



Eosinophilic Colitis in Recurrent Sigmoid Volvulus

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ABSTRACT

Eosinophilic colitis (EC) falls along the spectrum of a series of inflammatory gastrointestinal disorders in which eosinophils infiltrate the gut without known tissue eosinophilia. Eosinophilic gastrointestinal disorders include eosinophilic esophagitis, eosinophilic gastritis, and the least common EC. The presentation of EC is extremely variable in mucosal, submucosal, and transmural inflammation. We present a case of recurrent volvulus with histologic findings of eosinophilia.

INTRODUCTION

Primary eosinophilic gastrointestinal disorder is defined as the presence of intestinal inflammation and eosinophils in the absence of peripheral eosinophilia.^{1,2} Any portion of the gastrointestinal tract can be affected by eosinophilic colitis (EC), with the least common portion being the colon. Primary EC is very rare because the presence of colonic eosinophilia is most commonly seen in inflammatory bowel disease, immunoglobulin E-mediated food allergens, and parasitic infections.^{3,4} For this reason, EC remains a diagnosis of exclusion at this time. The differential of colonic eosinophilia includes inflammatory bowel disease, parasitic infections, and allergens. Primary EC typically presents as diarrhea in the mucosal form, whereas transmural has been associated with volvulus, intussusception, and even perforation in select cases.³ In previous literature, the cecal volvulus has been associated with EC. However, the prevalence of sigmoid volvulus has not been reported. We present a case of recurrent volvulus with subsequent sigmoidectomy with histologic findings of transmural and subserosal eosinophilic infiltration.

CASE REPORT

A 55-year-old woman with a previous sigmoid volvulus and resultant sigmoidectomy in 2017 presented with 1.5 weeks of abdominal pain, increasing abdominal distention, and constipation. She had not had a bowel movement for 5 days at the time of presentation and was no longer passing flatus. She stated that other than her abdominal symptoms, she had no other complaints. Per the patient, her symptoms were very similar to her previous volvulus in 2017, however, slightly less severe than her previous presentation. At that time, endoscopic decompression was successful with subsequent resection of 34.9 cm of the sigmoid colon approximately 2 weeks later and anastomosis achieved through staples. She had no abnormal laboratory values, and her vital signs were within normal limits. Her examination was notable for a distended, tympanic abdomen without any significant tenderness.

Initial abdominal and pelvic computed tomography with IV contrast showed an acute sigmoid volvulus causing a large bowel obstruction. This was also initially evident on a kidney, ureter, and bladder (Figure 1). The patient was initially evaluated by a general surgeon who recommended evaluation from a gastroenterologist before considering surgical intervention.

She was taken for an urgent attempt of endoscopic decompression with flexible sigmoidoscopy. The water immersion technique was used with normal-appearing mucosa until approximately 25 cm from the rectum at the sigmoid flexure. At this point, the examination showed a swirling appearance of the lumen, consistent with volvulus (Figure 2). At this point, the mucosa was friable, erythematous, and ulcerated concerning for a subacute-to-chronic appearance. There was a stricture approximately 2 cm in length at the transition point, which was traversed with a pediatric colonoscope with minimal resistance. Proximal to the stricture, a moderate amount of gas and stool was seen with otherwise normal-appearing mucosa. Multiple attempts of endoscopic decompression were



Figure 1. A kidney, ureter, and bladder x-ray with evidence of an acute sigmoid volvulus and dilated colon noted proximal to the site of volvulus.

unsuccessful. The surgical team was notified and recommended bowel preparation. The patient continued to have symptoms similar to presentation during preparation, and she was subsequently taken for an open sigmoidectomy with end-to-end anastomosis 5 days after initial presentation. During this sigmoidectomy, she was noted to have a redundant sigmoid and 16.5 cm of colon was resected and anastomosis achieved with an 80-mm gastrointestinal anastomosis stapler.

