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Primary aortoduodenal fistula: A case report and review of literature

Tohru Ishimine^{a,*}, Toshiho Tengan^a, Hiroshi Yasumoto^a, Akio Nakasu^a, Hidemitsu Mototake^a, Yuya Miura^a, Kyohei Kawasaki^a, Takashi Kato^b^a Department of Cardiovascular Surgery, Okinawa Prefectural Chubu Hospital, Japan^b Department of General Surgery, Okinawa Prefectural Chubu Hospital, Japan

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ABSTRACT

BACKGROUND: Primary aortoduodenal fistula (PADF) is an abnormal connection between the aorta and the duodenum and is a life-threatening condition. It is a very rare cause of gastrointestinal bleeding, which often leads to delay in its diagnosis. Prompt diagnosis and surgical treatment are crucial to improve the outcome of patients with PADF.

PRESENTATION OF CASE: An 82-year-old man with a history of untreated abdominal aortic aneurysm (AAA) presented to the emergency department with hematemesis. Computed tomography (CT) revealed an AAA with air within the thrombus wall and disruption of the fat layer between the AAA and duodenum, indicating PADF. Emergent surgery, in situ aortic reconstruction using a Dacron graft, and omental coverage were performed. Although the patient needed another surgery for postoperative chylous ascites, he made good recovery and was discharged 86 days after initial surgery.

DISCUSSION: In our case, the patient presented with hematemesis and a pulsatile abdominal mass on physical examination and had a history of untreated AAA, which helped in prompt diagnosis of PADF. CT findings suggesting PADF include disappearance of the fat plane between the aneurysm and duodenum, air in the retroperitoneum or within the aortic wall, and contrast enhancement within the duodenum. The recommended surgical approach for PADF consists of aortic reconstruction (in situ aortic reconstruction or extra-anatomical bypass) and duodenal repair.

CONCLUSION: Our report affirms that CT and open surgery are effective diagnostic and treatment options, respectively, for PADFs.

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1. Introduction

Aortoduodenal fistula (ADF) is an abnormal connection between the aorta and the duodenum and a rare and life-threatening condition. This disease is divided into two types: primary and secondary ADF. The incidence of primary ADF (PADF) is 10 times as less as that of secondary ADF, which results from previous aortic prosthetic reconstruction [1]. PADF usually occurs when a large abdominal aortic aneurysm (AAA) erodes into the duodenum. Given the rarity and non-specificity of abdominal signs and symptoms, the diagnosis of PADF is often delayed, which in turn results in high morbidity and mortality. We present a case of a PADF between the infrarenal AAA and the third part of the duodenum, which was promptly diagnosed and successfully managed with appropriate

surgical treatment. This case report has been written in line with the SCARE criteria [2].

2. Presentation of case

An 82-year-old man with a history of untreated AAA, hypertension, and gout was admitted to our emergency department with hematemesis. He had a blood pressure of 98/65 mm Hg, heart rate of 82 beats/min, body temperature of 35.8 °C, and respiratory rate of 23 breaths/min. He had a palpable pulsatile abdominal mass without tenderness. Laboratory investigations revealed anemia (hemoglobin, 9.5 g/dL), leukocytosis (white blood cell count, 14,000/mm³), and renal insufficiency (serum creatinine level, 1.83 mg/dL). Computed tomography (CT) revealed an infrarenal fusiform AAA with a diameter of 7 cm, which compressed the third part of the duodenum (Fig. 1). Although no contrast was seen within the duodenum, CT also revealed disruption of the fat layer between the AAA and duodenum and air within the thrombus and the aneurysmal wall. A diagnosis of PADF was made based on the clinical course and CT findings.

Emergent laparotomy revealed dense adhesion between AAA and the third part of the duodenum, which was considered to be

Abbreviations: AAA, abdominal aortic aneurysm; ADF, aortoduodenal fistula; CT, computed tomography; PADF, primary ADF.

* Corresponding author at: Department of Cardiovascular Surgery, Okinawa Prefectural Chubu Hospital, 281, Miyazato, Uruma-shi, Okinawa, 904-2293, Japan.

E-mail address: ishimine.toru@hosp.pref.okinawa.jp (T. Ishimine).

