An iliac-appendiceal fistula causing gastrointestinal bleeding

Steven D. Gurien, MD, Adam Stright, MD, Melissa Garuthara, MD, Jonathan D. S. Klein, MD, *and* Mihai Rosca, MD, *Manhasset, NY*

ABSTRACT

Aortoenteric fistulas are an uncommon cause of gastrointestinal bleeding, and iliac-appendiceal fistulas are an even rarer cause. We describe a case of an iliac-appendiceal fistula in a patient who presented several months after aortic reconstruction with gastrointestinal bleeding. An extensive workup revealed that the source of bleeding was localized to the appendiceal orifice. The patient underwent an appendectomy with a two-stage procedure involving the iliac graft for definitive repair and ultimately recovered well. Despite the rarity of aortoenteric and iliac-appendiceal fistulas causing gastrointestinal bleeding, keeping a high index of suspicion in patients with a prior vascular repair can prevent death. (J Vasc Surg Cases and Innovative Techniques 2019;5:107-9.)

Keywords: Aortoenteric fistula; Iliac-appendiceal fistula; Gastrointestinal bleeding

An aortoenteric fistula (AEF) is defined as a track between a branch of the aortoiliac system and the enteric system. It is uncommon and difficult to manage. AEFs can arise from the esophagus, stomach, small bowel, colon, or even the appendix.[1-4] AEFs can be further classified as either primary AEFs (PAEFs) or secondary AEFs (SAEFs). PAEFs are connections between the enteric system and the native vasculature. In contrast, SAEFs are connections between the enteric system and the aortoiliac vasculature secondary to aortic reconstruction, graft placement, or stent placement. PAEFs are rarer than SAEFs, with an incidence of 0.04% to 0.07%.[5] SAEFs occur with a rate of 0.3% to 2% after aortic procedures.[6] In both types, the duodenum is the most frequently involved portion of the gastrointestinal tract.[1,2,6] We present the case of an iliac-appendiceal fistula that has been reported only a few times in the literature. The patient agreed to publication of the case report.

CASE REPORT

A 67-year-old man presented with melena. He had a previous history of an abdominal aortic aneurysm, which was repaired in open fashion with an aortobifemoral prosthetic graft 13 years ago. He presented several months earlier with a thrombosed mycotic right popliteal artery aneurysm requiring a right guillotine above-the-knee amputation. The source of the infection was the aortobifemoral prosthetic graft, which required excision, washout, and replacement with an aortobifemoral human allograft. This was complicated by a pseudoaneurysm in the right iliac arterial limb of the homograft. An endovascular stent repair was performed as a bridging modality until the inflammation of the previous surgery diminished for a definitive repair of the probably reinfected graft.

He experienced multiple episodes of melena 4 months after explantation and placement of the aortobifemoral human allograft. An extensive diagnostic workup during the course of 5 weeks was undertaken to determine the source of his gastrointestinal bleeding. Multiple computed tomography (CT) angiography scans of the abdomen and pelvis were performed that did not identify the source of bleeding. The diagnostic workup also included three colonoscopies, a flexible sigmoidoscopy, an esophagogastroduodenoscopy, two bleeding scans, and a provocative mesenteric angiography using heparin and tissue plasminogen activator. The three colonoscopies were performed on admission days 3, 11, and 35. The first colonoscopy found hemorrhoids; the second showed a nonbleeding colonic angioectasia; the third saw a small amount of old blood in the cecum with clots at the appendiceal orifice (Fig 1). Given the findings of the final colonoscopy, there was concern that the patient's gastrointestinal bleeding was due to an appendiceal source that was possibly an arterioenteric fistula.

The patient became hypotensive and required multiple blood transfusions. He was taken urgently to the operating room for diagnostic laparoscopy on hospital day 35. A right iliac-appendiceal fistula was found (Fig 2).

At this point, an intraoperative vascular surgery consultation was obtained and laparotomy was performed. Appendectomy and partial cecectomy were performed using an Endo GIA stapler (Medtronic, Santa Rosa, Calif). The tip of the appendix, which had fused to the previously placed cadaveric graft, was excised en bloc with the associated portion of the cadaveric graft. This created a defect approximately 8 mm in diameter. The previously placed iliac limb of the endograft was visible through the defect. No purulence was noted, and the defect was closed primarily with a running Prolene suture. A well-vascularized omental patch was used to temporize and buttress

From the Department of Surgery, Donald and Barbara Zucker School of Medicine at Hofstra/Northwell.

Correspondence: Steven D. Gurien, MD, Department of Surgery, Donald and Barbara Zucker School of Medicine at Hofstra/Northwell, Northshore University Hospital, 300 Community Dr, Manhasset, NY 11030 (e-mail: sgurien@northwell.edu).

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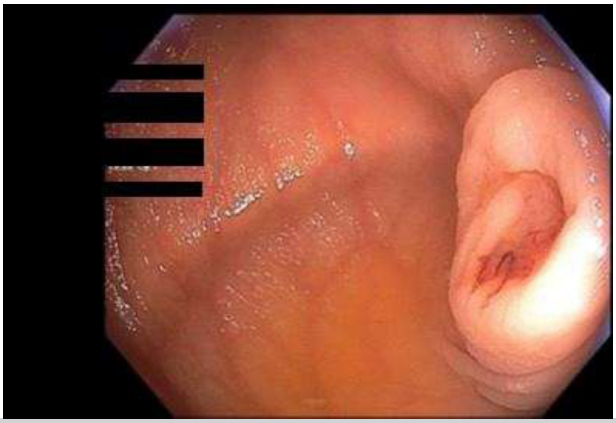

Fig 1. Appendiceal orifice with residual blood on colonoscopy.