Pathology of the surgical resection showed eosinophilic infiltration of the fibrotic submucosa with extension through the

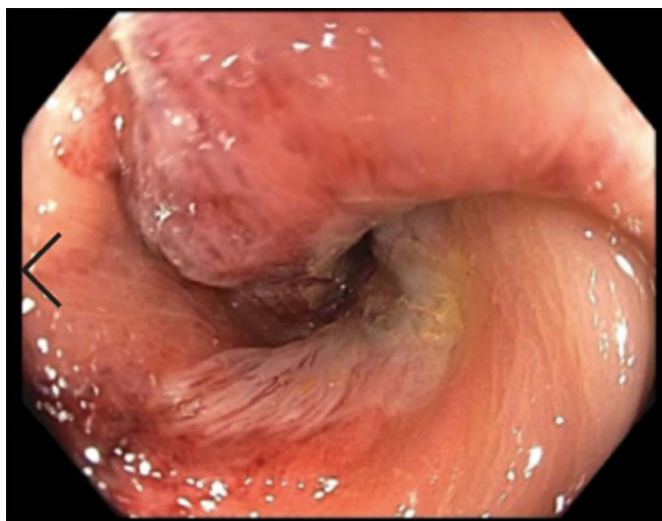


Figure 2. Endoscopic images of the sigmoid colon and swirling appearance of the lumen with friable, erythematous tissue.

muscularis propria into the subserosa and involving serosal adhesions (Figures 3 and 4). The findings were present in areas underlying mucosa with the ulceration at the anastomotic site and away from the anastomotic site in areas with normal overlying mucosa. No additional biopsies from other colonic sites were taken. Features to suggest inflammatory bowel disease were not identified in the mucosa, and there was no evidence of parasitic organisms. A review of the previous 2017 resection demonstrated no evidence of eosinophilic infiltrate. Since surgery, the patient has done very well with no further recurrence of symptoms.

DISCUSSION

EC is rare, with only a handful of cases reported in the literature since 1970.¹ At this time, no established diagnostic criteria are making definitive diagnosis challenging. Common presentations of EC include abdominal pain, cramping, diarrhea, and weight loss. Given that colonic eosinophils are present in allergies, inflammatory bowel disease (IBD), and parasitic/helminth infections, primary EC is a diagnosis of exclusion requiring extensive clinical and laboratory correlation.^{1,3} Our patient had no history of allergies, no symptoms or pathology consistent with IBD, and no history to support parasitic infection.

Primary EC more typically affects neonates and juveniles, with rare adult presentations. In younger patients, the disease is typically mild and self-limited. Adult presentations depend on the layer of colon affected. Mucosal infiltration typically presents with malabsorption, diarrhea, and protein-losing enteropathy. Transmural infiltration, on the other hand, presents with colonic thickening and intestinal obstruction.^{1,4} Histologic features demonstrate a dense eosinophilic infiltration of the colon in either a segmental or diffuse pattern. Diagnosis typically requires

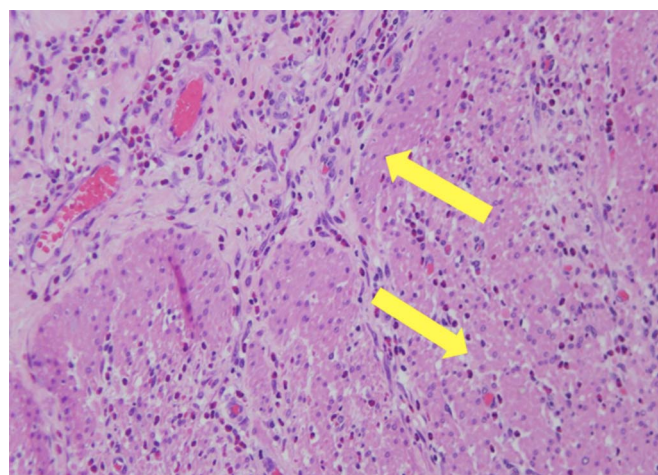


Figure 3. Sections away from the ulceration at the previous anastomotic site demonstrate a dense eosinophilic infiltrate in the fibrotic submucosa (yellow arrow, upper left of image) with extension into the muscularis propria (yellow arrow, lower right of image) (hematoxylin and eosin stain, 200× magnification).

