Radiology Case Reports 18 (2023) 1556–1559

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr

Case Report

A case report of primary aortoduodenal fistula: A forgotten cause of gastrointestinal bleeding ☆,☆☆

Pham Dang Hai, MD[a,*], *Pham Yen Nhi, MD*[a], *Nguyen Thu Huyen, MD*[a], *Nguyen Huu Thanh, MD*[b]

[a] *Medical Intensive Care Unit, 108 Military Central Hospital, Vietnam*
[b] *College of Health Sciences, VinUniversity, Vietnam*

ARTICLE INFO

Article history:
Received 14 December 2022
Accepted 27 December 2022

Keywords:
Primary aortoduodenal fistula
Gastrointestinal bleeding
Surgery
Endovascular aortic repair

ABSTRACT

Aortoenteric fistula is one of the uncommon emergencies and is challenging to navigate for diagnostic testing. Here, we present a clinical case of an aortoduodenal fistula with primary etiology. A 73-year-old female patient with a history of hypertension was admitted to the hospital because of a 1-day history of melena. Ultrasound showed an abdominal aortic aneurysm sized (33 × 46) mm and a hematoma on the wall of the aorta. The patient underwent a gastrointestinal endoscopy with no bleeding point detected. However, the patient suddenly fell into a hemorrhagic shock on day 3 of admission. We rapidly performed fluid resuscitation, blood transfusion, a second gastrointestinal endoscopy, and a computed tomography scan of the abdomen with contrast injection that revealed a fistula from the abdominal aorta into the second segment of the duodenum. The patient was indicated for urgent endovascular aortic repair. Although this technique was successful with 3 abdominal aortic stents, the patient died due to multiorgan failure. Delayed diagnosis is the root cause of primary aortoduodenal fistula treatment failure, so it is important for clinicians to keep aortoduodenal fistula in mind as a possible cause of gastrointestinal bleeding in any patient.

Introduction

Aortoduodenal fistula (ADF) is a rare condition but life-threatening, even fatal, if not diagnosed and treated promptly. Aortoenteric fistula (AEF) is defined as an abnormal connection between the aorta and the gastrointestinal tract. AEF is classified into 2 types: Primary aortoenteric fistula (PAEF) and Secondary aortoenteric fistula (SAEF). This article only addresses the primary abnormal communication between the

Abbreviations: ADF, Aortoduodenal fistula; AEF, Aortoenteric fistula; PAEF, Primary aortoenteric fistula; SAEF, Secondary aortoenteric fistula; PADF, Primary aortoduodenal fistula; AAA, Abdominal aortic aneurysm; EVAR, Endovascular aortic repair; GI, Gastrointestinal; EGD, Esophagogastroduodenoscopy.

☆ Funding: This research did not receive any grant funding.
☆☆ Competing Interests: The authors declare that there are no conflicts of interest.

* Corresponding author.
E-mail address: bsphamdanghai@gmail.com (P.D. Hai).

https://doi.org/10.1016/j.radcr.2022.12.064

abdominal aorta and the duodenum. More than 70% of primary aortoduodenal fistula (PADF) involves the duodenum, most located in the third and fourth parts [1]. It is often manifested as massive, acute, difficult-to-control gastrointestinal bleeding that leads to shock due to blood loss and fetal. Early diagnosis of this dangerous condition is not easy, especially for PADF, where up to two-thirds of PADF diagnoses are made in the operating room [2]. Abdominal aortic aneurysm (AAA) is considered the most common risk factor of PADF. The choice of treatment method is also controversial, but without treatment, PADF is 80%-100% fatal and has a perioperative mortality rate of 18%-63% [3].

Case report

A 73-year-old woman with a history of hypertension was admitted to our emergency department after a 1-day episode of melena. The initial evaluation showed blood pressure of 110/70 mmHg, a heart rate of 108 beats/min, a body temperature of 36.8 °C, and a respiratory rate of 18 breaths/min. She had no pain complaint and no abdominal mass discovered by physical examination. Laboratory investigations revealed red blood cell of 2.51 × 10^{12}/L, hemoglobin of 8 g/dL, white blood cell count of 21.75 G/L with 89% neutrophils, prothrombin of 80%, urea of 14 mmol/L, creatinine of 127 μmol/L. Abdominal ultrasound showed an infrarenal abdominal aortic aneurysm (AAA) in size of 33 × 46 mm with a partially thrombosed 60 × 16 mm at the anterior side; the rest of the analytical findings were unremarkable.

