Case Series

Aortoenteric fistula as a rare manifestation of rupture of an abdominal aortic aneurysm: Case report a diagnostic dilemma

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Abstract
Introduction: Secondary aortoenteric fistula (SAF) is an uncommon, but very important complication of abdominal aortic reconstruction. The complication often occurs months to years after aortic surgery. The clinical manifestation of the aortoenteric fistula is always upper gastrointestinal bleeding. Treatment of the disease is early surgical intervention. If operative treatment is not performed promptly, the mortality is high.

Case Report: A case of secondary aortoduodenal fistula found 6 years after aortic reconstructive surgery, with the clinical presentation of upper gastrointestinal bleeding because of the increasing number of elective aortic aneurysm repairs in the aging population, it is likely that more patients with SAF will present to the clinical physicians in the future. Hence, a high index of suspicion is necessary for prompt diagnosis and treatment of this life-threatening event. The patient treated medically and finally expiration of the patient and review of the literature currently available in Medline.

Conclusion: The aim of this case report is to emphasize early diagnosis and management of all gastrointestinal bleeding in patients who have a history of aortic reconstructive surgery.


Introduction
Sudden hematemesis is a life-threatening emergency that directs a physician’s attention toward various causes of gastrointestinal bleeding. Aortoenteric fistula is an uncommon but life-threatening complication of aortic reconstructive surgery.1 Communications between the aorta and the intestine resulting from disease at either site are referred to as aortoenteric fistulas. Fistula formation between the aorta and the intestinal tract was first described in 1839 in reference to a man with a “pulsating tumor and a discharge of bloody stool,” who died suddenly. At autopsy, it was noted that “the jejunum had adhered to the aneurismal bag and that sac had ulcerated into the intestine.”2

Fistulas occurring after aortic reconstructive surgery, also called aortic graft-enteric fistulas, are considered secondary aortoenteric fistulas (SAF). Before 1960, the most common cause of abdominal aortoenteric fistulas was aortic aneurysm, followed by infectious aortitis due to syphilis or tuberculosis.3 However, over the past three decades or so, erosion of the intestine by prosthetic vascular grafts has become a much more common cause, with an incidence of up to 4%.4,5 The complication often occurs months to years after the original surgery. Bastounis et al. that the mean interval from the initial operation to the onset of upper gastrointestinal bleeding was 32 months.6 The 20 years’ experience with SAF at the Johns Hopkins Medical institution showed the average to be 2.8 years.6 The first reported SAF was reported by Brock in a case involving an aortic homograft and the duodenum.7 Claytor et al. presented the first aortoenteric fistula caused by a prosthetic graft of the aorta.8 In Mac Kenzie et al.9 demonstrated the first

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successful repair of an SAF between a synthetic graft and the intestine. Owing to the anatomic proximity, the majority of cases involve the duodenum, with the proximal suture line of an aortic prosthesis. Prompt diagnosis with surgical intervention is the only possible treatment that preserves the patient’s life. Due to the nonspecific nature of the clinical history and physical findings, diagnosis of aortoenteric fistula is difficult to make preoperatively. There is no single diagnostic investigation that has a very high specificity and sensitivity, including upper computed tomography (CT), angiography, or gallium-67 CT.

Gastrointestinal endoscopy is the most helpful method for diagnosis. If findings are negative, this test is meaningless unless another source of bleeding is found. Nevertheless, exploratory laparotomy is the only method that can definitely confirm the diagnosis.

Case Report

We present a case of secondary aortoduodenal fistula found 6 years after aortic reconstructive surgery, with the clinical presentation of upper gastrointestinal bleeding. The patient was a 70-year-old man who complained of hematemesis and melena. He gave a history of aorto-femoral reconstructive surgery in Tehran, Iran, 6 years ago. There was no history of peptic ulcer disease or any other gastrointestinal pathology.

On physical examination, the patient appeared pale with cold, clammy skin in a pre-shock condition. His vital signs were recorded as pulse rate 112 beats per minute regular, respiratory rate 22 breaths/min, and blood pressure 90/60 mm Hg. Chest wall, heart, and lungs were normal on physical examination. There was a median linear scar on his abdomen showing previous abdominal surgery. Epigastrium was tender on palpation. None of the abdominal viscera was palpable. His past surgical records revealed aorto-femoral graft 6 years ago. CT showed an aneurysmal mass around the graft (Figure 1). He was admitted with massive hematemesis and hypovolemic shock. The clinical examination, blood tests, electrocardiogram, and chest roentgenogram were unremarkable only with abdominal tenderness emergency gastroscopy could not identify a bleeding source due to massive amounts of clot and fresh blood in the stomach. However, bright red blood was seen in the stomach and injection therapy with repeated upper endoscopy was not possible due to massive amounts of clot and fresh blood in the stomach (Figure 2).

