Case Report

Fresh Bloody Discharge from Rectal Cap Polyposis: A Rare Case Report and Literature Review

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Cap polyposis is a rare gastrointestinal disease also occurring elsewhere in the colorectum. Mostly, it appears in the distal colon and rectum, characterized by erythematous, inflammatory colonic polyps covered by a cap of fibrinopurulent mucous. The pathogenesis of cap polyposis is not well understood, but the pathological hypothesis includes infection, mucosal ischemia or T cell-mediated inflammation or mechanical stimulation by abnormal bowel motility and repeated trauma to the colonic mucosa caused by straining. The exact incidence is unknown, but the incidence of age reports range from 11 months to 76 years. As most patients with cap polyposis appear asymptomatic, diagnosis is difficult, and symptomatic patients usually present unspecific symptoms such as rectal bleeding, mucoid diarrhea, abdominal pain, tenesmus, weight loss and constipation. Herein, we present a 25-year-old male patient presenting with 3-month rectal bleeding where multiple rectal sessile polyps were found via colonoscopy and managed successfully with transrectal polypectomy.

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Key Words
Rectal cap polyposis;
Colonoscopy;
Transrectal polypectomy;
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Cap polyposis (CP) is a rare and under-recognized condition with distinct clinical, endoscopic and histopathological features. It was first described by William et al.[1] in 1985, and is characterized by inflammatory polyps usually located from the rectum to the distal descending colon. Most cases occur during the fifth decade of life with intermittent frequency,[2] although the exact incidence and pathogenesis are unknown. The most common symptoms include constipation, mucous discharge, diarrhea, abdominal pain and rectal bleeding[3-5] that are unspecific and overlap with inflammatory bowel disease, making cap polyposis difficult to diagnose.

Rectal bleeding combined with the presence of a

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rectal mass has been traditionally associated with presence of malignant disease, although cap polyposis is a relatively young and still undefined rare entity that mainly involves the rectosigmoid. Diagnosis is usually set by exclusion of ulcerative colitis after poor response to mesalamine and steroids where surgical resection is considered the optimal treatment.[3] We herein present a case of a 25-year-old male who presented with unexplained chronic long-standing anemia secondary to intermittent rectal bleeding where histopathological examination confirmed it to be a case of CP of the rectum.

Case Report

This 25-year-old male had no medical history other than anemia. He presented with fresh blood in his stool intermittently over a 3-month period. He visited our outpatient department (OPD) and the associated symptoms were occasional abdominal fullness and pain although he denied suffering from diarrhea or constipation. Digital rectal examination showed a palpable mass in the lower rectum, so colonoscopy was arranged, revealing multiple sessile polypoid lesions over the rectum that were partially removed by polypectomy for complete pathological examination (Fig. 1). Biopsy was performed revealing inflammatory polyps with dilated or degenerated crypts with abundant mucoid deposits and inflammatory exudate simulating rectal cap polyp or polyposis (Fig. 2). After discussion with the patient, transrectal polypectomy was performed (Figs. 3-5), with the pathology report indicating compatibility with rectal cap polyposis. After the operation, the patient recovered well and has had regular follow-up in our OPD with no further rectal bleeding being found. He has also received a colonoscopy every year following the surgery, with no sign of recurrence till now.

Discussion

Colorectal cap polyposis is a rare condition characterized by erythematous, inflammatory colonic polyps covered by a cap of fibrinopurulent mucous. It always presents with multiple sessile polyps occurring in distal areas like the sigmoid colon and rectum. Cap polyposis is generally known as a benign lesion, and there is no documented malignant potential of these polyps[6] although the exact incidence is unknown because cases in both males and females are rarely reported, while still lacking statistical significance. The age of re-

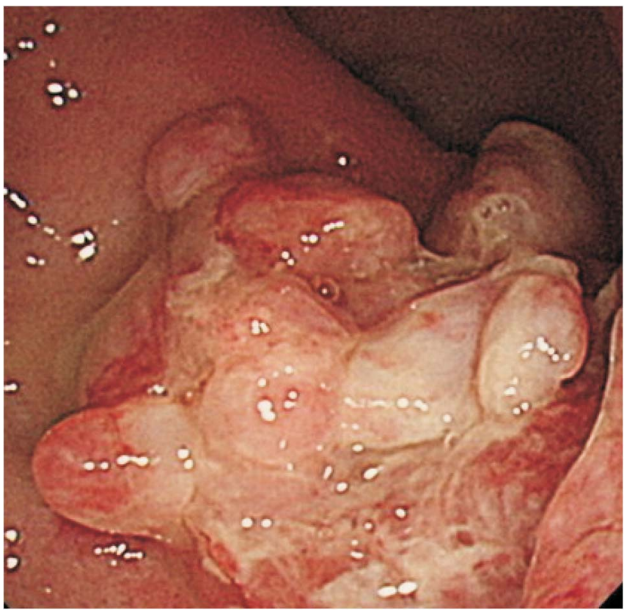

Fig. 1. Colonoscopic picture: multi-polyp cover with an erythematous surface and a yellow-white coating under colonoscopy.