After hours of presentation, esophagogastroduodenoscopy (EGD) was performed, and observed a small curvilinear ulcer with red scars and atrophic gastritis; the colonoscopy found no bleeding point. The patient was diagnosed with moderate upper gastrointestinal (GI) bleeding may be due to gastric ulcer, and was resuscitated with intravenous fluid administration, 350 mL of red blood cells and 350 mL of platelets transfusion, continuous proton pump inhibitor infusion, and fasting. During one and a half days in the Gastrointestinal Department, her hemodynamics remained temporarily stable, and no more hematochezia. Suddenly, on the third day of admission, the patient complained of upper abdominal pain, dizziness, hypotension, systolic pressure dropped rapidly to approximately 60 mmHg, heart rate of 120 beats/min, SpO_2 of 90% in room air condition, and gastric drainage with much bright red blood. She was immediately transferred to the Intensive Care Unit in a state of coma and required a second endoscopy after aggressive attempts at resuscitation with normal saline plus 700 mL of packedred blood cell, 500 mL of fresh plasma, vasopressor, and tracheal intubation.

Nonetheless, the stomach was filled with blood clots at the endoscopy, which provided inadequate gastric visualization. An abdominal computed tomography (CT) scan was immediately indicated and showed an infrarenal fusiform AAA with a size of 75 × 53 mm and 25 mm thick anterior wall thrombus, ruptured and leaked into the second part of the duodenum confirming the diagnosis of hemorrhage complicated by aortoduodenal fistula condition (Fig. 1). The patient underwent urgent endovascular aortic repair (EVAR) involves the placement of 3 stent graft components delivered via the femoral arteries and plugging the ruptured aneurysm with histoacryl, which was effectuated successfully after 2 hours of intervention duration with no more presence of contrast leak into the gastrointestinal tract (Fig. 2). However, the patient died after 10 hours of intervention due to multiple organ failures complicated by reversible hypovolemic shock.

Discussion

Our case is one of the PADF, which Cooper first introduced; the first case report was presented by Salman [4,5]. PADF is an abnormal communication between the aorta and gastrointestinal tract, which is uncommon but life-threatening. The incidence of PADF is very low, accounting for 0.04%-0.07%, and 53 cases of PADF were presented between 1951 and 2010 [6]. PADF is more common in males than females, with a median age of 70 and a range of 23-89 years old [7].

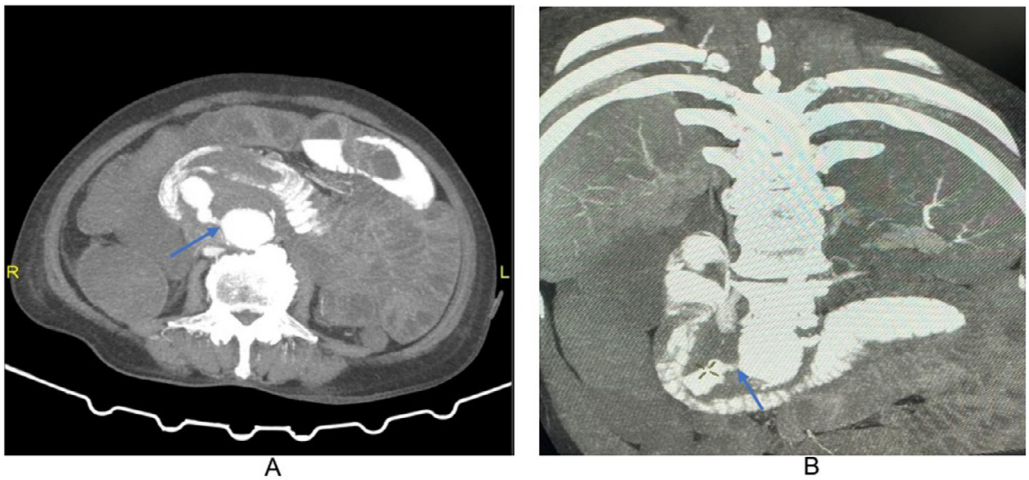

Fig. 1 – Infrarenal fusiform AAA with a size of 75 x 53 mm in a 73-year-old woman adjacent and ruptured into the second part of the duodenum on abdominal CT scan.

