A CASE REPORT ON AORTOJEJUNAL FISTULA: A RARE CAUSE OF GASTROINTESTINAL BLEEDING

CASE REPORT CATEGORY

ABSTRACT

Aortoenteric fistula is an exceptionally rare condition, with a global incidence estimated at approximately <1%, and results in significant mortality if undetected. A primary aortoenteric fistula typically develops when a large abdominal aortic aneurysm is in close proximity to bowel loops that cause erosion and result in catastrophic bleeding if left untreated. This report aims to present a case of a 65-year-old male with a known abdominal aortic aneurysm who presented with hematochezia and abdominal pain. The patient underwent exploratory laparotomy, where intraoperative findings confirmed the presence of an aortoenteric fistula at the level of the jejunum. An open primary aortic repair using a bifurcated Dacron graft soaked with Rifampin solution and omental coverage with side-to-side anastomosis were performed. Aorto-enteric fistulae should always be considered as one of the differential diagnoses in the setting of an acute upper gastrointestinal hemorrhage with no apparent cause.

Aortoenteric fistula, gastrointestinal bleeding

INTRODUCTION

Gastrointestinal bleeding is a common problem in people over 60 years old, comprising up to 35–45% of patients with upper gastrointestinal (GI) bleeding.[1] Elderly people are a much vulnerable population owing to their multiple comorbidities; polypharmacy, particularly intake NSAIDs, antiplatelets and anticoagulants.GI Bleeding can originate in the upper or lower GI tract. Clinically, it presents as hematochezia, melena , hematemesis, epigastric pain and may also present as occult bleeding.[1,2] Most common causes of gastrointestinal bleeding would include peptic ulcer disease, NSAID gastritis, Mallory Weiss tears, and esophageal varices. Less common causes would include neoplasms, vascular ectasias , vasculitis , diverticula and aortoenteric fistulas. [3]

Aortoenteric fistula is an exceptionally rare condition, with a global incidence estimated at approximately 0.007%. It can be classified as either primary or secondary, with the latter often occurring as a complication of open aortic repair or any aortic surgery. A primary aortoenteric fistula typically develops when a large abdominal aortic aneurysm or a penetrating aortic ulcer is in close proximity to bowel loops, most commonly the third or fourth portions of the duodenum. Over time, prolonged pressure from the aneurysm gradually erodes into the bowel or esophageal wall, leading to the formation of the fistula. [5] Classically, aortoenteric fistula often presents initially with a minor "herald bleed," characterized by a transient gastrointestinal hemorrhage and followed by a subsequent catastrophic and life-threatening gastrointestinal hemorrhage. [5,6]

This paper aims to provide a comprehensive overview of the patient's medical history, symptomatology, clinical presentation, and management of aortoenteric fistula. It specifically outlines the surgical interventions performed and emphasizes the critical role of a thorough clinical history and physical examination, particularly in cases with atypical presentations of this rare condition.

This case report adds to the existing body of knowledge on aortoenteric fistula by emphasizing its clinical features, diagnostic challenges, and management. Furthermore, It recognizes the importance of recognizing subtle initial symptoms, such as a "herald bleed", which may precede catastrophic gastrointestinal hemorrhage. Moreover, this report underscores that early recognition, maintaining a high index of suspicion, timely use of advanced imaging techniques, and prompt intervention are essential for improving patient outcomes.

CASE

We present a case of 65 year old male adult presenting at the emergency room with 1 month history of hematochezia and melena associated with on and off left sided abdominal pain. He is diagnosed with Type 2 diabetes mellitus and hypertension for > 5 years with maintenance medications of only Losartan 50 mg once daily, Atorvastatin 40 mg once daily. He had a history of transient ischemic attack 8 months prior to this admission with no residual deficits. He was also diagnosed with pituitary macroadenoma with no surgical management done. The patient denies any history of intake of NSAIDs, steroids, antiplatelets and anticoagulants. He is a 30-pack year smoker, and an occasional alcoholic beverage drinker. The patient initially had a consult with a surgeon and was managed as Lower GI bleeding probably secondary to bleeding internal hemorrhoids vs diverticulosis. The patient was advised for further imaging hence, a non-contrast computed tomography (CT) scan of the abdomen revealed calcific plaques along the abdominal aortic wall, along with a lobulated soft tissue lesion encasing the infrarenal abdominal aorta, measuring 7.1 × 7.4 × 6.6 cm (AP × T × CC), consistent with an abdominal aortic aneurysm (AAA). Additionally, the scan showed mild diverticulosis in the ascending and proximal sigmoid colon without evidence of diverticulitis, a calcified nephrolithiasis in the right kidney, a fat-containing left inguinal hernia, and atherosclerosis of the abdominal aorta. The patient was advised to undergo surgical intervention but declined and chose to go home against medical advice. Subsequently, the patient experienced recurrent episodes of hematochezia, leading to two hospital admissions, during which they were still managed under the impression of diverticulitis. A repeat CT

scan of the abdomen with contrast noted a presence of a large distal/ infrarenal abdominal aortic aneurysm approx. 7.1 x 7.4 x 6.6 cm, with irregular intraluminal thrombus. (Figure 1, 2 respectively).