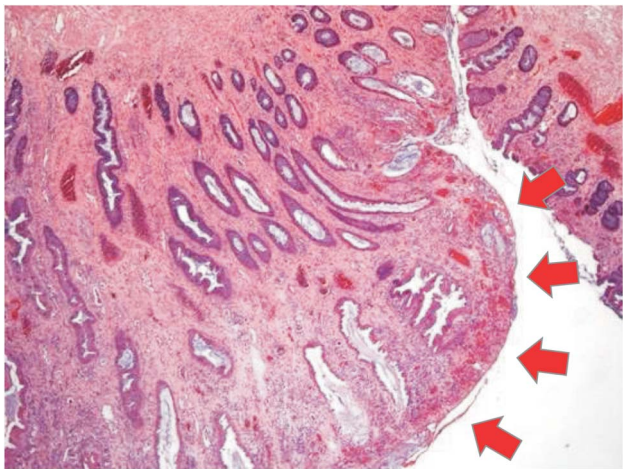

Fig. 2. Pathological section: lower-power view of cap polyposis showing elongated hyperplastic appearing glands and a cap of overlying mucosa composed of mucus, fibrin, and leukocytes (red arrow).

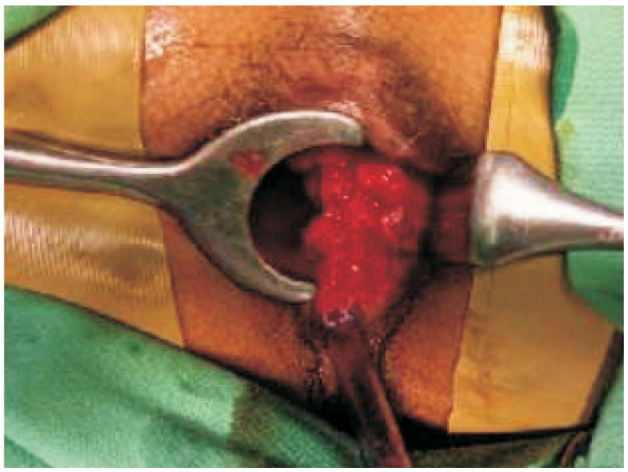

Fig. 3. Intra-operative picture: multiple sessile polypoid lesions were found during operation over rectum 3 cm from anal verge.

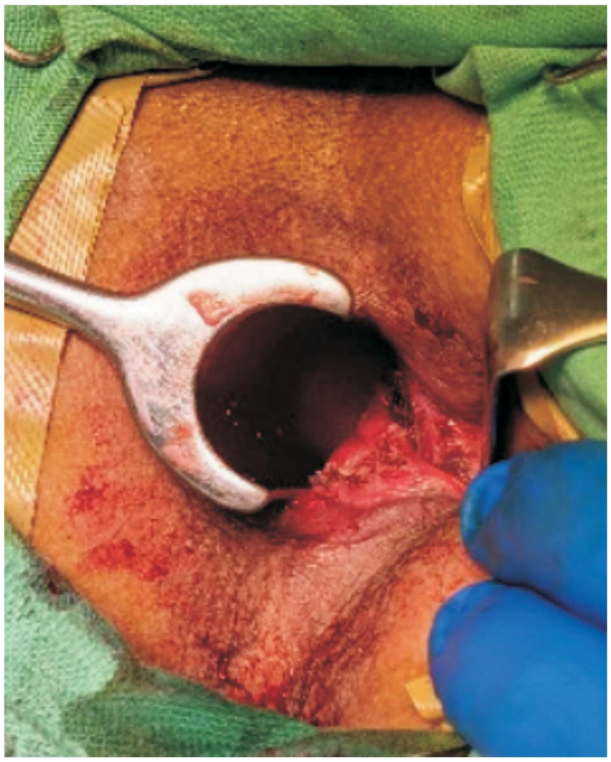

Fig. 4. Post-operative picture: surgical wound after transanal rectal polypectomy.

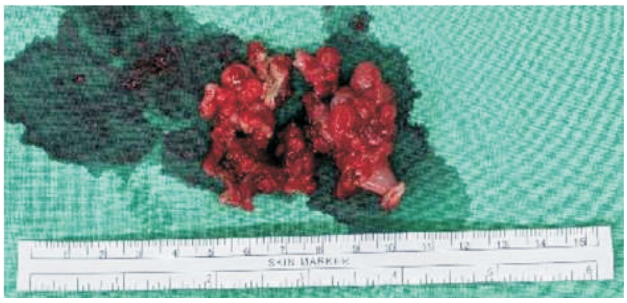

Fig. 5. Gross view of specimen: rectal cap polyps.

ported cases has ranged from 11 months to 76 years.[2,6]

The pathogenesis of cap polyposis is not well understood, with hypothesized causes including infection, mucosal ischemia, T cell-mediated inflammation, mechanical stimulation by abnormal bowel motility, and repeated trauma to the colonic mucosa caused by straining when defecating. Microscopically, the polyps consist of elongated, tortuous and distended crypts covered by a "cap" of inflammatory granulation tissue.

