



# “Not All That Fistulizes Is Crohn’s Disease”: A Rare Case of Ischemic Jejunocecal Fistula

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## ABSTRACT

Enterocolic fistulae lead to abnormal diversion of gastrointestinal contents from the small intestine to the colon, causing diarrhea and protein energy malnutrition. We describe a rare case of a 40-year-old patient presenting with chronic diarrhea, unintentional weight loss, sitophobia, and severe abdominal pain in the setting of extensive thrombosis due to JAK2 V617F mutant disorder and associated tobacco use. Computed tomography enterography demonstrated an enterocolic fistula (jejunum to cecum), and a colonoscopy confirmed a 4 mm fistula in the cecum. The patient was nutritionally optimized with plans for a surgical fistula takedown in the future.

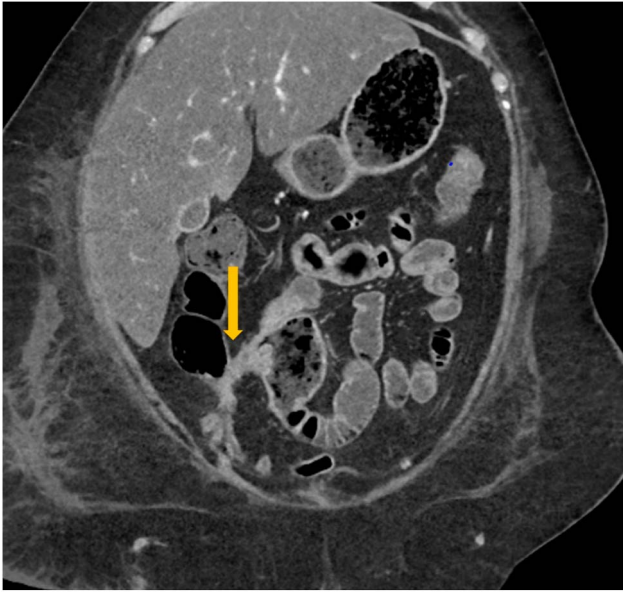
**KEYWORDS:** jejunocecal fistula; enterocolic fistula; chronic mesenteric ischemia

## INTRODUCTION

An intestinal fistula is an abnormal communication between the intestine and an adjacent organ or surface. Enterocolic fistulae lead to an abnormal diversion of gastrointestinal contents from the small intestine to the colon, causing diarrhea and nutrient malabsorption.<sup>1</sup> They may be caused by inflammatory conditions such as inflammatory bowel disease (eg, Crohn’s disease) but can also be a complication of a prior surgery, foreign bodies, diverticulitis, or malignancy.<sup>2</sup> Rarely, enterocolic fistulae can be caused by ischemic injury.<sup>3</sup> We describe a case of a jejunocecal fistula due to chronic mesenteric ischemia leading to severe chronic diarrhea.

## CASE REPORT

A 40-year-old woman with morbid obesity, nicotine dependence, and type 2 diabetes mellitus was evaluated for epigastric pain and chronic diarrhea. She also had sitophobia and postprandial urgency, with a weight loss of 30 kg over the past 6 months with a medical history of splenic infarct 5 years ago. Her computed tomography (CT) angiography showed non-flow-limiting segmental and extensive thrombosis involving the abdominal aorta with extensions into the celiac artery, common hepatic artery, left gastric artery, and splenic artery. A peripheral blood myeloproliferative neoplasm panel identified a JAK2 V617F mutant disorder with an allele fraction of 0.7%. In addition, her bone marrow biopsy revealed a hypercellular bone marrow (70%–80%) with adequate-to-increased trilineage hematopoietic precursors and no overt features of an underlying myeloproliferative neoplasm. Considering the worsening of her symptoms (sitophobia, weight loss, JAK2 mutation) in the setting of extensive arterial thrombosis, she was diagnosed with chronic mesenteric ischemia. Consequently, her decreased food intake resulted in nutritional deficiency and muscle weakness, which limited her ambulation. Despite identifying chronic mesenteric ischemia as an underlying cause of her sitophobia, abdominal pain, and weight loss, her chronic diarrhea remained an enigma. For the evaluation of her chronic diarrhea, she underwent an esophagogastroduodenoscopy with gastric and small bowel biopsies, which were normal. Although she previously had a CT angiography of the abdomen and pelvis, a CT enterography was recommended to further evaluate the small bowels for any organic causes of chronic diarrhea. The CT enterography demonstrated an enterocolic fistula (jejunum to cecum) (Figure 1). She underwent a colonoscopy that confirmed a 4 mm fistula in the cecum (Figure 2). Random colonic biopsies were normal, and biopsies from the fistula showed active chronic inflammation (Figure 3). Her fecal calprotectin level was measured to be 2,600 (μg/mg). The overall evaluation favored this isolated fistula in the setting of complex atherothrombotic disease to be from an ischemic origin rather than



**Figure 1.** Computed tomography enterography demonstrating an enterocolic fistula (yellow arrow).

related to Crohn's disease. The patient is currently being nutritionally optimized with total parenteral nutrition, which improved her diarrhea. She is also planned for a surgical fistula takedown in the future.