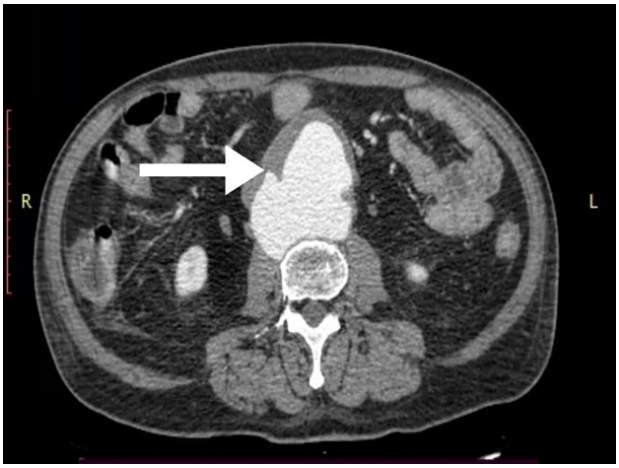

Figure 1. Axial contrast CT scan showed Infrarenal aortic aneurysm with intraluminal thrombus (as pointed by white arrow)

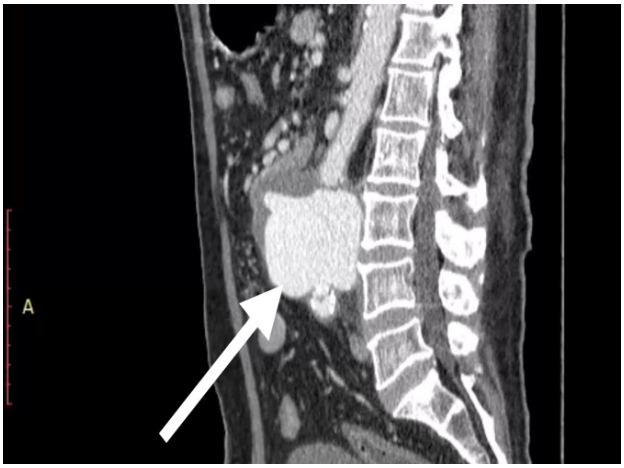

Figure 2. Sagittal axis. Contrast CT scan showed Infrarenal aortic aneurysm with intraluminal thrombus at the level of L3 vertebrae (as pointed by white arrow)

On the night before admission, the patient experienced another episode of hematochezia and was subsequently brought to the emergency room. Upon arrival, vital signs were recorded as follows: blood pressure ranging from 80/60 mmHg to 110/60 mmHg in both upper extremities, heart rate of 82 beats per minute, body temperature of 36.5°C, and respiratory rate of 21 breaths per minute. There was a palpable pulsatile abdominal mass with tenderness on slight palpation. On further examination, both lower extremities were cool and distal pulses were unappreciated. Laboratory investigations revealed anemia (Hemoglobin 9.3 g/dL), and renal insufficiency (creatinine 1.68 g/dL , EGFR 45 ml/min. 1.73 m2). Other laboratory parameters were unremarkable.

An emergency exploratory laparotomy was performed and revealed an adhesion between the AAA and the jejunum , about 5 cm from the ligament of Treitz suggesting a primary aortoenteric fistula (figure 3).

