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CASE REPORT | INFLAMMATORY BOWEL DISEASE

Ulcerative Colitis in Colonic Interposition for Esophageal Atresia

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ABSTRACT

A 38-year-old male with a history of colonic interposition for esophageal atresia as an infant presented with dysphagia and abdominal pain. On the basis of endoscopy findings, pathology, and response to therapy, he was found to have ulcerative colitis of the colonic conduit.

INTRODUCTION

Colonic interposition is one of the best-established treatments for long-gap esophageal atresia. Earlier studies had shown superiority of colonic interposition over gastric pull-up, but a recent meta-analysis showed that colonic interposition and gastric pull-up for treatment of esophageal atresia have comparable results in terms of postoperative mortality, anastomotic complications, and graft loss.[1,2] Long-term studies have shown that the conduit does not show significant changes on exposure to gastric acid.[3] Complications in colonic conduit may arise many years after placement of the graft, prompting long-term follow-up.[4] These complications include graft necrosis, ischemia, stricture, gastric acid reflux, and chest infections. Late complications like graft redundancy, anastomotic stricture, and adhesional obstruction, which require medical or surgical intervention, may arise well after conduit placement.[5-7]

CASE REPORT

A 38-year-old male with chronic hepatitis B carrier state and a remote history of colonic interposition for childhood esophageal atresia in Jordan presented with a 1-day history of dysphagia and abdominal pain. The patient never smoked and was not using any medications at home. The patient was hospitalized 4 weeks prior with abdominal pain and nonbloody diarrhea. On colonoscopy, he was found to have linear and noncircumferential right-sided colonic ulcerations with moderate wall edema and loss of vascular pattern, as well as a few diverticuli in the sigmoid colon. The transverse colon, descending colon, and terminal ileum were normal in appearance. Computed tomography with contrast showed changes consistent with mild right-sided colitis. Due to severe pain and concern of ischemic bowel, the patient underwent exploratory laparotomy, which showed normal intraabdominal viscera. Stool cultures and serum serologies were negative for infectious etiologies including *Shigella*, *Aeromonas*, *Plesiomonas*, *Salmonella*, *Campylobacter*, *Entamoeba histolytica*, and *Cytomegalovirus*. Histology showed acute and chronic inflammatory exudate, architectural distortion, cryptitis, a normal muscularis mucosa, and absent granulomas.

The patient was administered ciprofloxacin and metronidazole upon admission and remained on antibiotics for 6 days until infections were ruled out. With high suspicion of inflammatory bowel disease on biopsy, the patient was given mesalamine (500 mg twice daily). With symptomatic improvement, the patient was discharged home with a recommendation for outpatient follow up with gastroenterology in 2 weeks. The patient returned to the hospital with complaints of dysphagia 2 weeks after discharge. Dysphagia was present for both solids and liquids. Physical examination demonstrated mild tenderness in epigastrium. Electrolytes, blood counts, and hepatic chemistries were normal. Chest, abdominal, and pelvic computed tomography showed only mild wall thickening of the colon.

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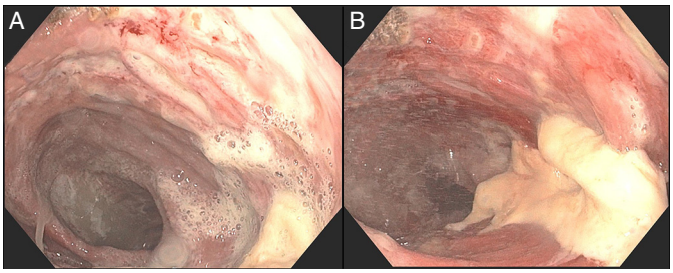

Figure 1. (A) EGD showed diffuse erythema with overlying exudate and mucus. (B) The colonic conduit had regions of severe ulceration but no signs of necrosis on endoscopy.

Esophagogastroduodenoscopy (EGD) showed that the 7-cm segment of native esophagus appeared normal. The esophagocolonic anastomosis was at 25 cm and appeared circumferentially ulcerated and severely strictured. The colonic conduit was diffusely erythematous with overlaying exudate, mucus, and regions of severe ulceration (Figure 1). A cologastric anastomosis at 35 cm appeared normal. The stomach and duodenum appeared normal. The esophagocolonic anastomotic stricture was dilated using controlled radial expansion balloon dilation to 10 mm. Biopsies from the colonic conduit showed colonic mucosa with architectural disarray with crypt distortion, paneth cell metaplasia, acute and chronic inflammatory exudate, and granulation tissue, suggestive of inflammatory bowel disease. Infectious workup including aspirate from colonic conduit for bacterial pathogens (*Salmonella*, *Shigella*, *Campylobacter*, *Aeromonas*, *Plesiomononas*, and *Clostridium difficile*) and biopsy for the herpes simplex virus were negative. Computed tomography angiography did not show vascular stenosis or signs of ischemia in colonic conduit.

