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Single Case

Cap Polyposis Syndrome Imitating Inflammatory Bowel Disease with Polyps Extending to the Terminal Ileum

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Keywords

Cap polyposis syndrome · Inflammatory cap polyposis · Mucosal prolapse syndrome

Abstract

Introduction: Cap polyposis syndrome is a rare subtype of mucosal prolapse disease characterized by erythematous, inflammatory colonic polyps covered by a cap of fibrinopurulent mucous. Although a benign condition, patients may present with significant symptoms that can be suggestive of inflammatory bowel disease or colorectal cancer. ***Case Presentation:*** We describe the case of a 34-year-old male who presented with a 5-month history of diarrhea and 40-lb weight loss following hospitalization for enterotoxigenic *Escherichia coli* colitis. The patient had a past medical history significant for prior colonoscopy revealing hundreds of polyps and a father who died of colorectal cancer at age 45. Multiple repeat infectious stool workups were negative, and antibiotics failed to resolve the patient's symptoms. The patient underwent endoscopy which revealed numerous polyps from the rectum to the terminal ileum that appeared similarly to pseudopolyps giving concern for inflammatory bowel disease. Subsequent histology demonstrated surface erosion and inflammation without dysplasia. Review of endoscopy showed inflammatory polyps with a cap of fibrinopurulent mucous. In the absence of chronic inflammation (C-reactive protein was within normal limits following hospitalization), endoscopic and histologic findings were suggestive of cap polyposis syndrome. ***Conclusion:*** Cap polyposis is diagnosed endoscopically and histologically. While most cases of cap polyposis are confined to the distal colon and rectum, we believe that this is the first case of cap polyposis syndrome extending to the terminal ileum. Treatment of cap polyposis syndrome is dependent on the severity of symptoms.

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Introduction

Cap polyposis syndrome is a rare condition characterized by erythematous, inflammatory colonic polyps covered by a cap of fibrinopurulent mucous [1]. Patients have a nonspecific clinical presentation that may mimic inflammatory bowel disease or colorectal cancer. Patients usually present with rectal bleeding, mucoid diarrhea, and abdominal pain [2] with normal C-reactive protein (CRP) levels [3]. However, some patients may present asymptomatically [4]. Diagnosis requires colonoscopy and subsequent histological evaluation. Treatment is dependent upon severity of the patient's symptoms.

Case Presentation

Patient is a 34-year-old male with past medical history of hypertension, hyperlipidemia, and psoriasis who presented for clinic follow-up after a 5-day hospital admission for enterotoxigenic *Escherichia coli* colitis treated with azithromycin. Hospitalization was complicated by prolonged diarrhea and acute kidney injury, nearly requiring dialysis. He had persistently loose stools, a 40-lb weight loss, and intermittent lower abdominal pain for 4 months following discharge. His primary care physician prescribed a course of cefixime which failed to provide relief. Repeat infectious stool workups, including repeat *E. coli*, *Clostridium difficile*, and stool ova and parasite were negative, and ultimately, the patient's symptoms resolved.

The patient has a family history significant for colon cancer in his father diagnosed at age 45. He received a colonoscopy at an outside hospital 1 year prior to his hospitalization and had over 100 hyperplastic polyps in the rectosigmoid colon and one adenomatous polyp in the descending colon. The patient was instructed to repeat his colonoscopy in 1 year. At his follow-up colonoscopy (5 months following his hospitalization and 1 month following resolution of symptoms), numerous polyps were noted in the terminal ileum and throughout the colon but spared the rectum.

Endoscopically, the polyps appeared similarly to pseudopolyps giving concern for the presence of inflammatory bowel disease. Several cold snare polypectomies were performed throughout the colon revealing inflammatory polyps. The polyps were nonneoplastic lesions. The patient has no extraintestinal clinical features (i.e., ocular changes, dermatological findings, or arthralgias), no evidence of chronic inflammation (no reactive thrombocytosis and CRP = 0.5 following hospitalization), and the remainder of his laboratories were unremarkable. Gross images of the colonic inflammatory polyps (Fig. 1) were noted to have a cap of fibrinopurulent mucous, a specific finding for cap polyposis. Histology of the inflammatory polyps (Fig. 2) demonstrated surface erosion, inflammation, and vascular congestion without dysplasia of the epithelium and crypts. There were no non-caseating granulomas, altered crypt architecture or abscesses, giant cells, or lymphocytic infiltration suggestive of IBD. None of the samples showed histologic findings consistent with colitis, ileitis, or adenomatous changes. The endoscopic and histological features in the absence of inflammatory bowel disease are suggestive of cap polyposis. The patient denied rectal bleeding, severe diarrhea, and weight loss; thus, there was no indication for surgery or pharmacological management. The patient was also advised to repeat colonoscopy in 2–3 years based on his clinical course and family history of a first degree relative with colorectal cancer as well as genetic testing due to multitude of polyps.

