Primary aortoappendiceal fistula: Case report and review of the literature

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We report a case of primary aortoappendiceal fistula in a patient with chronic, relapsing abdominal symptoms and acute lower gastrointestinal hemorrhage. The diagnosis was evident with abdominal computed tomographic scan results. The patient’s condition was successfully managed with appendectomy, abdominal aortic aneurysm resection, and extranatomic bypass grafting. Review of the literature revealed this to be the first report of a true primary aortoappendiceal fistula. (J Vasc Surg 2002;35:1284-6.)

Gastrointestinal bleeding because of a primary aortoenteric fistula (PAEF) is rare. Most often, a fistula occurs between the duodenum and the aorta. We report a case of a primary aortoappendiceal fistula presenting as chronic relapsing abdominal pain and acute lower gastrointestinal hemorrhage. We discovered only one other reference to primary aortoappendiceal fistula in a literature review.

CASE REPORT

A 66-year-old man was admitted with the acute onset of passage of bright red blood per rectum and diffuse abdominal pain. The patient was weak and lightheaded but had no loss of consciousness. He reported chronic fatigue and several episodes of lower abdominal pain associated with low-grade fevers in the 8-month period before admission. Prior evaluation results had yielded a diagnosis of recurrent urinary tract infections treated with several courses of oral antibiotics. The patient had had no previous episodes of gastrointestinal hemorrhage. He had no history of immunocompromise nor was he receiving immunosuppressive medication. Blood pressure was 121/67 mm Hg, and pulse rate was 68 beats per minute. The abdomen was soft, nontendend, and diffusely tender. A pulsatile mass was palpable. Hemoglobin was 12.8 gm/dL after two units of packed red blood cells had been given at another hospital.

Ultrasound scan results of the abdomen showed a 5.9-cm abdominal aortic aneurysm (AAA), with the suggestion of gas within the AAA. Computed tomographic (CT) scan results of the abdomen and pelvis revealed a 5.8-cm AAA; gas was visible within the luminal thrombus (Fig 1). Two periaortic fluid collections with enhancing rims were noted: one adjacent to the distal right AAA and the other extending from the AAA into the left psoas muscle (Fig 1). This constellation of findings led to a diagnosis of PAEF with abscess formation. At later review of the CT scan results, the thickened, enhancing appendix was visible adherent to the anterior distal AAA (Fig 2).

The patient was taken immediately to the operating room. At laparotomy, there was a large amount of blood filling the entire colon beginning at the cecum. No blood was in the small bowel (Fig 3). The appendix was stuck to a complex inflammatory mass overlying the distal AAA and extending into the left retroperitoneum. The inflammation was limited to these areas adjacent to the distal AAA and did not have the characteristic appearance of an inflammatory aneurysm. There was no concentric thickening of the anterior aortic wall or periaortic tissues. The duodenum, jejunum, ileum, and colon were not adherent to the aneurysm. After proximal control of the infrarenal aorta and distal control at both common iliac arteries were gained, the AAA was opened longitudinally. The AAA was inflamed and extremely friable. Thrombus was removed from the AAA, and there was a retroperitoneal abscess exiting from the aorta into the left iliopectineus muscle. The appendix was firmly attached to the AAA, and there was an easily demonstrable fistula tract between the tip of the appendix and the aortic lumen (Fig 4). There was a second retroperitoneal abscess posterior to the appendix. Because of the intense inflammation, friability of the aortic wall, and the two adjacent abscesses, it was elected to resect the anterior and lateral portions of the AAA, oversew the infrarenal aorta and the common iliac arteries, and drain the two abscesses. An appendectomy was performed, omentum placed over the aortic stump, and the abdomen closed. An axillary-femoral, femoral-femoral bypass grafting procedure was completed with polytetrafluoroethylene graft. The total elapsed time from aortic clamping to completion of revascularization was 3 hours and 5 minutes. There was no evidence of lower extremity compartment syndrome or vascular compromise after surgery. Mild postoperative pancreatitis and prolonged ileus that needed total parenteral nutrition resolved without any complications. Intravenous levofloxacin and clindamycin were administered for 14 days. The patient was discharged after 42 days in the hospital and remains well at 15 months. He is taking no antibiotics and has had no further episodes of abdominal pain or fever. An abdominal and pelvic CT scan was performed 9 months after repair, and the aortic stump appeared healthy.

Histologic examination results of the aortic wall showed atherosclerosis with chronic inflammation. The appendix showed acute appendicitis with fistula formation and periappendicitis. Cul-
ture from the abscesses at the time of operation grew alpha streptococcal species. Culture of the aortic wall grew Fusobacterium species, Prevotella species, and Actinomyces israelii. These anaerobic bacteria are common colonic flora.

