



An unusual case of proctosigmoiditis secondary to arteriovenous fistulization

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ABSTRACT

Refractory chronic colitis presents a medical and surgical challenge, and underlying etiologies are diverse with potential for misclassification as inflammatory bowel disease. We present an unusual case of chronic proctosigmoiditis with rare vascular etiology. A 48-year-old Caucasian male presented with severe diarrhea, weight loss, and abdominal pain. Computed tomography (CT) suggested proctosigmoiditis. Colonoscopy and biopsy findings were non-specific but were suggestive of ischemic etiology and venous congestion. He was initially treated with antibiotics, steroids, and mesalamine, which did not show any improvement. Mesenteric angiography showed a fairly large irregular and bizarre vessel consistent with a large arteriovenous fistula (AVF) associated with one of the branches of the inferior mesenteric artery. AVF was too large to be embolized, and he underwent a laparoscopic low anterior resection with creation of a coloproctostomy and protective diverting loop ileostomy. An AVF was found at the origin of the ascending left colic artery. Inferior mesenteric vein thrombosis and arteriovenous fistulization are rare vascular causes of chronic proctosigmoiditis, but these should be considered in refractory cases. Both initial diagnosis and surgical treatment can be challenging.

Keywords: Proctosigmoiditis, inferior mesenteric vein thrombosis, arteriovenous fistula

INTRODUCTION

Refractory chronic colitis presents a medical and surgical challenge, and underlying etiologies are diverse with potential for misclassification as inflammatory bowel disease. An unusual case of chronic proctosigmoiditis with rare vascular etiology is presented. Patient consent and Institutional Review Board approval were obtained for this study.

CASE PRESENTATION

A 48-year-old Caucasian male presented with a 2-month history of intermittent fecal incontinence and, urgency, severe diarrhea, weight loss, and abdominal pain. His past medical history was not significant. Computed tomography (CT) of the abdomen and pelvis demonstrated thickening of the left colon and rectum. Initial flexible sigmoidoscopy revealed mild macroscopic colitis, with unremarkable mucosal biopsies. Oral antibiotics, steroids, and mesala-

mine (Ferring Pharmaceuticals, Parsippany, NJ, USA) were started, as well as mesalamine enemas, for a presumed diagnosis of ulcerative colitis. The regimen did not provide any relief of symptoms.

Approximately 2 months into his illness, he presented to our institution for a second opinion. Prior to presentation, he had undergone multiple endoscopic procedures and CT scans. Laboratory examination results revealed normal hemoglobin level, white blood cell count, electrolytes, liver biochemistries, and C-reactive protein level. Stool studies ruled out infectious colitis. CT enterography was consistent with colitis, with continuous involvement of the left colon and rectum and the most pronounced inflammatory changes in the rectum and sigmoid (Figure 1). Mesenteric arteries were patent; however, the inferior mesenteric vein (IMV) was not visualized. Radiologic studies from an outside hospital did not note an IMV occlusion. Re-