The patient was seen by gastroenterology regularly following his initial endoscopy and received a repeat endoscopy 2 years later. In the interim, he continued to have normal stools without hematochezia, unintentional weight loss, or lower abdominal pain. Repeat

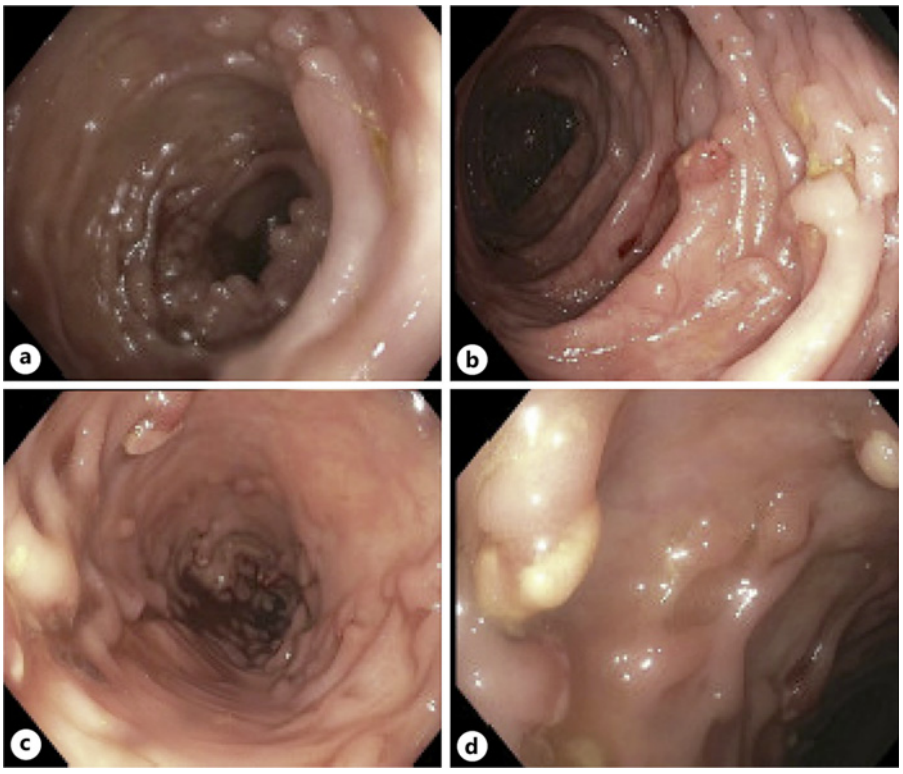

Fig. 1. Inflammatory polyps covered with a cap of fibrinopurulent mucous located in the terminal ileum (**a**), transverse colon (**b**), descending colon (**c**), and sigmoid colon (**d**). This figure was previously published in the American Journal of Gastroenterology and has been reproduced with permission from the author and publisher [5].

endoscopic and histologic findings were stable and consistent with cap polyposis syndrome. Endoscopically, multiple polyps were identified in the terminal ileum and pseudopolyps were present throughout the entire colon, but overall assessment was limited due to large volumes of liquid stool. Multiple polypectomies were performed during the procedure, and histologically, there were no signs of colitis, ileitis, or adenomatous changes. The patient was instructed to repeat endoscopy in 1 year due to suboptimal bowel preparation and obtain genetic testing for familial colon cancer.

Discussion

Cap polyposis syndrome is a subtype of mucosal prolapse syndrome and is an extremely rare condition occurring in <1 in 1,000,000 individuals worldwide [6]. The pathophysiology of cap polyposis syndrome is unclear; however, several contributing factors have been postulated including *Helicobacter pylori* infection [3, 7], altered gut microbiome [8], chronic inflammation (through TNF-α) [9], mucosal prolapse [10], mechanical stimulation by abnormal bowel motility [11], repeated trauma to the colonic mucosa secondary to straining [3, 7], and following pelvic surgery [12].

The clinical presentation of cap polyposis syndrome is nonspecific and can present asymptomatically [4] or may mimic more sinister conditions such as inflammatory bowel disease [13] or colorectal cancer [14]. Patients most commonly present with abdominal pain, mucoid diarrhea, and rectal bleeding [2]. Patients may also present with weight loss,