LITERATURE REVIEW

In 1980, Steffes and O’Leary\(^1\) reviewed the world’s literature on 196 PAEFs, and 186 were reviewed in detail. One of these cases was reported to be a primary aortoappendiceal fistula. However, review of the original paper revealed that this patient had peritonitis from a perforated appendix. The base of the appendix and cecum were adherent to a large AAA.\(^2\) In an attempt to separate the inflamed tissue from the AAA, a fistula between the cecum and the AAA was entered. The cecum was repaired, an appendectomy performed, and the AAA resected with Dacron graft prosthesis insertion. The patient died 13 hours after the procedure. Extensive reviews of PAEF by Sweeney and Gadacz\(^3\) in 1984 and Voorhoeve et al\(^4\) in 1994 did not include any cases of primary aortoappendiceal fistula. With a Medline search from 1966 to the present, we found no additional cases, which makes the case described in our report the first true primary aortoappendiceal fistula described in the literature. In our review, we were able to find reports of secondary aortoappendiceal fistulas. In 1999, Chiche, Pitre, and Sarfati\(^5\) reported a secondary aortoappendiceal fistula and found less than 10 previous reports of secondary aortoenteric fistulas involving the appendix in the literature.

The case described by Chiche, Pitre, and Sarfati\(^5\) involved an 83-year-old man with painless hematochezia and fever 11 years after AAA replacement with a bifurcated Dacron graft. Preoperative study results were not diagnostic, and exploratory laparotomy showed a blood-filled colon, with the tip of the appendix adherent to the right iliac...
anastomosis. The patient underwent successful treatment with appendectomy, right limb of graft excision, and in situ placement of a rifampin-bonded Dacron graft. These authors concluded that secondary aortoappendiceal fistula is related to mechanical factors placing the appendix near an anastomotic pseudoaneurysm, rather than perigraft infection as the result of appendicitis.

DISCUSSION

First described by Sir Ashley Cooper in 1829, PAEF is a rare condition. An incidence rate of 0.04 to 0.07 has been reported in a large autopsy series. The most common causative factors before the antibiotic era were tuberculosis and syphilis. Other rare causes are ulcers, carcinomas, diverticulitis, foreign body perforations, and gallstones. Currently, the most common risk factor is the presence of an aortic aneurysm. Most cases reported describe fistulas between the duodenum and aorta. Other gastrointestinal locations have included the stomach, jejunum, ileum, and colon. We found only one previously reported case of primary aortoappendiceal fistula, which actually involved the caecum in a patient with perforated appendicitis.

The classic presentation of aortoenteric fistula is a triad of abdominal pain, gastrointestinal bleeding, and a pulsatile mass. Other features include fever and flank pain. A “herald bleed” may be manifested by hematemesis, melena, or hematochezia. Our patient had symptoms of chronic, relapsing appendicitis with episodic abdominal pain, fevers, and fatigue and acute lower gastrointestinal hemorrhage. The importance of a good physical examination cannot be overemphasized because 50% of patients with PAEF will have a pulsatile mass on examination results. It was the finding of a pulsatile abdominal mass in our patient that led to the rapid evaluation, which led to identification of the aortoenteric fistula.

Options for evaluation of aortoenteric fistula include esophagogastroduodenoscopy, colonoscopy, ultrasound scan, CT scan, and aortography. Esophagogastroduodenoscopy is primarily used for the identification of another obvious source of gastrointestinal hemorrhage. Occasionally, a fistula with or without an ulcer may be visualized. Ultrasound scan can accurately document an AAA and may show gas in the lumen of the aorta, as was seen in our patient. Findings on abdominal CT scan results suggestive of PAEF include AAA, gas in the lumen of the aorta, inflammation of the aorta, and an inflammatory mass next to the aorta. Occasionally, the demarcation between the aorta and an adjacent gastrointestinal structure is indistinct or shows inflammation, which suggests a possible fistula tract. Any patient with AAA and gastrointestinal bleeding, without another clear-cut cause on the aforementioned studies, should raise suspicion of PAEF.

The diagnosis of PAEF mandates surgical exploration. Patients with PAEF who do not undergo surgical repair may exsanguinate hours to weeks after the herald bleed. Surgical options include primary repair of the gastrointestinal perforation and resection of the AAA with in situ placement of an aortic graft. In the presence of frank purulence or a friable aortic wall, the diseased aorta should be resected, the aortic stump oversewn, and extraanatomic reconstruction performed. In our case, the gastrointestinal involvement was treated with appendectomy and the AAA was resected with aortic stump closure and extraanatomic bypass grafting because of gross contamination, aortic wall friability, and retroperitoneal inflammation. At 15 months after repair, the patient is well and is not taking antibiotics. Periodic evaluation of the aortic stump for signs of pseudoaneurysm or infection is planned with abdominal ultrasound and CT scans.

REFERENCES