## DISCUSSION

Intestinal fistulae are complex conditions with high morbidity and mortality rates. These fistulae may be classified into lateral/side fistulae, which allow the normal progression of the intestinal content, and end fistulae, which result in a complete loss of intestinal continuity and generally require surgery for correction.<sup>4</sup> In addition, there are multiple classification systems for gastrointestinal fistulae (eg, anatomical, output volume, etiological), which carry specific implications about several

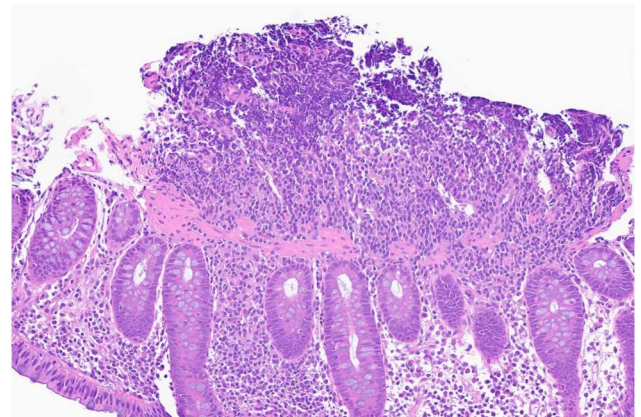
outcomes including prognosis, spontaneous closure, operative timing, and nonoperative care planning.<sup>4</sup> Intestinal fistulae are most commonly caused by abdominal surgery (75%–85%), in the setting of infection or breakdown of an intestinal anastomosis due to ischemia, tension, or distal obstruction.<sup>5</sup> Spontaneous fistula formation is uncommon (15%–20% of cases), and causes include inflammatory bowel disease (specifically Crohn's disease), diverticular disease, cancer, radiation enteritis, and pancreatitis.<sup>4</sup> Ischemia is a very rare cause of spontaneous intestinal fistulization, with scant case reports described in the literature regarding acute ischemia.<sup>3</sup> However, to the best of our knowledge, there has not been any report of an enterocolic fistula in the setting of chronic ischemia (eg, chronic mesenteric ischemia). The pathophysiology of fistulae occurrence after ischemia is still not well-understood, but likely in the setting of ischemic injury causing tissue breakdown.

The diagnosis of an intestinal fistula can be challenging because clinical clues can be varied and often be subtle. Patients can present with pain; diarrhea; and weight loss; and complications such as protein-energy malnutrition, abscesses, and sepsis.<sup>6</sup> Fistulae can be diagnosed by multiple modalities; however, little data are available regarding the best imaging modality in these cases.<sup>4</sup> CT imaging may be performed first, especially in acute settings of intestinal fistulae, and may guide the planning for surgical correction.<sup>7</sup> However, the high accuracy of magnetic resonance imaging in the evaluation of intestinal fistulae (eg, perianal fistulae in Crohn's disease<sup>8</sup>) may be preferred when other modalities fail to diagnose the fistulae.<sup>7</sup>

The management of patients with a gastrointestinal fistula requires a comprehensive team approach. Generally, conservative management is preferred over any surgical intervention. This includes maintaining fluid and electrolyte balance and providing bowel rest with nutrition support.<sup>9</sup> In fact, fluid and electrolyte replacement is of utmost importance when the fistula effluent drainage reaches greater than 1,000 mL/d (eg, fistula located in the proximal region of the small bowel).<sup>10</sup> In



**Figure 2.** Colonoscopy image of a fistula.



**Figure 3.** Biopsies from around fistulous opening showing active inflammation.

certain cases of intestinal fistulae, enteral nutrition is recommended to avoid mucosal villi atrophy and allow for a normal bowel function.<sup>10</sup> Medical management might include medications such as octreotide to decrease the fistula output by inhibiting the release of certain gastrointestinal hormones (eg, gastrin).<sup>11</sup> Surgical management might be needed for fistula takedown in certain cases with failure of conservative treatment or high-output/highly symptomatic fistulae.<sup>7</sup> Endoscopic treatments (eg, over-the-scope clip) represent a newer and minimally invasive technique that have shown promising results in sealing visceral perforations in several clinical settings (eg, postsurgical and traumatic fistulae).<sup>12</sup> In this case, our patient only required conservative management to resolve the associated symptoms with the colonic fistula.

In conclusion, enterocolic fistula is a rare but serious complication that can be caused by various conditions including chronic ischemia. Most fistulae are managed medically, but may need surgical or endoscopic takedown in highly symptomatic cases.

## DISCLOSURES

**Author contributions:** All authors wrote, edited, and approved the final version before submission. V. Chedid is the article guarantor.

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