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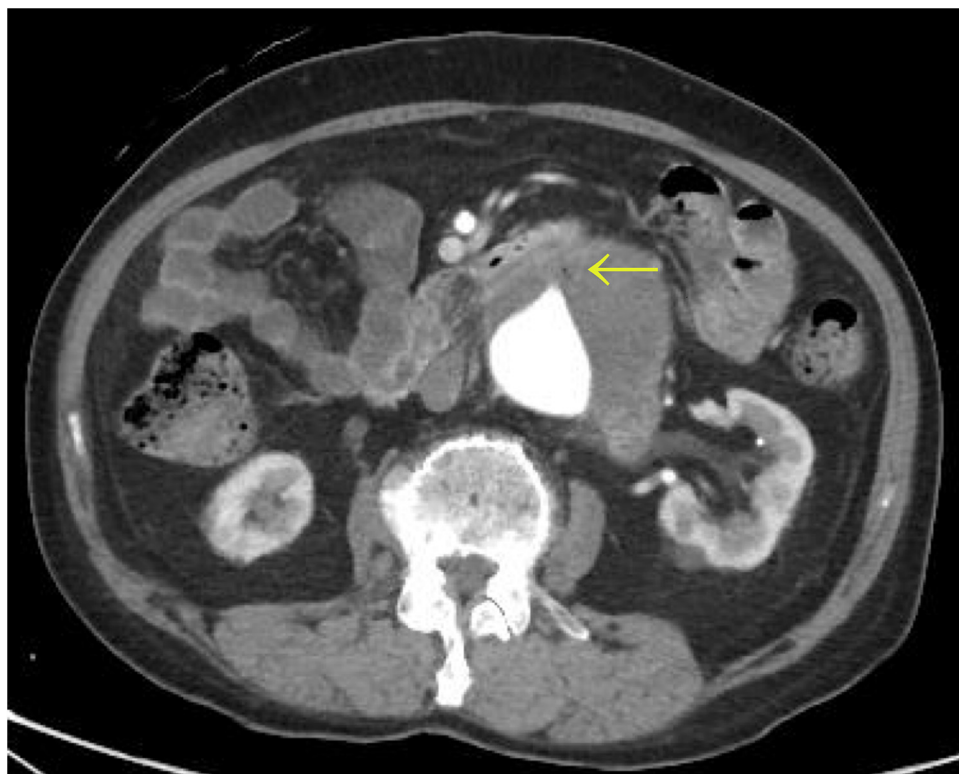


Fig. 1. Axial contrast-enhanced computed tomography image of the abdomen showing a large abdominal aortic aneurysm attached to the third part of the duodenum. Air within the wall thrombus of the aneurysm can be seen (arrow).

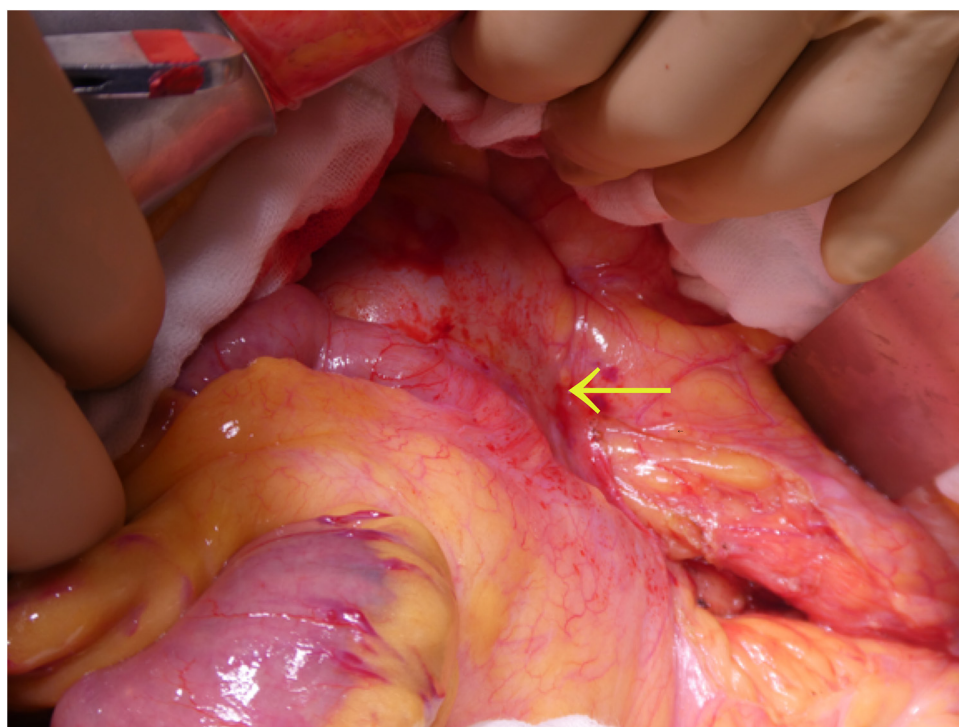


Fig. 2. Intraoperative findings of the primary aortoduodenal fistula (PADF). The PADF is located between the posterior wall of the third part of the duodenum and the anterior wall of abdominal aortic aneurysm (arrow).