The patient was started on oral balsalazide (4.8 g daily), pantoprazole (40 mg daily), and sucralfate (1 mg twice daily). The patient showed some improvement in symptoms, but the symptoms recurred after 2 weeks, at which time budesonide (9 mg daily) was started. With symptoms persisting over the next 3 weeks, the medication was changed to prednisone (40 mg daily) for 2 weeks. The patient's symptoms responded well and resolved in 2 weeks. Follow-up EGD 2 months after diagnosis revealed remarkable improvement in mucosal ulceration and inflammation of the colonic conduit (Figure 2). Colonoscopy performed at the same time showed improved right-sided colonic ulceration and normal terminal ileum. After resolution of symptoms, the patient was lost to follow-up as he did not show up for his appointments.

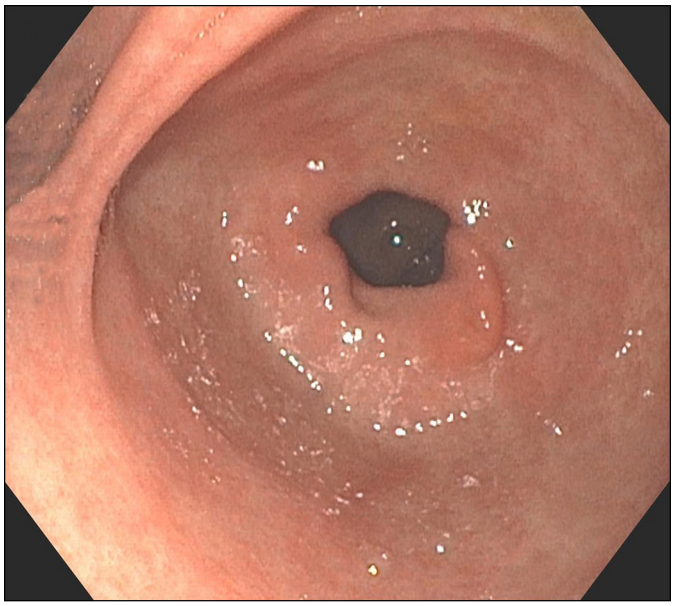

Figure 2. Follow-up EGD after treatment with balsalazide and steroids revealed remarkable improvement in mucosal ulceration and inflammation of the colonic conduit.

DISCUSSION

While the etiology of inflammatory bowel disease is not known, it is thought to arise in part from the combination of exposure to infection or toxins and the genetic susceptibility of the host, combined with defective downregulation of the inflammatory process with subsequent tissue destruction and fibrosis.[8]

In this case, the combined endoscopic and histologic findings of both the colonic conduit and remnant colon, and the exclusion of other causes, were used to determine inflammatory bowel disease to be the most probable diagnosis. Other possible causes of isolated colonic conduit inflammation, as discussed above, include ischemia, graft necrosis, infections, diverticular disease-related inflammation, and gastric acid reflux-induced inflammation. Concomitant inflammation of colonic conduit and remnant colon narrows the differential to concomitant infections and systemic diseases like inflammatory bowel disease or vasculitis.

In this case, features favoring diagnosis of Crohn's disease include focal right-sided colonic ulcerations with preservation of transverse and descending colon. While Crohn's disease is also a possibility, ulcerative colitis was considered to be more likely than Crohn's disease given the lack of extracolonic findings (considering colonic conduit is still colonic tissue histologically with normal rest of the upper gastrointestinal tract on EGD and normal ileum on colonoscopy), the endoscopic appearance of the ulcerations, and the lack of granulomas with normal muscularis mucosa on histology. There have been case reports of ulcerative colitis developing in colonic transpositions for vaginal reconstruction,[9-11] but to the authors' knowledge, there has been no published literature on inflammatory bowel disease in colonic conduits for esophageal atresia.

DISCLOSURES

Author contributions: HMS Arshad wrote the manuscript and is the article guarantor. E. Tetangco and I. Elkhatib reviewed and edited the manuscript.

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Informed consent could not be obtained due to hardship of finding the patient. All identifying information has been removed to protect patient privacy.

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