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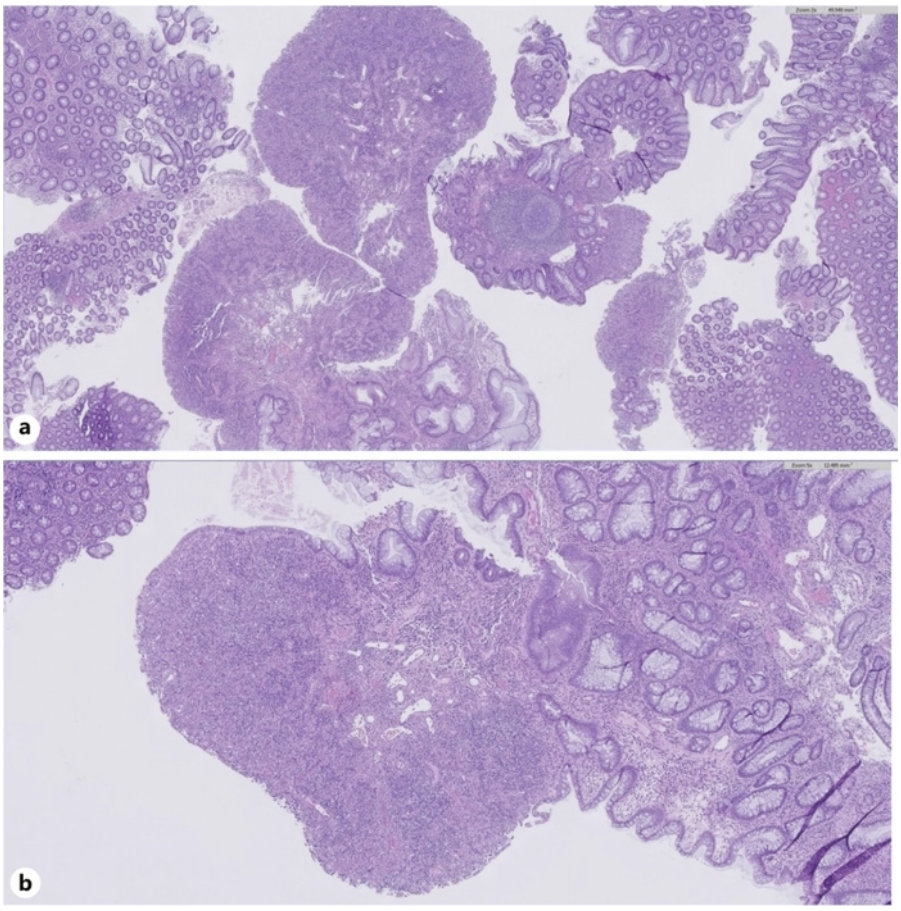

Fig. 2. a, **b** Multiple polyps are seen in the colon with histological features of surface erosion, inflammation, vascular congestions without dysplasia of the epithelium and crypts. This figure was previously published in the American Journal of Gastroenterology and has been reproduced with permission from the author and publisher [5].

tenesmus, and constipation [9]. Lower extremity edema can occur due to a protein-losing enteropathy [11]. Laboratory values may reveal an iron deficiency anemia (secondary to rectal bleeding), hypoproteinemia (from protein-losing enteropathy), and a normal C-reactive protein [4]. Similarly, our patient presented with chronic weight loss, diarrhea, and abdominal pain with a normal CRP. Prior to diagnosis, his symptoms resolved entirely, and he has remained asymptomatic.

Cap polyposis syndrome is definitively diagnosed with colonoscopy and histology. Polyps in patients with cap polyposis are commonly found in the distal colon and rectum and appear erythematous with an adherent mucoid cap on endoscopy [3]. While cap polyposis has been documented in the cecum [4], we believe our patient demonstrates the first case of cap polyposis extending from the colon to the terminal ileum. Histology of cap polyposis shows nonneoplastic lesions with elongated and tortuous glands, a mixed inflammatory infiltrate containing smooth muscle fibers in the lamina propria, and a cap of inflammatory granulation tissue with fibrinopurulent exudate that covers the polyps [1].

There is no official recommendation for treatment of cap polyposis syndrome. Management is dependent upon severity of symptoms. Asymptomatic patients require no treatment and should schedule their follow-up colonoscopy according to their pertinent colorectal cancer screening guidelines. Symptomatic patients with *H. pylori* infection display improved symptoms

following *H. pylori* eradication therapy [3, 7]. Symptomatic patients with a small number of polyps can undergo snare polypectomy for resection. Symptomatic patients with a large number of polyps require colectomy [2]. The role of anti-inflammatory agents in treating cap polyposis is less certain; however, there has been success using corticosteroids [15] and TNF-α inhibitors such as infliximab [9]. This may be considered in the patient presented if he develops symptoms in the future. Spontaneous resolution of cap polyposis has also been reported in several cases [16].

Conclusion

Cap polyposis syndrome is a rare condition characterized by inflammatory, fibrinopurulent polyps. In this report, we presented a patient presenting with clinical features concerning inflammatory bowel disease; however, upon further histologic evaluation, a diagnosis of cap polyposis syndrome was more consistent with his presentation. This is the first case in which cap polyposis was found in the terminal ileum. Cap polyposis is an important consideration in patients presenting with symptoms suggestive of inflammatory bowel disease, but without findings of systemic inflammation. Recognizing the findings on colonoscopy is critical to prevent misdiagnosis and subsequent treatment on patients with this benign condition. The CARE Checklist has been completed by the authors for this case report, attached as online supplementary material (for all online suppl. material, see https://doi.org/10.1159/000545182).

Statement of Ethics

This study protocol was reviewed and the need for approval was waived by the Institutional Review Board at University of Texas Medical Branch at Galveston. Written informed consent was obtained from the patient for publication of the details of their medical case and any accompanying images.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Author Contributions

G.T. Coleman designed and wrote the manuscript. R. Dayah, S. Qiu, and G. Luthra critically reviewed and revised the manuscript. All authors reviewed and approved of the final version.

Data Availability Statement

All data generated or analyzed during this study are included in this article and its online supplementary material files. Further inquiries can be directed to the corresponding author.

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