PADF (Fig. 2). Intraoperative gastroduodenal endoscopy revealed an extrinsic pulsatile mass with a central ulceration in the third part of the duodenum (Fig. 3). The involved part of the duodenum was resected using a linear stapler to avoid spillage of bowel con-

tents and was removed from the wall of AAA after aortic clamping. As there were no findings of contamination and infection, in situ aortic reconstruction with a 20 × 10 mm bifurcated Dacron graft

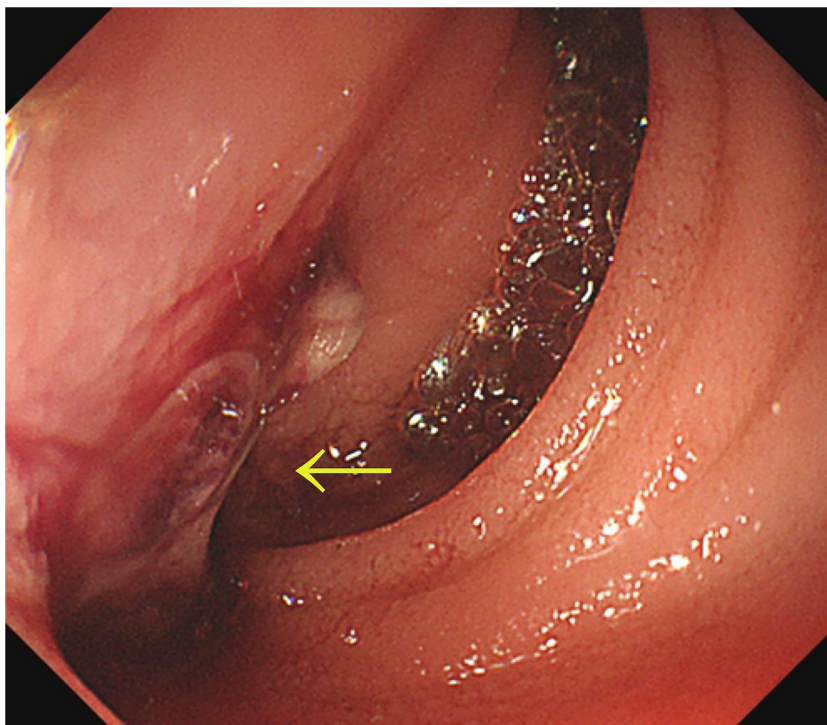


Fig. 3. Intraoperative gastroduodenal endoscopy findings. Endoscopy shows a pulsating bulge with an adjacent ulcer (arrow) in the third portion of the duodenum.

(J Graft SHIELD NEO) and omental coverage were performed. The duodenum was anastomosed in a side-to-side fashion.

Re-exploration and suture ligation of leaking lymph channels for chylous ascites resistant to conservative treatment were performed on postoperative day 48. The patient was discharged on postoperative day 86. At 8 months follow-up, he was alive and healthy.

3. Discussion

PADF is an abnormal connection between the aorta and the duodenum and a very rare and life-threatening condition. PADF is believed to result mostly from direct wear and inflammatory destruction of an aortic wall [3]. The most common cause of PADF is AAA. Gad reported that 73% of PADFs were from atherosclerotic aneurysms and 26% from traumatic or mycotic aneurysms [4]. Other possible causes are infection, foreign body, radiotherapy, and tumors [5]. Because of anatomical proximity, the segment most commonly involved is the third part of the duodenum (two-thirds of cases), as in our case, followed by the fourth part (one-third of cases) [6]. The classical triad of PADF is upper gastrointestinal bleeding (64%), abdominal pain (32%), and a pulsatile abdominal mass (25%) [7]. However, the classical triad is concomitantly present in only 11% of cases [6], which makes diagnosis of PADF difficult. Other symptoms may include back pain, fever, sepsis, and melena. The first upper gastrointestinal bleeding known as herald bleeding is usually self-limited and observed in about 30% of patients with PADF [7]. Herald bleeding is thought to be transient bleeding from a small fistula stopped by blood clot formation; massive bleeding ensues after the clot is removed. The secondary massive hemorrhage occurs within the next 6 h in one-third of the cases [1].

