Filiform polyposis associated with sigmoid diverticulitis in a patient without inflammatory bowel disease

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Abstract

Filiform polyposis (FP) of the colon is an uncommon entity that is occasionally encountered in patients with inflammatory bowel disease (IBD). FP is morphologically characterized by multiple slender worm-like projections consisting of submucosal cores lined with normal mucosa. To date, only two cases of FP have been reported in patients with inflammatory conditions other than ulcerative colitis or Crohn’s disease. We report an additional case of FP occurring in an 83-year-old man with no history of IBD. The patient underwent anterior resection of the sigmoid colon for perforated diverticulitis. Around the diverticular orifice, localized FP involving a 13-cm colonic segment was observed. The filiform polyps consisted of submucosal fibrous cores lined with normal mucosa without epithelial dysplasia. To our knowledge, this is the first reported case of FP associated with colonic diverticulitis in a patient without IBD.

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1. Introduction

Filiform polyposis (FP) of the colon is a distinct and uncommon entity that is occasionally encountered in patients with a history or evidence of inflammatory bowel disease (IBD).1 It is morphologically characterized by multiple slender worm-like projections consisting of submucosal cores lined with normal mucosa. To date, only two cases of FP have been reported in patients with inflammatory conditions other than ulcerative colitis or Crohn’s disease. Here, we report a case of FP associated with sigmoid diverticulitis in an 83-year-old man with no history or evidence of IBD, as well as a review of the current literature.

2. Case report

The patient was an 83-year-old man who initially presented with lower abdominal pain. He did not have any personal or family history of colon polyps, colon cancer or IBD. He denied having nausea, vomiting, diarrhea, melena, hematochezia or...
changes in bowel habits. The physical examination was significant for abdominal tenderness with guarding and rigidity, and laboratory examination revealed an elevation of the white blood cell count to 14,500/μL, with 89.1% segmented neutrophils. Chest roentgenogram and abdominal computed tomographic scan demonstrated free intraperitoneal air beneath the bilateral hemidiaphragms, indicating bowel perforation. During laparoscopic exploration, a perforated diverticulum was discovered in the sigmoid colon. Gross spillage of fecal material and purulent fluid into the peritoneal cavity were also identified. Anterior resection of the colon with a temporary colostomy was performed.

The resected specimen was a 19-cm colonic segment that included the sigmoid colon and upper rectum. Grossly, there was a single 0.8×0.4-cm solitary perforated diverticulum in the proximal sigmoid colon, which was located 0.7 cm from the proximal resection margin. Distal to the diverticular orifice, numerous slender worm-like mucosal projections were identified and ranged in size from 0.2×0.2×0.1 cm to 2.2×0.3×0.2 cm. These filiform polyps had occasional bridging and fusion between adjacent polyps. The involved segment of the proximal and middle sigmoid colon measured 8 cm in length (Fig. 1A). Microscopically, the perforated diverticulum showed expansion of the lamina propria with neutrophilic infiltrates, vascular congestion and subserosal granulation tissue. The filiform polyps consisted of submucosal fibrovascular cores lined by normal mucosa (Fig. 1B). Some of the polyps had dense fibrosis and large-caliber blood vessels in their stalks. No epithelial dysplasia was identified, and the number of chronic inflammatory cells in the lamina propria was minimally increased (Fig. 1C). There was neither evidence of arborizing smooth muscle or hyperplastic mucosa to suggest Peutz–Jeghers polyps nor cystic, dilated glands or expansion of the lamina propria to suggest juvenile polyps. There were no increases in smooth muscle fascicles, nerves or ganglion cells. The intervening mucosa showed minimal lymphocytic infiltrate with a few lymphoid aggregates in the lamina propria.

Seven months later, the patient underwent colostomy take-down. The resected specimen, measuring 12 cm in length, also showed approximately 20 slender, worm-like mucosal projections near the distal resection margin, which involved a 5-cm segment of the distal descending colon (Fig. 1D). The histologic features were identical to those of the filiform polyps found in the sigmoid colon. No epithelial dysplasia was observed.