Figure 1. There is a defect in the anterior wall of the (repaired) aortic aneurysm

Figure 2. Endoscopy view of esophagus and duodenum without any pathologic finding
Surgical consultation was requested, but the surgeon refused operation due to inappropriate condition for surgery. The patient rebled, and another upper endoscopy was performed, but with no successful results and the patient was expired. This case illustrates intensive care treatment of a life-threatening hemorrhage from an aortoenteric fistula.

There is a defect in the anterior wall of the (repaired) aortic aneurysm contiguous with the third part of the duodenum. Increased attenuation material is present in the third part of the duodenum (the patient was not given oral contrast). Small bubbles of gas in the aneurysm sac and periaortic inflammatory tissue are also present. Note also the presence of peritoneal fluid with some dilated small bowel loops may be due to massive bleeding. The esophagogastroduodenoscopy showed no pathologies except for a clot in the stomach with mucosal tearing at the esophagogastric junction (Mallory-Weiss syndrome). This patient did not survive probably due to massive blood loss, very old age, and infection.

**Discussion**

The diagnosis and the treatment of aortoenteric fistula are difficult and represent a big problem for a vascular surgeon. Nevertheless in a patient with hematemesis and melena who underwent an aortobifemoral bypass or aortic interposition grafting without esophagogastroduodenal pathologies, a diagnosis of aortoenteric fistula should not be overlooked. In the present clinical case, the available clinical, instrumental, and radiological supports made the hypothesis of such a diagnosis very much presumable.

The esophagogastroduodenoscopy showed no pathologies except for a clot in the stomach with mucosal tearing at the esophagogastric junction (Mallory-Weiss syndrome). These signs, associated with high gastroesophageal bleeding and the history of aortobifemoral bypass grafting 6 years previously lead to the diagnosis of aortoenteric fistula.

The longest postoperative interval for an aortoenteric fistula was 23 years after aortofemoral bypass surgery; the shortest postoperative interval was 2 days, recorded in 1974, in which a para-prosthetic enteric fistula developed after resection of a ruptured abdominal aortic aneurysm with graft interposition. In our case, the complication presented 6 years after aortic aneurysm reconstruction.

Both in situ and extra-anatomic bypass grafting have been described in the literature. The treatment of choice is aortic ligature and axillofemoral bypass. It was reported that once the fistula identified, the surgical procedures most commonly used are graft excision, over sewing of the aortic stump, repair of the gut defect, and placing a new graft in situ or use of extra-anatomic bypass. The mortality rate during surgery and in the postoperative period is relatively high, averaging approximately 50-60%.

Chang et al. from Taiwan reported a similar case. An SAF developed in an 80-year-old patient as an immediate postoperative complication after aortic reconstruction surgery; the patient died on the 20th day after primary surgery. This patient did not survive probably due to massive blood loss, very old age, and infection. Our patient is similar and presented after 6 years with melena and hematemesis, which was diagnosed and managed with delay and the patient died on the 7th day.

In general 2 types of SAF have been described. Type 1, termed a true aortoenteric fistula or graft enteric fistula, with or without a pseudoaneurysm, develops between the proximal aortic suture line and the bowel. This type of fistula is the most common and often initiates massive gastrointestinal hemorrhage. The main clinical manifestation of this type is always upper gastrointestinal bleeding (76%), which might be either hematemesis or melena with equal frequency. Sepsis and abdominal pain are relatively rare with this type of fistula. The present case is appearing 6 years after aortic surgery was of this type.

Type 2, or a para-prosthetic enteric fistula, develops no communication between the bowel and the graft. It accounts for 15-20% of
Aorto-enteric fistula, aortic aneurysm

SAF. In this type of fistula, bleeding occurs from the edges of the eroded bowel by mechanical pulsations of the aortic graft. Sepsis is more frequently associated with this type of fistula (75%). In addition to sepsis, gastrointestinal hemorrhage (30%), abdominal pain (20%), septic emboli in the lower extremities, septic arthritis, multicentric osteomyelitis, and hypertrophic osteoarthritis (HOA) have been described.\textsuperscript{13,15}

The aim of this case report is to emphasize early diagnosis and management of all gastrointestinal bleeding in patients who have a history of aortic reconstructive surgery. Possibility of aortoenteric fistula should be considered in such cases. In selected cases, aortic reconstruction with patch graft, duodenorrhaphy, and omentoplasty can represent a valid alternation and easy choice for aortoenteric fistula without any complication.

Conflict of Interests
Authors have no conflict of interest.

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