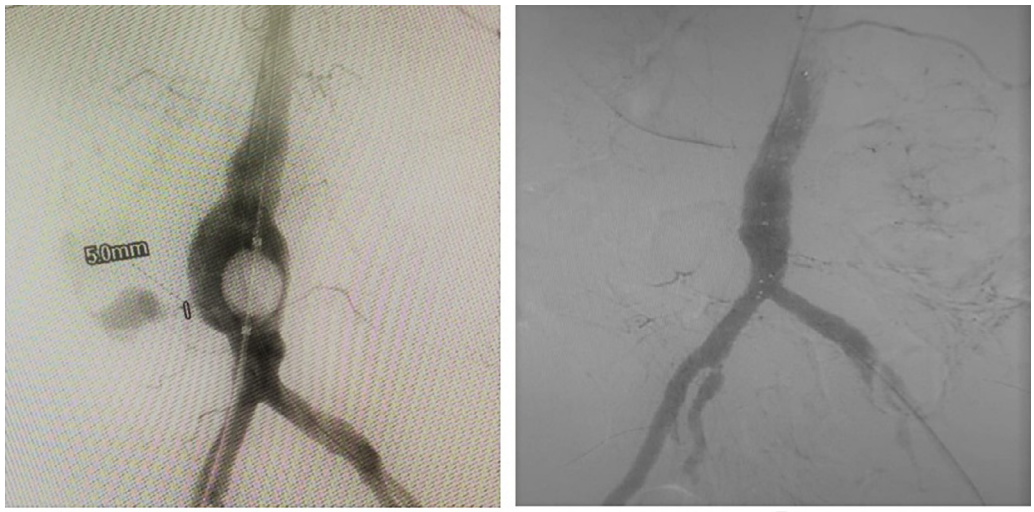

Fig. 2 – (A) Digital subtraction angiography of the abdominal artery demonstrated a infrarenal AAA. (B) Endovascular aortic repair (EVAR) of the infrarenal AAA involving the placement of 3 stent graft components show complete eradication of the aneurysm.

There are multiple potential causes of PADF, including aneurysms (most common), foreign body, tumors, radiation therapy, and infection such as tuberculosis and syphilis [8]. The proposed mechanism of this condition is likely a direct compression and inflammation between the abnormal aorta and the surrounding gastrointestinal structures. However, it is considered that aortic inflammation or infection plays a more important role [9]. Regarding the anatomical distribution, more than 70% of PAEFs involve the duodenum, particularly the third part of the duodenum, and less than 1% of lesions in the duodenal second section [1]. Diagnosis of PADF can be challenging due to its rarity, nonspecific symptoms, and difficulty confirming with available imaging modalities. Two-thirds of patients with PAEF were diagnosed in the operating room [2]. Laparotomy may be false in approximately 50% of cases [7]. Patients with PADF were generally admitted to hospitals with gastrointestinal bleeding and other presentations, including malaise, weight loss, overt sepsis, and other nonspecific symptoms [5]. The classic triad of gastrointestinal bleeding, abdominal pain, and palpable mass was demonstrated in only 6%-12% of patients with AEFs and only 3/40 cases of patients with PADFs [7,10]. Therefore, it is critical to maintain a high suspicion index in all upper GI bleeding, especially in patients with known AAA. The first diagnostic tools for patients with PADFs are EGD and CT scans. Patients with acute gastrointestinal bleeding and hemodynamically stable generally undergo upper gastrointestinal endoscopy. However, the sensitivity of upper gastrointestinal endoscopy for diagnosing AEF is less than 50%, as its common lesions are located in the third part of the duodenum and were not visualized due to angulation [11]. Otherwise, for hemodynamically stable patients with known repaired or unrepaired AAA, CT angiography is considered a valuable diagnostic study with a sensitivity of 50–94% and a specificity of 85%-100% [4,5,11]. The specific CT findings indicate the presence of PAEF, including ectopic gas in the aorta, bowel wall thickening, loss of the aortic wall, disappearance of the fat plane between duodenum and aorta, and leakage of contrast material into the bowel lumen [4,12]. The urgent ultrasound may help identify the presence of AAA but will not confirm the PAEF in patients with massive and unstable hemodynamics, which should be transferred directly to the operating room to make the diagnosis [2]. Moreover, a blood culture should be obtained whenever a diagnosis of PAEF is suspected [12]. The most common pathogens in mycotic aneurysms are *Salmonella* or *Klebsiella* [13]. Other modalities, such as arteriography, magnetic resonance imaging, and colonoscopy, are of limited value [14].