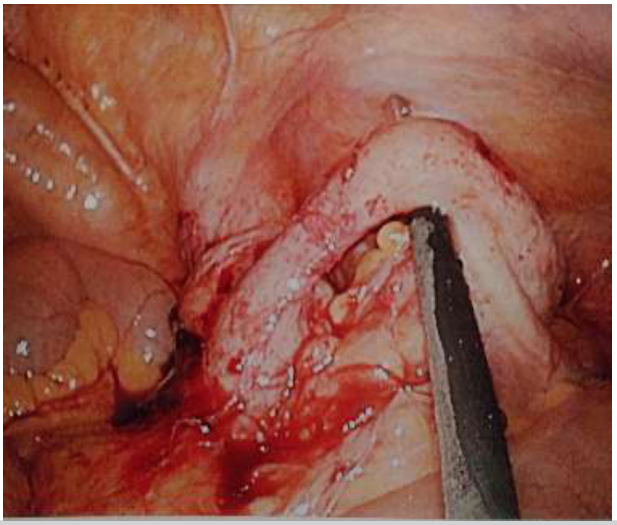

Fig 2. Iliac-appendiceal fistula during laparoscopy.

the repair, with definitive treatment planned for the following day once a replacement cadaveric graft could be obtained. A cadaveric graft and not an autologous femoral vein conduit was decided as the definitive treatment as it was thought taking the femoral vein from his one functional leg would cause significant morbidity. The patient was therefore brought back to the operating room within 24 hours to undergo excision of the right iliac cadaveric graft. There were no signs of active hemorrhage before the operating room; however, on re-entering the abdomen, 1 L of hemoperitoneum was encountered, with active hemorrhage coming from a rupture of the previously repaired fistula defect. The right iliac limb cadaveric graft was excised along with the underlying stent and replaced with a segment of aortic homograft attached proximally and distally in end-to-end fashion. A tongue of omentum was sutured over the graft with interrupted Vicryl stitches. At the end of the case, the patient had a triphasic Doppler signal distal to the anastomosis. The remainder of his hospital stay was uneventful. One month postoperatively, he was found to have a strong right femoral pulse.

DISCUSSION

AEFs are rare causes of gastrointestinal bleeding but can be catastrophic. The health care team must maintain a high clinical index of suspicion in patients with a previous aortoiliac intervention, even a remote history. A systematic review found that the median amount of time from the initial placement of a graft to the occurrence of fistulization was 2 years, with ranges as short as 1 week to as long as 26 years.[7]

AEFs are also difficult to diagnose. The classic triad of symptoms includes gastrointestinal bleeding, abdominal pulsatile mass, and abdominal pain, but the triad is seen in only 11% to 13% of patients.[8,9] The most common presenting symptom is gastrointestinal bleeding.[6-10] Approximately 25% of patients present with infectious complaints.[7] When performed, gastroscopy and colonoscopy elicited the diagnosis only 25% to 76% and 10% of the time, respectively.[7,9,10] CT scan findings of AEFs and perigraft infections are similar. However, findings of ectopic gas, periprosthetic fluid collections, and extravasation of aortic contrast material into the enteric system and of oral contrast material into the paraprosthetic space are highly suggestive of an AEF.[10,11] CT scans have been found to have a detection rate of 61% in patients with a PAEF[9] and 79% in patients with SAEFs.[7] The low incidence of AEFs and the difficulty in diagnosis lead to delays in making a definitive diagnosis. Deijen et al[12] found that the median time to make a diagnosis of AEF with a herald bleed as the presenting symptom was 14 days, with a range of 2 to 137 days. For this patient, it took 35 days of admission with multiple diagnostic scans and procedures to diagnose, locate, and surgically treat the iliac-appendiceal fistula.

AEFs that do not undergo surgical treatment are universally fatal.[6,13,14] Chopra et al[15] found that after operative intervention, 25% of their patients died each month for the first 2 months. After 2 months, mortality rates decreased. They also found that a gastrointestinal complication (which included gastrointestinal leaks and mesenteric ischemia) increased mortality rates by threefold. The surgical approach can be broken up into an open approach and an endovascular approach. The open surgical repair involves proximal and distal control, débridement of infected or necrotic tissue, and reconstruction with either extra-anatomic bypass graft or in situ graft. Placement of a graft in a noninfected field is possible with an extra-anatomic bypass, but it still carries a risk of reinfection as well as the possibility of an aortic stump blowout.[16] Another option is an in situ aortic graft replacement with a rifampin-soaked Dacron graft. One study, which looked only at patients with aortic graft enteric erosions or fistulas without excessive perigraft purulence, showed a mortality rate of 9% and a reinfection rate of 4% when patients were surgically treated

with the placement of in situ rifampin-soaked Dacron grafts.[17] The endovascular approach includes an endograft placement with or without staged open repair.[2,18] Kakkos et al[2] found in a pooled data analysis that in-hospital mortality of 823 reported cases was 30.7%. Open surgery vs endovascular approach had a significantly higher in-hospital mortality of 33.9% vs 7.1%, respectively. However, this difference becomes diminished with time as the 2-year survival is 40% for open surgery and 51% for an endovascular approach.

CONCLUSIONS

Reoperation for explantation and subsequent endovascular intervention for a pseudoaneurysm repair in a chronically infected field resulted in persistent inflammation that led to the development of an iliac-appendiceal fistula. In patients with an occult gastrointestinal bleed who have a medical history of a previous vascular repair involving the aorta or iliac vessels, it is important to consider the presence of an AEF. Whereas this diagnosis is rare, it can be fatal if it is not recognized. By keeping this diagnosis in mind, it may lead to quicker decision-making for a diagnostic laparoscopy and a decrease in repetitive interventions, hospital length of stay, and mortality.

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