



Case report: Primary aortosigmoid fistula – A rare cause of lower gastrointestinal bleeding

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ABSTRACT

INTRODUCTION: A primary aortoenteric fistula (PAEF) is a communication between aorta and the gastrointestinal tract. It is a rare condition that is difficult to diagnose and therefore associated with a high mortality.

PRESENTATION OF CASE: We report a rare case of a 66-year old man with an unstable bleeding PAEF. A salvage procedure was performed with a covered stent. Later, a sigmoidectomy was performed with resection of the fistula. Postoperatively, the patient suffered an infection that was treated well with antibiotics and he was discharged two weeks later with complete resolution of the fistula.

DISCUSSION: Aortoenteric fistulas are more common secondary to previous vascular surgery of aorta, however, PAEF's involve the sigmoid in 2%. Seldom, fistulization can be due to diverticulitis that can be difficult to diagnose.

CONCLUSION: Retroperitoneal bleeding from the left iliac artery is more common due to a ruptured aneurysm. This case, however, demonstrates a special PAEF formation as a very rare complication of diverticulitis. The pathophysiology of the PAEF is very unique along with the anatomic localization in the sigmoid colon and left external iliac artery.

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1. Introduction

A primary aortoenteric fistula (PAEF) is a spontaneous formation of communication between the native aortoiliac vascular system and the gastrointestinal tract [1]. In 1829, Sir Astley Cooper described this pathological condition for the first time as a potential complication of aortic aneurysms [2]. In recent times, fistulas are most frequent seen as a complication to aortic aneurysm repair with a prosthetic graft, termed secondary aortoenteric fistulas (SAEF) [3], it can, however, seldom be seen as a complication of diverticulitis in the colon. Acute rectal bleeding, a pulsating mass and abdominal pain can be the first symptoms of a PAEF, many patients are, however, asymptomatic making diagnosis difficult [4]. A PAEF is a potential lethal condition when left untreated. In this case we report an uncommon case of an unstable bleeding from a PAEF developed by penetration of diverticulitis in the sigmoid colon to the left external iliac artery. The anatomic location and pathophysiology of the PAEF make this case very unique. With the patients written informed consent, we decided to describe the case in a repeat, using principles of the SCARE guidelines [5].

2. Case presentation

A 66-year old Caucasian male was at the hospital for an elective colonoscopy due to ongoing pain during defecation for 2 months as the only symptom, however hematochezia was observed during bowel preparation. The medical antecedent included coronary artery bypass surgery, percutaneous coronary intervention, chronic obstructive pulmonary disease, diabetes, myelo-dysplastic syndrome and 75-pack years of smoking. He had no familial diseases. His medications included Plavix, ASA and 4 antihypertensive drugs.

During colonoscopy, the patient suffered an unstable bleeding in the sigmoid colon and was acutely admitted to the hospital for stabilization. He was alert, however hypotensive with normal heart rate and fever of 38.9 °C. Tenderness in the lower left quadrant was observed at physical examination. He was anaemic (haemoglobin 6.0 mmol/L) and infected. With intravenous fluid he got stabilized, blood cultures were made and intravenous antibiotics were given. An acute contrast-enhanced computer tomography (CT) revealed retroperitoneal bleeding from the left external iliac artery (Fig. 1) that was treated with an emergency damage control endografting (12 mm wide and 8 cm long Fluency covered stentgraft, Bard, Tempe, AZ, USA) by an experienced vascular surgeon. Three days later, temperature was fluctuating between 37–39 °C, the patient experienced intermittent left-sided abdominal pain and got more septic despite continuous intravenous antibiotics. He

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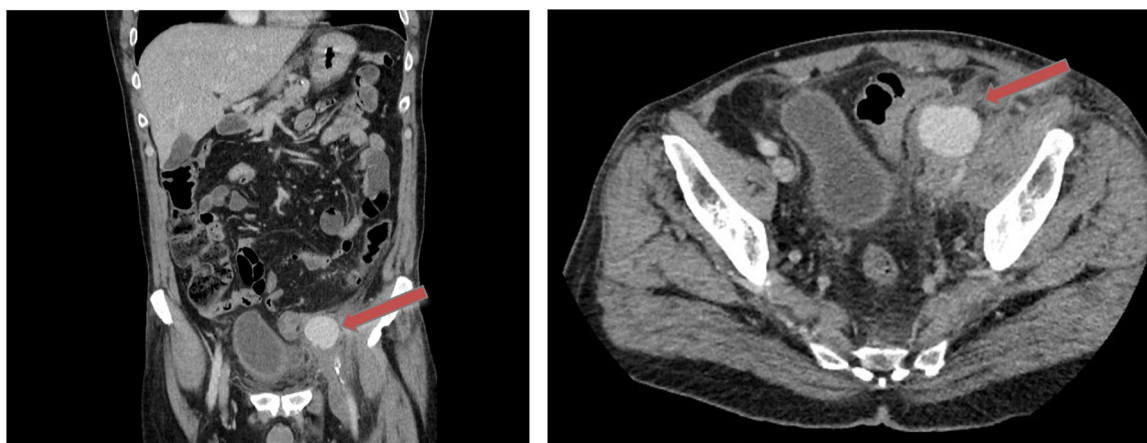


Fig. 1. A contrast-enhanced CT-scan showed contrast fluid extravasation (arrow) from the left external iliac artery.