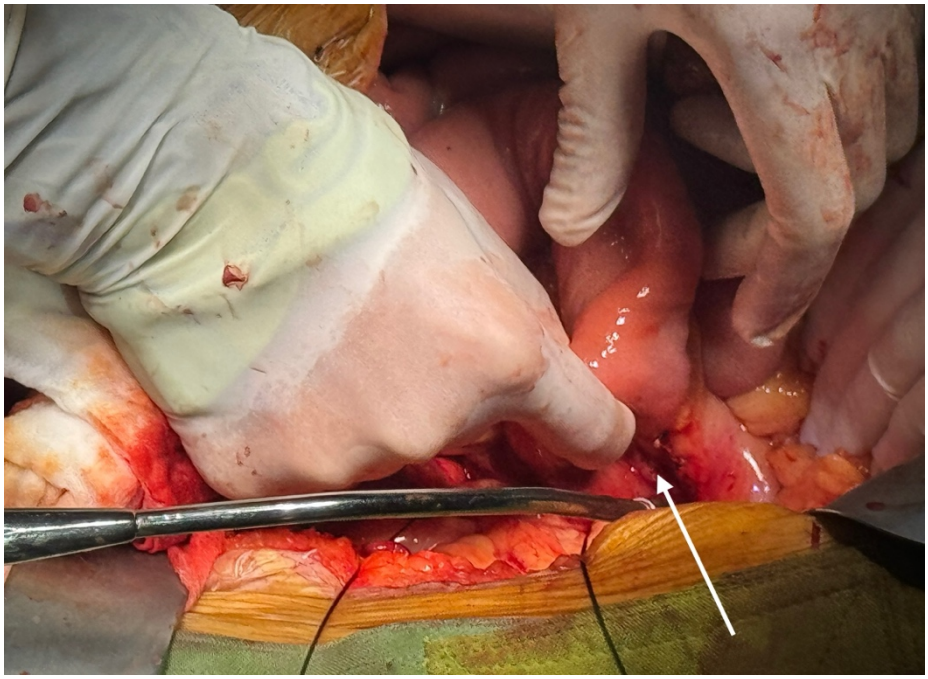

Figure 3. Intraoperative findings of Aortoenteric fistula (aortojejunal) 5 cm from the ligament of Treitz (as pointed by white arrow)

The part of the jejunum involved was resected to avoid spillage of bowel contents, and it was removed from the wall of AAA after aortic clamping. They also noted that the aneurysmal part of the aorta was already adherent to the adjacent vertebrae owing to the condition's chronicity and the aneurysm's inherent size. Intraoperatively, there were no findings of contamination and infection during the resection of the adhered segment.

In situ aortic reconstruction with a 15 × 9 mm bifurcated Dacron graft soaked with Rifampin solution and omental coverage were performed (figure 4).

The jejunum was debrided and was anastomosed in a side-to-side fashion. There were no untoward events that occurred intraoperatively and immediately post-operative. On the 2nd day post-operative day, oliguria was noted with increasing creatinine levels.

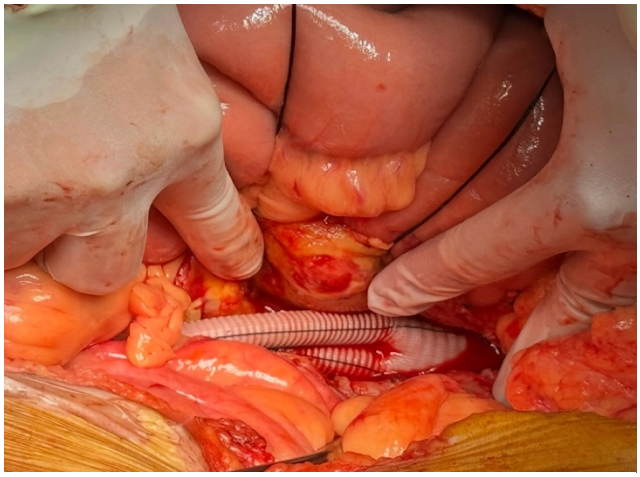

Figure 4. In situ aortic reconstruction with Dacron graft and omental coverage

Early hemodialysis was instituted to address acute kidney injury. There was notable improvement in urine output with a steady decline in serum creatinine levels, indicating improved renal function. Empiric antibiotic therapy was initiated upon admission and was continued for a total of 10 days. Throughout treatment, the patient's clinical condition significantly improved, hence, discharged on the hospital day 10.

DISCUSSION

Primary aortoenteric fistula (PAEF) is an abnormal connection between the aorta and the portion of the gastrointestinal tract , most commonly the 2nd and 3rd part of the duodenum. Less common sites would include the jejunum, ileum, colon and even esophagus. A primary aortojejunal fistula is exceptionally rare accounting for < 1% of cases worldwide and is a life-threatening condition.[7, 20] PAEF is an unusual cause of gastrointestinal bleeding and delay in the diagnosis can result to a catastrophic bleeding.[7,10] It is believed to be a result mostly from direct wear and destruction of an aortic wall causing an erosion to the adjacent organs.[7,8] The anatomical proximity of the infrarenal aorta to the distal part of the duodenum, combined with factors such as arteriosclerosis and mechanical trauma, significantly increases the risk of developing an aortoenteric fistula.[9] The clinical triad of primary aortoenteric fistula (PAEF) includes gastrointestinal bleeding, a pulsating abdominal mass, and abdominal pain, which were exhibited by the patient. This condition should be included in the differential diagnosis, particularly in patients with a known abdominal aortic aneurysm [10,11] Notably, the classic triad is observed in only 11% of cases of primary aortoenteric fistula, which adds to the diagnostic challenge of this complication. [8] Classically, all forms of aortoenteric fistula often presents initially with a minor “herald bleed,” characterized by a transient gastrointestinal hemorrhage which is self-limiting in approx. 30% of the patients and followed by a subsequent catastrophic and life-threatening gastrointestinal hemorrhage. [5,6,10]

Primary aortoenteric fistula (PAEF) poses a significant diagnostic dilemma and carries a high mortality rate if left untreated. [8,9,10] This underscores the importance of suspecting primary aortoenteric fistula (PAEF) primarily in all patients presenting with gastrointestinal bleeding with a history of abdominal aortic aneurysm (AAA). In our case, the patient presented with hematochezia and a pulsatile abdominal mass on physical examination, alongside a history of untreated AAA and left sided abdominal pain which was mistaken for a diagnosis of diverticulitis. These factors facilitated a prompt diagnosis of PAEF allowing for urgent and life-saving treatment.

