Ileal artery pseudoaneurysm: a rare cause of gastrointestinal bleed

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ABSTRACT
Aorto-enteric fistulas are life-threatening conditions that occur commonly between the aorta and duodenum. Our case describes a fistula between the ileal artery and small bowel. To our knowledge, ileal artery fistulas are more commonly described in surgical cases, unlike our case. This case emphasizes early diagnosis of an aorto-enteric fistula causing life-threatening gastrointestinal bleeding to improve morbidity and survival.

1. Case
A 67-year-old woman with a remote history of colon cancer status postcolectomy and recent Stage IV endometrial adenocarcinoma treated with chemoradiation, pyelonephritis s/p left nephrostomy tube was transferred to our facility for an emergent interventional radiology (IR) embolization for a substantial gastrointestinal bleed (GIB) that led to Hgb 5 g/dL and further hemodynamic instability.

The events that transpired prior to her transfer included development of melena with a significant drop in hemoglobin requiring a total of 7 units packed red blood cells (RBC) transfusion over the course of 9 days. An upper endoscopy, colonoscopy, and tagged RBC scan were all unrevealing for any source of active or recent bleed. On hospital day 8, she again developed recurrent melena with associated hypotension with systolic blood pressure in the 50s. An urgent CT angiogram (CTA) demonstrated small contrast blush within mid-ileal bowel loops along the lower anterior abdomen suggestive of possible fistula between a left iliac aneurysm and small bowel (Images A, B and C).

On arrival to our facility, physical examination findings were significant for an alert but pale female with conjunctival pallor, intact pulses, mild suprapubic pain, and stable hemodynamics. She underwent emergent embolization of ileac artery fistula by IR and subsequently underwent stenting of the iliac artery aneurysm. Postprocedure, she had no further bleeding and tolerated advancement to a regular diet. She was transferred back to her primary facility in stable condition.

2. Discussion
Arterio-enteric fistulas (AEFs) are rare life-threatening abnormal communications between a vessel and any part of the bowel, but most commonly involving the aorta and duodenum. They may be of primary or secondary type. Primary arterio-enteric fistulas (PAEF) are rare (0.07% or less) and are due to an aneurysm that erodes as a result of malignancy, radiotherapy, infection, or inflammation. Secondary AEF are slightly more common (0.5–2.3%) [1] and include erosion of a prosthetic graft, as described in surgical cases.

A review by Saers and Scheltinga found that a fistula was associated with the presence of an aneurysmal aorta in 83% of patients [2]. At least 50% of these fistulas involved the duodenum. The incidence of secondary aortoenteric fistula due to erosion of a previously placed prosthesis makes up 0.36–1.6% of the cases [3].

Our patient had a primary aortoenteric fistula, likely due to her history of colon cancer, metastatic endometrial cancer to the colon, and abdominal radiation treatments. Additionally, she had a complex abdominal history and multiple insults to the bowel in the setting of abdominal and pelvic surgeries such as nephrostomy stenting, vascular interventions, and sepsis. This case is exceptionally unique due to the rare involvement of the ileal artery rather than mesenteric vessels causing a fistulous communication between the pseudoaneurysm and bowel, leading to intermittent hemorrhage. In our literature search, cases of an ileal artery fistula have mostly been reported as uretero-arterial fistulas secondary to an ileal conduit following cystectomy or cystoprostatectomy, unlike our case of ileo-enteric fistula.
Since these fistulas are extremely rare, early diagnosis can be challenging. One should have high index of suspicion if common etiologies are ruled out with routine diagnostic tests. In Saers and Scheltinga’s literature review, gastroduodenoscopy was pursued in 59 of 81 patients with detection of PAEF in only 25% \[2\]. CT with IV contrast is becoming the accepted initial diagnostic test for suspicion of PAEF as it can help establish a diagnosis preoperatively in over one-third of patients \[4,5\]. Positive CT findings typically includes bowel wall thickening, para-aortic or intra-aortic gas, and extravasation of contrast into the bowel lumen \[3\]. In secondary AEF, the ability of CT to diagnose advanced graft infection such as periprosthetic abscess or aortoenteric fistula can have a specificity of 100% \[6\]. In a retrospective study performed by Deijen et al from 1994–2013, a CTA identified a fistula in 35% of cases where as an EGD identified a fistula in only 25%. Some patients had multiple EGDs, resulting in an increased detection rate \[7\]. Colonoscopy was performed in 6 patients; none of which revealed fistula \[7\].

Because of high mortality rates associated with aortoenteric fistulas, making the diagnosis early is crucial. The differential diagnosis for a patient presenting with ‘herald bleed’ where a spontaneous bleed stops abruptly, only to recur days to weeks later should include PAEF. Other risk factors that should clue a physician to this diagnosis include history of aortic aneurysms, prior abdominal and vascular surgeries, presence of diverticulosis, and history of radiation therapy.

In about 30% of patients, the time period between herald bleed and massive bleed can be as little as 6 hours \[2\]. However, only about 11% of patients present with the classic triad of herald bleed: gastrointestinal bleeding, abdominal pain, and pulsatile abdominal mass. Among these, GI bleed is the most common and can present as hematemesis and or melena secondary to a rupture of the adjacent aorta into the GI tract \[3\].

Treatment for PAEF has moved toward an endovascular approach, although rebleeding can occur. This approach enables rapid bleeding control and can reduce perioperative morbidity and improve early survival. Stent grafts have shown to control bleeding in 80% of cases but may still require reintervention\[8\]. The perioperative mortality rate of PAEF can be 18–63% whereas untreated mortality rate can be as high as 80–100% \[1,9\]. Imperative to note that in patients that have good-life expectancy but are poor surgical candidates due to any coexisting medical conditions or critically ill presentation, endovascular approach should be considered as a bridging treatment towards a more definitive open surgery \[8\]. In our patient, an iliac artery stent was placed (Image D) which successfully stopped bleeding. Although an endovascular approach can potentiate infections, our patient is not an operative candidate due to the extent of her metastatic cancer.
Disclosure statement
No potential conflict of interest was reported by the authors.

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Image C. 3D visualization of pseudoaneurysm projecting from the left common iliac artery.

Image D. Angiogram visualizing balloon expandable endoprosthesis deployed over site of pseudoaneurysm.