Our case raises an interesting learning point about the diagnosis. Although abdominal ultrasound suggested AAA with the hematoma on the wall of the aorta, this patient presented with moderate GI bleeding and normal findings on EGD. Therefore, there is not to urgently do further tests. It is consistent with the typical “herald bleed,” which can be self-limited, then precedes a catastrophic episode of hemorrhage later. Unfortunately, due to the very low incidence of PDAF, physicians often put PDAF in the very low differential diagnosis, particularly in young or nonspecific clinicians. If there is sufficient awareness of this high-risk condition, experienced doctors may try to observe the duodenum carefully on EGD or do a CT scan earlier.

The management of PADF includes initial resuscitation, antimicrobial therapy, bleeding control, and aortic repair if possible [7]. Without treatment, almost all patients with PADF are fatal. Surgery remains important in managing PADFs with a perioperative mortality rate of 18%-63% [3]. In addition to vascular control and reconstruction, surgery can remove the debridement of infected necrotic tissue and restorative gastrointestinal continuity, which should be performed in SAEFs. However, for PADFs treatment, there is no consensus on an approach based on large studies [7].

With the high morbidity and mortality of traditional open surgery, there has recently been the development of EVAR as a temporary method in emergency settings and as definitive management of PADF. Resuscitative Endovascular Balloon

Occlusion of the Aorta has been considered an effective intervention to control bleeding from the aorta in unstable patients, which provides time for resuscitation and a bridge to definitive care like open surgery in a delayed and controlled condition [5]. EVAR is a minimally invasive procedure with lower morbidity and mortality than operative intervention. Kakkos et al. show that 30-day mortality was 8.5%, and 12- and 24-month mortality rates were 15% and 19%, respectively [15]. Although the presence of late EVAR infection is a persistent concern, particularly with SAEF, patients with PAEF have insufficient clinical and radiographic evidence of EVAR reinfection in many studies [16].

Furthermore, infection control plays an essential role in the post-intervention period. Antibiotics should be used for 7-10 days if the culture is negative and 4-6 weeks if positive [5,13,14]. Long-term antimicrobial therapy may be recommended, but this field still lacks consensus.

Our patient followed this approach. She has undergone the EVAR to quickly control the bleeding and maybe bridge to open surgery later. At first, the balloon in the aorta successfully stopped the contrast leakage out of the aorta, which helped her hemodynamics become more stable. Unfortunately, the first stent was not fit entirely into the wall of the aorta, followed by the presence of contrast in the abdomen. Therefore, 2 more stents were placed, which prolonged the time of the procedure, and the patient was exposed to a higher level of contrast and eventually died due to multiple organ failure. When reviewing the case, we think the worse outcome for this patient may result from the severity of this disease, the delayed diagnosis, and the prolonged duration of EVAR procedure that gets more difficult for resuscitation. Evaluating the anatomy of AAA is also essential, which determines how to repair the AAA and predicts the success of the intervention. Furthermore, surgery may be considered in this case at the beginning of the presentation.

In conclusion, PADF is a rare, life-threatening condition with high mortality that can be reduced by timely diagnosis and early management. Diagnosis of PADF should be suspected in all patients with AAA or no clear sources of GI bleeding, particularly with young and nonspecific clinicians. The CT scan with intravenous contrast is considered the gold standard for diagnosis of this entity. The intervention depends on the clinical picture, the doctor's experience, and resources. Recently, EVAR has become the growing method for controlling bleeding in unstable patients or bridging procedures to a controlled setting. However, open surgery still remains important in managing PADF, especially in difficult AAA and suspected infected fistula. We hope that the increasing reviews and case reports can both remind physician's clinical sense and contribute to the database for more extensive studies.