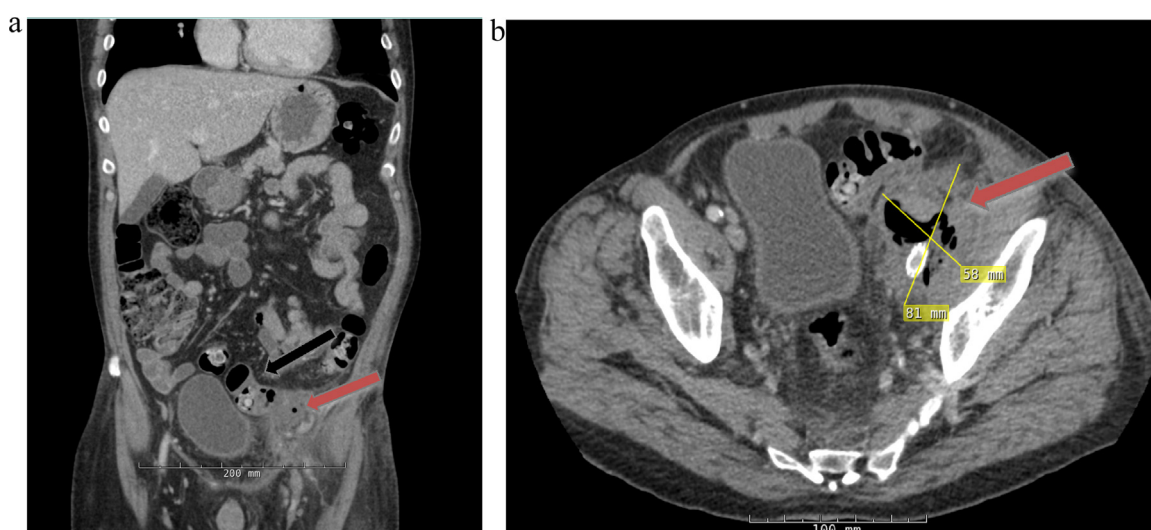


Fig. 2. (a) A contrast-enhanced CT-scan showed a hematoma including air (red arrow) near the covered stent and in close relation to the sigmoid colon (black arrow). (b) Axial slide of hematoma (5.8 × 8.1 cm) with air and reaction near the covered stent (arrow) indicating an abscess with communication between the external iliac artery and sigmoid. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

underwent another CT-scan that showed a hematoma including air near the covered stent and the sigmoidal colon in close relation (Fig. 2). *Clostridium tertium* bacterium was found in blood cultures and white blood count and C-reactive-protein concentration were decreasing.

After nine days a control CT-scan revealed regression of the infected hematoma and thickening of the sigmoid colon, indicating a possible communication to the left external iliac artery with infection around the stent area. He was informed about his surgical options that included both vascular surgery and an explorative laparotomy. He underwent an exploratory laparotomy that confirmed a fistula and a Hartmann's procedure was performed. The infected stent was removed and a femoro-femoral bypass operation from the right to the left side was performed to ensure blood flow to the left leg. He was in the intensive care unit for 5 days due to hypotension and pain with slow and good recovery hereafter. Medical specialists with the necessary expertise and experience performed the procedures.

Histological examination revealed diverticulitis with abscess, inflammation and perforation through the colon wall representing a fistula. Three weeks post-operatively, he showed signs of infection in the left groin confirmed by a CT-scan. He was treated with intravenous antibiotics and re-incision with drainage and rinsing

of the abscess cavity. He was discharged with another 2 weeks of oral antibiotics. After 2 weeks a control appointment showed no signs of vascular prosthesis infection and the antibiotics were discontinued with no further follow-up. The patient experienced stress especially with regard to the colostomy however expressed gratefulness about his recovery.

3. Discussion

PAEF's involving the sigmoid colon are uncommon and previously reported in only few cases (2%), however involvement of the third and fourth part of duodenum are more common [6].

Previously, aortoenteric fistulas (AEFs) were seen as a complication of infectious diseases and atherosclerotic aneurysms [7]. In recent times SAEF's are more frequent following repair of aortic aneurysms with a prosthetic graft [8].

Diverticulitis with breaching abscesses can cause fistulization to adjacent structures most frequent to the urinary bladder [9], ureter, vagina and other parts of the intestines [10]. This case demonstrates fistulization arising from diverticulitis complicated with abscess erosion into the external iliac artery, as seen in a CT-scan. An unstable retroperitoneal bleeding can be provided with a covered stent as a salvage procedure until laparotomy. One important learning

point from this case is that early diagnosis and treatment of PAEF's are important with a salvage treatment for the unstable bleeding patient that is described as a safe procedure with e.g. infected areas arising from e.g. pseudo-aneurysms in the groins [11].

4. Conclusion

We report a rare case of a PAEF causing pain during defecation that is an uncommon manifestation of diverticular disease or diverticulitis. The patient presented with an unstable bleeding during a colonoscopy that required a salvage procedure stenting before the fistula was resected. Early and correct diagnosis combined with surgical treatment is life saving for this patient with an unstable lower gastrointestinal bleeding. The anatomic location makes this case very unique along with the pathophysiology of diverticulitis causing a spontaneous aortosigmoid fistula. It is important that surgeons consider this as a possibility in patients known with diverticulitis with an unstable bleeding.

Conflicts of interest

There are no conflict of interest.

Funding

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Ethical approval

Non applicable.

Consent

Written informed consent has been obtained from the patient prior to the writing of the case report.

Author contribution

Both authors have written the paper together. Figures, design ect. was made by 1st author.

Guarantor

Both authors, Chirin Khalaf and Kim C. Houliand

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