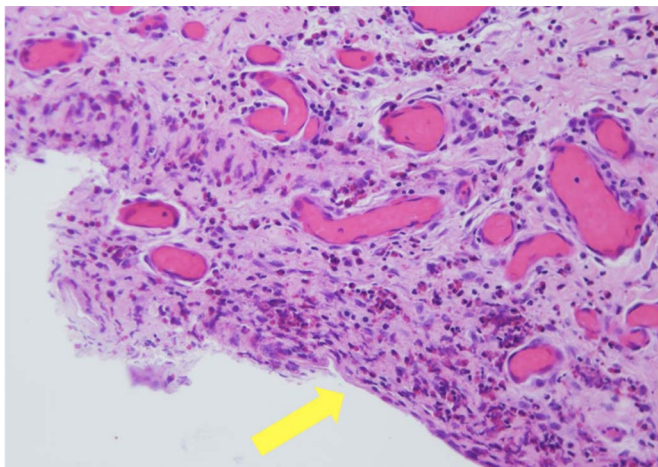


Figure 4. The eosinophilic infiltrate (yellow arrow) extends through the muscularis propria (not pictured) into the subserosa and involves serosal fibrous adhesions (hematoxylin and eosin stain, 200× magnification).

multiple biopsies and elimination of any secondary causes of colonic eosinophilic infiltration.^{1,3}

Therapeutic treatment of EC is not standardized because of the rarity of diagnosis, and current management is based on the limited number of case reports available in the literature. Typical treatment of EC includes dietary modifications, given the common presentation with immunoglobulin E-related food allergens and avoidance of amino acid-based foods. Diet modifications typically involve elimination with slow reintroduction of food groups. In addition to dietary modifications, symptomatic EC can be treated with glucocorticoids and azathioprine through inhibition of eosinophil growth factors.^{2,4} Additional medical intervention includes leukotriene receptor antagonists as well as antihistamines and mast cell stabilizers.^{2,4}

In our patient, the main question of her presentation revolves around EC as the cause of recurrent sigmoid volvulus. The

initial volvulus in 2017 had no evidence of eosinophilic infiltrates on pathology; however, dense transmural infiltration was seen in the 2020 sample. There have been no previous reports on EC-related sigmoid volvulus. Given she had no history of IBD, no food allergens, and no symptoms/history to support parasitic infection, the main differentials for her recurrent volvulus are primary EC vs an allergic reaction and the staples used in her previous anastomosis site. Since the second resection, she has had no gastroenterological symptoms, supporting the 2 remaining differentials rather than a secondary cause of colonic eosinophilia.

DISCLOSURES

Author contributions: K. Zucker wrote article. F. Pradhan, A. Gomez, and R. Nanda edited the article. R. Nanda is the article guarantor.

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Informed consent was obtained for this case report.

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REFERENCES

1. Alfadda A, Storr M, Shaffer E. Eosinophilic colitis: An update on pathophysiology and treatment. *Br Med Bull.* 2011;100(1):59–72.
2. Okpara N, Aswad B, Baffy G. Eosinophilic colitis. *World J Gastroenterol.* 2009;15(24):2975–9.
3. Alfadda A, Storr M, Shaffer E. Eosinophilic colitis: Epidemiology, clinical features, and current management. *Ther Adv Gastroenterol.* 2010;4(5):301–9.
4. Impellizzeri G, Marasco G, Eusebi L, Salfi N, Bazzoli F, Zagari R. Eosinophilic colitis: A clinical review. *Dig Liver Dis.* 2019;51(6):769–73.

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