic LAG is one such approach, which holds promise as a first-line treatment for refractory lymphatic fistula.

Sources of funding

There are no funding sources for the study.

Ethical approval

The ethical approval was given from our institution.

Consent

Written informed consent was obtained from the patient's next of kin for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Takao Tsuneki: Operating Surgeon and writing the paper.
Yasuhiro Yuasa: Assistant Surgeon, Supervisor and writing the paper.
Takeshi Nishino: Writing the paper.
Atsusi Tomibayashi: Writing the paper.
Tatsuo Motoki: Writing the paper.
Yoshiaki Fukumura: Assistant Surgeon and Writing the paper.

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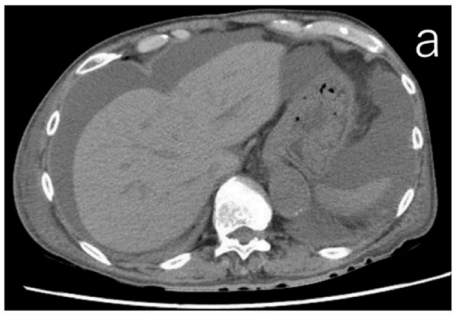

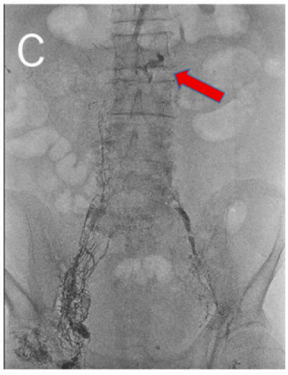

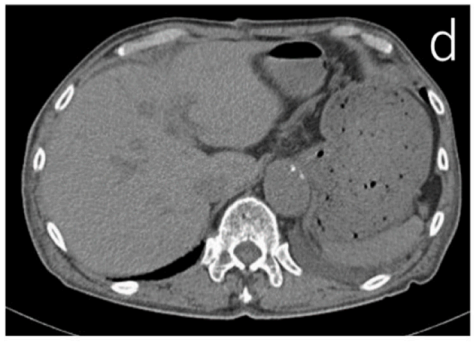

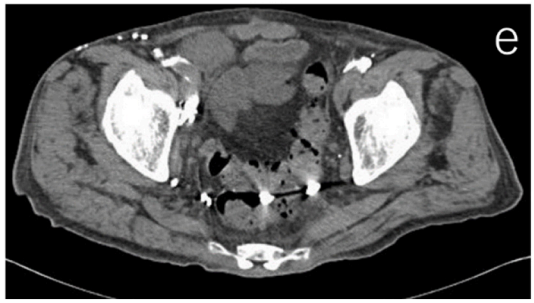

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Fig. 5. a, b. Large-volume ascites was noted on CT on D16.
c. During the therapeutic LAG, lipiodol was injected into the right inguinal lymph nodes, lipiodol perfused the lower circulation within a relatively short time; lipiodol leakage was observed anterior to the L2/3 vertebrae.
d, e. Seven days post-LAG, a significant reduction in ascitic volume was confirmed on CT.

Research registration

This is not the first in man study.

Guarantor

Takao Tsuneki.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Declaration of competing interest

No conflict of interest.

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