Untreated PADF has a high mortality rate of approximately 100%, making the difficulty in diagnosing PADF a significant challenge [8]. This necessitates suspecting PADF in all patients with upper gastrointestinal bleeding, particularly in patients with a history of

AAA. In our case, the patient presented with hematemesis and a pulsatile abdominal mass on physical examination and had a history of untreated AAA, which helped in prompt diagnosis of PADF, and emergency treatment was provided.

CT scanning, endoscopy, and angiography are commonly used in the diagnosis of PADF. Of these, the most useful diagnostic modality is CT with intravenous contrast, which has a specificity of 85%–100% and a sensitivity of 50%–94% [9]. Moreover, CT is less invasive and poses no danger of dislodging the thrombus compared to endoscopy or angiography. CT findings strongly suggesting PADF include disappearance of the fat plane between the aneurysm and duodenum, air in the retroperitoneum or within the aortic wall, and contrast enhancement within the duodenum [6].

Endoscopy should be performed only if the patient is hemodynamically stable. The findings of an ulcer or erosion adjacent to a blood clot with an extrinsic pulsatile mass in the duodenum are highly indicative of PADF, as demonstrated in our case [8]. In contrast, the diagnostic rate of PADF using endoscopy is low (25%); this could be attributed to the difficulty in visualization of a fistula distal to the third part because of the acute angle between the third and fourth parts of the duodenum [10]. Therefore, a negative endoscopy finding does not exclude the possibility of a PADF. Endoscopy is useful in ruling out other causes of upper gastrointestinal bleeding, such as gastroduodenal ulcers and varices. In our case, although CT and intraoperative findings indicated PADF, intraoperative endoscopy was performed to rule out other diseases and confirm the diagnosis of PADF.

Owing to the recent advancements in CT imaging, its superior imaging resolution, and less invasiveness, use of angiography in the diagnosis of PADF is currently limited.

The recommended surgical approach for PADF consists of aortic reconstruction (in situ aortic reconstruction or extra-anatomical bypass) and duodenal repair. In case of PADF with no or mild contamination, as in our case, an in situ aortic reconstruction using a Dacron or polytetrafluoroethylene graft with omental coverage is preferred [11]. Omental coverage of a prosthetic graft is criti-

cal to prevent infection [12]. Rodrigues dos Santos et al. in their multivariate analysis of 791 patients with primary and secondary aortoduodenal fistula (ADF) reported that omental coverage and in situ aortic reconstruction are independent predictors of survival [12]. The survival rate among patients undergoing in situ aortic reconstruction is 61%–77% [13]. By contrast, in case of a primary mycotic ADF or PADF with massive contamination, an extra-anatomical bypass graft with extensive debridement is feasible [14]. However, this surgical procedure is related to low survival rates of 40%–60% because of hemorrhage from the suture line of the aorta and stump blow-out [1,15].

Regardless of evidence of infection, empirical antibiotics should be initiated to treat the most probable microbial causes. During surgery, tissue specimens for culture must be collected to determine the appropriate antibiotic therapy. Even if cultures are negative, the administration of antibiotics for 7–10 days is recommended [6]. If cultures are positive, antibiotics should be selected based on the results of sensitivity test and administered for 4–6 weeks after surgery [7].

Recently, successful endovascular treatment of PADF has been reported as another option for unstable patients who are not suitable for open surgery [16]. However, placement of an endovascular stent graft, which is a foreign body, within the aorta connected to the duodenum, is not favorable owing to the high risk of infection. Keunen et al. reported that removal of endovascular stent was required for a patient with PADF several months after successful initial endovascular treatment [17]. Although endovascular treatment is useful to control bleeding immediately, performing definitive surgery (open aortic reconstruction and duodenal repair) following endovascular repair is desirable to obtain good long-term outcome.

Based on our case and review of literature, we recommend CT as a highly effective diagnostic modality for PADF and open surgical treatment as the preferred treatment.

Conflicts of interest

All authors declare no conflict of interest.

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Ethical approval

This case report was approved by the Ethics Committee of Okinawa Prefectural Chubu Hospital.

Consent

Written informed consent was obtained from the patient 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

Tohru Ishimine: Study design, data collection, writing the paper.
Toshiho Tengan: Review manuscript.
Hiroshi Yasumoto: Review manuscript.

Akio Nakasu: Data collection, obtain images.
Hidemitsu Mototake: review manuscript.
Yuya Miura: Data collection, obtain images.
Kyohei Kawasaki: Data collection, obtain images.
Takashi Kato: Data collection, obtain images.

Registration of research studies

Not applicable for case report.

Guarantor

Tohru Ishimine.

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