Patients with cap polyposis might be mostly asymptomatic as symptomatic patients usually present with rectal bleeding (82%), followed by mucoid discharge (46%), constipation, diarrhea and abdominal pain.[6,8] Other symptoms include tenesmus, straining during defecation, the sensation of incomplete defecation, the need for manual stool evacuation, weight loss and protein-losing enteropathy have also been reported,[5,7] with the symptoms appearing present for weeks to months prior to the diagnosis. Laboratory studies are typically normal, and diagnosis is always difficult because the symptoms overlap with inflammatory bowel disease, which causes delay and inappropriate treatment for such patients, although cap polyposis can be directly diagnosed by colonoscopic evaluation and confirmed by biopsy. It can be found anywhere in the colon but is mostly discovered in the rectosigmoid colon where single or multiple polyps varying in size and shape might be observed.

Treatment for asymptomatic patients is unnecessary. Treatment for symptomatic patients includes endoscopic polypectomy and colectomy according to severity and distribution of the polyps.[4] Snare polypectomy with argon plasma coagulation or endoscopic mucosal resection is recommended, and is associated with complete resolution of symptoms. Some reports have shown polyposis regrowth for patients who received polypectomy, especially in patients with multiple polyps. Repeated colonoscopy has been recom-

mended for these patients, while colectomy is indicated for those with multiplicity of polyps precluding endoscopic resection. Complete colonoscopy for the entire colon should be performed prior to surgery to make sure no other colonic disease coexists. Some case reports have revealed successful treatment with amino-salicylates,[17] anti-inflammatory agents (eg., topical and systemic steroids),[12,13] antibiotics (eg., metronidazole)[10,11] and immunomodulators (eg., infliximab),[8,14,15] implying a possible cause of inflammation in the pathogenesis of cap polyposis; however, the evidence is not sufficient to support these treatment strategies.

Interestingly, a possible relationship between *Helicobacter pylori* infection and cap polyposis of the colon was proposed by Akamatsu et al. in 2004, who demonstrated that some patients with cap polyposis receiving eradication therapy for *Helicobacter pylori* infection also showed endoscopic improvement of cap polyposis. All of these patients had been diagnosed with *H. pylori* infection in the stomach; however, *Helicobacter pylori* was not detected in the biopsy specimens from the colonic inflammatory polyps.[9]

Conclusion

Cap polyposis is a rare condition with unknown incidence and pathogenesis with the most common symptoms including rectal bleeding and mucoid diarrhea. Medication treatment is an option, especially in children, and for those symptomatic cases and patients failing in medication treatment or recurrence, surgical intervention with endoscopic polypectomy should be recommended. Colectomy is indicated for patients with multiplicity of polyps that makes endoscopic polypectomy inappropriate. Testing for *Helicobacter pylori* infection is also recommended because of the efficacy of eradication therapy. The outcome of cap polyposis is not clear because of the small case number with limit long-term follow-up data. Moreover, chronic recurrent symptoms requiring multiple treatments with polypectomy as well as spontaneous resolution, have also been reported.[13,16]

Ethical Approval

The study was approved by the Institutional Review Board of Kaohsiung Medical University Hospital (KMUHIRB-E(I)-20200036).

Declaration of Patient Consent

The authors certify that they have obtained all appropriate patient consent forms where the patient has given his consent for the images and other clinical information to be reported in the journal while understanding that his name and initials will not be published and all due efforts will be made to conceal his identity, but anonymity cannot be guaranteed.

Data Availability Statement

Data sharing is not applicable to this article because no data sets were generated or analyzed during the current study.

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Conflicts of Interest Statement

The authors declare that they have no conflict of interest with regard to the content of this article.

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病例報告

導因於直腸帽狀瘜肉症所引起血便：一個罕見案例報告及文獻回顧

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帽狀瘜肉症是一種少見的胃腸道疾病，它有可能出現在結直腸任何一處。帽狀瘜肉症常見於遠端結腸及直腸，主要特徵為紅斑性，發炎性的大腸瘜肉包覆於帽狀膿性纖維蛋白黏液下。病理機轉不明，目前認為有可能之原因為感染、黏膜缺血、T 細胞介導的發炎、腸蠕動異常引起的機械性刺激、過度用力引起的結腸黏膜反覆損傷。確切的盛行率不明，但是年齡大都介於 11 個月到 76 歲之間。帽狀直腸瘜肉症的診斷十分不容易，主要原因為大部份病人可能無症狀。我們在此報告一例 25 歲年輕男性，因為持續 3 個月的直腸出血，經大腸鏡診斷出帽狀直腸瘜肉症，最後通過經直腸瘜肉切除術成功治療這位病患，並整理回顧相關文獻。

關鍵詞 帽狀直腸息肉症、大腸鏡、經直腸息肉切除術、病例報告、文獻回顧。