Computed tomography (CT) scan, endoscopy, and angiography are commonly utilized in the diagnosis of primary aortoenteric fistula (PAEF). Among these, CT with intravenous contrast is the most effective diagnostic tool, offering a sensitivity of 50%–94% and a specificity of 85%–100%.[10,12] In this case, a contrast-enhanced CT scan was performed; however, the presence of an aortoenteric fistula was not identified in the imaging report. Endoscopy can be performed in hemodynamically stable patients, however, was not performed in this patient. It may reveal findings highly suggestive of primary aortoenteric fistula (PAEF), such as an ulcer or erosion near a blood clot accompanied by an extrinsic pulsatile mass in the duodenum. However, the diagnostic yield of endoscopy for PAEF is relatively low. This limitation may be due to the difficulty in visualizing a fistula located distal to the third part of the duodenum, where the acute angle between the third and fourth segments hampers direct visualization of the lesion. Consequently, a negative endoscopy result does not totally rule out the possibility of PAEF. Despite its limitations,

endoscopy remains valuable for excluding other potential causes of upper gastrointestinal bleeding, such as peptic ulcers and esophageal varices. [8,10,11,12]

Traditionally, the preferred treatment for primary aortoenteric fistula is surgical intervention, which typically involves aortic reconstruction—either in situ or through extra-anatomical bypass—along with repair of the affected duodenum or jejunum, as in this case. In cases of primary aortoenteric fistula (PAEF) with no or minimal contamination, as observed in this case, in situ aortic reconstruction using a Dacron or polytetrafluoroethylene (PTFE) graft with omental coverage is the preferred approach. [13, 14] In a review by Baril et al., conventional open repair continues to be associated with significant perioperative morbidity and mortality rates and lengthy hospitalizations as compared to endovascular repair. [15] During surgical aortic reconstruction, acute renal failure and respiratory failure are the most commonly encountered complications. These arise from organ hypoperfusion during aortic clamping, contributing to a mortality rate of 0.2%. Risk factors for mortality among patients post repair include age > 60 years old, baseline serum creatinine > 2.0 mg/dl, concomitant coronary artery disease, aneurysmal rupture, complementary renal revascularization. [12,13, 22] Acute renal failure was observed in this case, with noted improvement after hemodialysis.

Endovascular strategies provides an alternative therapeutic option to control bleeding and early outcomes are comparable to open surgery. However, the benefit is lost during long term follow up implying that staged approach and conversion to in situ repair may achieve best results in selected patients. [16] Furthermore, in an analysis of 791 patients with aortoduodenal fistula by Rodrigues dos Santos et al. , they reported that omental

coverage and in situ aortic reconstruction are independent predictors of improved survival outcomes . The survival rate among patients undergoing in situ aortic reconstruction is 61%–77% . [14,17]

Empiric antibiotic therapy should be initiated to target the most likely microbial pathogens. During surgery, tissue specimens must be collected for culture to guide the selection of appropriate antibiotic therapy. If cultures are negative, it is recommended to continue antibiotics for a minimum of 7–10 days. However, if cultures yield positive results, antibiotics should be tailored based on sensitivity testing and continued for 4–6 weeks postoperatively to ensure effective infection control.[11,12] As observed in this case, soaking a gelatin-sealed Dacron graft in rifampin solution evidently prevents early bacteremic graft infection and secondary foci of infection. This is done to prevent prosthetic vascular graft infections[18,19, 21] In this case, cultures were negative, and the patient showed significant improvement during the course of admission. A one-month follow-up revealed no abnormalities, though graft surveillance is still pending.

CONCLUSION

Primary aortoenteric (aortojejunal) fistula (PAEF) is a rare but life-threatening condition that poses significant diagnostic challenges due to its rarity and nonspecific presentation. A high index of suspicion is crucial for timely diagnosis and management. Surgical repair remains the mainstay of treatment, with in situ aortic reconstruction and omental coverage offering favorable outcomes in selected cases. This case highlights the importance of considering PAEF in the differential diagnosis of gastrointestinal bleeding, particularly in patients with a history of abdominal aortic aneurysm, and the importance of urgent surgical intervention to prevent fatal complications.

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