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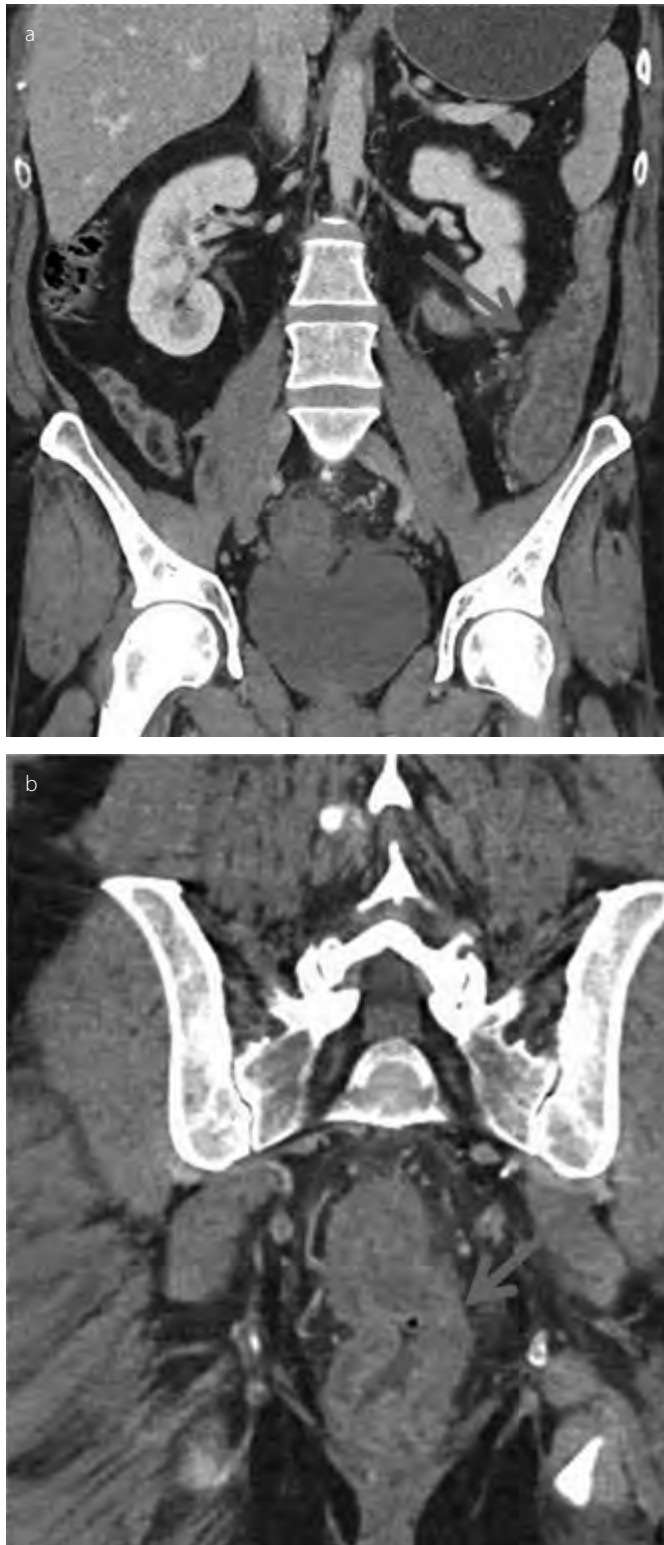


Figure 1. a, b. CT enterography with inflammatory changes in the rectum and sigmoid

CT: computed tomography

peat sigmoidoscopy demonstrated a diffuse area of severely congested, erythematous, ulcerated mucosa extending to 50 cm above the anal verge (Figure 2). Biopsies revealed patchy congested vessels in the lamina propria with fibrin thrombi and fibrinoid changes in the vessel walls, a pattern suggestive

of a thrombotic microangiopathy injury. Immunostaining for Immunoglobulin-A (IgA) was positive in the fibrinoid vessel walls and negative for cytomegalovirus. No clotting abnormalities were identified, and evaluation for vasculitis did not reveal any abnormalities. Differential diagnosis included drug effects, infection, autoimmune conditions, and ischemic colitis from passive congestion.

Computed tomography angiography demonstrated a patent celiac axis, superior mesenteric artery (SMA), and inferior mesenteric artery. Venous phase demonstrated hypervascularity around the left colon with multiple tortuous vessels enhancing in the arterial phase, along with prominent left gonadal veins (Figure 3). IMV was again not visualized, and superior mesenteric vein and portal vein were patent and without abnormality.

Further evaluation with mesenteric and renal angiography showed normal bilateral renal arteries and a widely patent and normal-appearing SMA. Delayed venous phase imaging revealed a patent SMV and portal vein. However, there was a fairly large irregular and bizarre vessel that enhanced very early from either the ascending branch of the left colic artery or one of the sigmoid branches. This did not appear to be arterial, and was filled earlier than the other venous structures. The appearance was consistent with a large arteriovenous fistula (AVF) (Figure 4). IMV again was not visible on imaging.

Therapeutic enoxaparin (Sanofi-Aventis) was initiated; however, after 6 weeks, his symptoms did not significantly improve. AVF was too large to be embolized, and he underwent a laparoscopic low anterior resection with creation of a coloproctostomy and protective diverting loop ileostomy. In surgery, the mesentery of the left colon was thickened, edematous, and foreshortened to the level of the anterior peritoneal reflection. A sharp demarcation in the wall of the intestine was found at the junction of the descending and sigmoid colon, where a pink and healthy-appearing proximal colon transitioned into an extremely thickened and erythematous distal bowel. Engorged venous collaterals were encountered between the left gonadal veins and colonic mesentery. The AVF was noted to be at the origin of the ascending left colic artery.