3. Discussion

The term FP was first coined by Appelman et al.4 who used it to describe a syndrome involving the radiographic appearance of numerous long slender worm-like or filiform defects in the colon with a normal haustral pattern. FP often presents as multiple mucosal projections that can reach up to 9 cm in length and can have bridging between adjacent polyps.1,4,5 FP

![Figure 1](image-url)
can be localized, or it can involve the entire length of the colon; diffuse colonic FP can endoscopically mimic familial adenomatous polyposis. Furthermore, numerous congested polyps may give the appearance of a fungating mass; hence, it has been confused with cancer on colonoscopy and radiologic studies. Although the presence of dysplasia or malignant transformation has never been reported in FP, in some cases, the polyps are difficult to distinguish from villous adenomas, and biopsies are needed to make the diagnosis. The pathogenesis of FP is uncertain, but since the majority of cases occur in the setting of IBD, it is generally believed to form as a result of an exuberant post-inflammatory reaction.

Although FP typically occurs in patients with IBD, it is important to realize that rare cases have been documented in patients without IBD. To date, there have been two cases of FP occurring in association with inflammatory conditions other than ulcerative colitis or Crohn’s disease. One case, reported in 1985, was that of a 60-year-old woman with a history of Langerhans cell histiocytosis X who presented with intermittent bloody diarrhea and abdominal pain. A double-contrast barium enema showed multiple filiform filling defects throughout the colon. A colonoscopy revealed that, in addition to polyps, there were superficial ulcerations and inflammatory changes in the mucosa, which histologically consisted of numerous atypical histiocytes admixed with eosinophils and lymphocytes. Another case, reported in 1988, was that of a 22-year-old woman with pulmonary and colonic tuberculosis who presented with diarrhea and abdominal pain. A double-contrast barium enema demonstrated multiple worm-like branching filling defects in the transverse colon, characteristic of FP, as well as the ulceration, deformation and contracture that is typical of tuberculosis. The patient responded well to antituberculous medication, but FP was still present 11 months after the ulcers had resolved. The present case is the first reported case of FP associated with perforated diverticulitis of the sigmoid colon in a patient without IBD.

There was a previous report describing a case of localized FP with multiple deep diverticula in a patient with active ulcerative colitis. The authors described polyps that involved the proper muscle layer and showed a budding appearance with diffuse neutrophilic and plasmacytic infiltrates and partial loss of the submucosa. It was presumed that the pathogenesis of the deep diverticular lesions with FP was probably due to the repair of the regenerative mucosa after deeper ulcerations due to active ulcerative colitis. In contrast, in the present case, the polyps and intervening mucosa had no evidence of features that would suggest IBD such as acute inflammation, ulceration, granulomas, architectural distortion of crypts or chronic mucosal injury. Based on these findings, we propose that the pathogenesis of FP without IBD may not be related to the post-inflammatory reparative process; instead, it might be a localized response to cytokines and growth factors generated by the inflamed diverticular mucosa.

Diverticulitis-associated FP should be distinguished from prolapsing mucosal folds, which are often found in the peridiverticular mucosa. These folds typically have mucosal edema with crypt hyperplasia and elongation, muscularized lamina propria, hypertrophic muscularis mucosae, hemosiderin deposition or superficial erosions. Normal to minimally inflamed mucosa without changes in cryptal architecture and muscularis mucosae exclude the possibility of prolapsing mucosal folds.

Interestingly, we observed residual FP in the specimen obtained at colostomy take-down seven months later. Together with the previous finding that FP remained after the resolution of tuberculosis, this observation raises the possibility that elimination of the causative inflammatory condition may not bring about a spontaneous resolution of FP.

In summary, we present a case of localized FP associated with perforated diverticulitis of the sigmoid colon. To our knowledge, this is the first reported case of diverticulitis-associated FP in a patient without IBD. It is important to recognize that FP can be associated with non-IBD intestinal inflammation. When encountering FP in a patient without IBD, awareness of this type of lesion can prompt physicians to look for a causative inflammatory condition.

References