Patient consent

A written consent was obtained from the patient's relative for publication of this case and any accompanying images.

REFERENCES

[1] Lozano FS, Muñoz-Bellvis L, San Norberto E, Garcia-Plaza A, Gonzalez-Porras JR. Primary aortoduodenal fistula: new case reports and a review of the literature. J Gastrointest Surg 2008;12(9):1561–5.

[2] Dossa CD, Pipinos, Shepard AD, Ernst CB. Primary aortoenteric fistula: Part I. Ann Vasc Surg 1994;8(1):113–20.

[3] Šumskienė J, Šveikauskaitė E, Kondrackienė J, Kupčinskas L. Aorto-duodenal fistula: a rare but serious complication of gastrointestinal hemorrhage. a case report. Acta Med Litu 2016;23(3):165–8.

[4] Guner A, Mentese U, Kece C, Kucuktulu U. A rare and forgotten diagnosis of gastrointestinal bleeding: primary aortoduodenal fistula. BMJ Case Rep 2013;2013:bcr2013008712.

[5] Bissacco D, Freni L, Attisani L, Barbetta I, Dallatana R, Settembrini P. Unusual clinical presentation of primary aortoduodenal fistula. Gastroenterol Rep (Oxf) 2015;3(2):170–4.

[6] Lin TC, Tsai CL, Chang YT, Hu SY. Primary aortoduodenal fistula associated with abdominal aortic aneurysm with presentation of gastrointestinal bleeding: a case report. BMC Cardiovasc Disord 2018;18(1):113.

[7] Georgeades C, Zarb R, Lake Z, Wood J, Lewis B. Primary aortoduodenal fistula: a case report and current literature review. Ann Vasc Surg 2021;74:518.e13–518.e23.

[8] Monteiro AS, Martins R, Martins da Cunha C, Moleiro J, Patrício H. Primary aortoesophageal fistula: is a high level of suspicion enough? Eur J Case Rep Intern Med 2020;7(8):001666.

[9] Bala M, Sosna J, Appelbaum L, Israeli E, Rivkind AI. Enigma of primary aortoduodenal fistula. World J Gastroenterol 2009;15(25):3191–3.

[10] Calligaro KD, Bergen WS, Savarese RP, Westcott CJ, Azurin DJ, DeLaurentis DA. Primary aortoduodenal fistula due to septic aortitis. J Cardiovasc Surg (Torino) 1992;33(2):192–8.

[11] eijen CL, Smulders YM, Coveliers HME, Wisselink W, Rauwerda JA, Hoksbergen AWJ. The importance of early diagnosis and treatment of patients with aortoenteric fistulas presenting with herald bleeds. Ann Vasc Surg 2016;36:28–34.

[12] Perks FJ, Gillespie I, Patel D. Multidetector computed tomography imaging of aortoenteric fistula. J Comput Assist Tomogr 2004;28:343–7.

[13] Genovés-Gascó B, Torres-Blanco Á, Plaza-Martínez Á, Olmos-Sánchez D, Gómez-Palonés F, Ortiz-Monzón E. Primary aortoduodenal fistula in a patient with pararenal abdominal aortic aneurysm. Ann Vasc Surg 2012;26(5):730.e1-5.

[14] Ishimine T, Tengan T, Yasumoto H, Nakasu A, Mototake H, Miura Y, et al. Primary aortoduodenal fistula: a case report and review of literature. Int J Surg Case Rep 2018;50:80–3.

[15] Kakkos SK, Papadoulas S, Tsolakis IA. Endovascular management of arterioenteric fistulas: a systemic review and meta-analysis of the literature. J Endovasc Ther 2011;18:66–77.

[16] Lonn L, Dias N, Veith Schroeder T, Resch T. Is EVAR the treatment of choice for aortoenteric fistula? J Cardiovasc Surg (Torino) 2010;51(3):319–27.