Gross pathologic examination demonstrated hemorrhagic serosa with mild fibrinous exudate, edematous mucosa with patchy dusky areas, and multiple pink firm nodules of 0.2-0.5 cm in size. Histologic examination found focal erosion and mucosal changes consistent with ischemic bowel disease, with submucosal blood vessel proliferation.

DISCUSSION

Recent clinical guidelines introduced by the American College of Gastroenterology raise the question of the mere existence of



Figure 2. a-c. Sigmoidoscopy with severely congested, erythematous, and ulcerated mucosa



Figure 3. CT angiography with venous hypervascularity around the left colon
CT: computed tomography

chronic ischemic colitis as a unique entity. Therefore, this case is of particular interest as it meets strict criteria described in the guidelines, including the duration of more than 3 months and histologic confirmation (1).

Both IMV occlusion and arteriovenous fistulization have been implicated in chronic colitis in previous reports, and usually not noted seen at the same time (2). Arteriovenous fistulization in the IMA territory is not a commonly recognized etiology of colonic ischemia (CI), with only 26 cases identified in a recent review (3). Pathophysiology is thought to be the result of simultaneous shunting of left hemicolon arterial perfusion and congestion of venous circulation (3). Mesenteric venous occlusion is another uncommon cause of CI. A recent series of confirmed CI identified venous occlusion as a cause in only one out of 313 patients (4). IMV occlusion due to other etiologies has been described as a causative factor of CI. Ookura et al. (5) presented two patients with remarkable similarities to our patient. Both had an occluded IMV and evidence of AVF on angiography, with a similar pattern of colitis. They speculated that arteriovenous fistulization is not infrequent and could have been

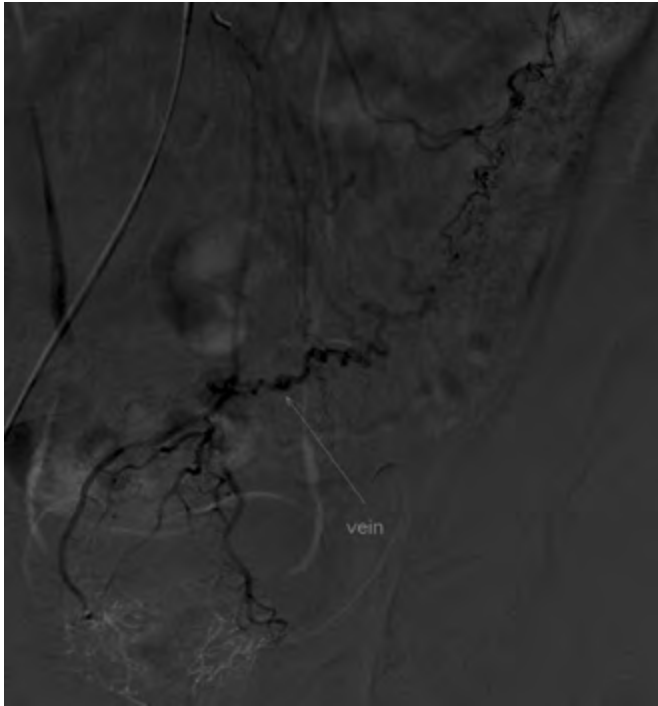


Figure 4. Mesenteric angiography with arteriovenous fistula

formed by impingement between the distended vein and neighboring artery. However, they concluded that it is impossible to determine if AVF is primary or secondary to venous occlusion and that venous congestion certainly worsened because of the development of the fistula.

Our report demonstrates the difficulties in diagnosing this rare entity, and a vascular etiology should be kept in mind as a potentially treatable cause of refractory colitis. In general, vascular imaging is not indicated in most cases of suspected CI because it typically develops in the absence of major vasculature occlusion, although mesenteric angiography may be necessary in order to clearly identify the cause of refractory colitis, similar to our case.

Refractory chronic colitis presents a medical and surgical challenge, and underlying etiologies are diverse with potential for

misclassification as inflammatory bowel disease. IMV thrombosis and arteriovenous fistulization are rare vascular causes of chronic proctosigmoiditis, but these should be considered in refractory cases. Both initial diagnosis and surgical treatment can be challenging.

Informed Consent: Written informed consent was obtained from the patient who participated in this study.

Peer-review: Externally peer-reviewed.

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