CASE REPORT

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Remission of Cap Polyposis Maintained for More Than Three Years after Infliximab Treatment

Eun Sun Kim, Yoon Tae Jeen, Bora Keum, Yeon Seok Seo, Hoon Jai Chun, Soon Ho Um, Chang Duck Kim, and Ho Sang Ryu

Department of Internal Medicine, Korea University College of Medicine, Seoul, Korea

Cap polyposis is a rare disorder with characteristic endoscopic and histological features; its etiology is still unknown, and no specific treatment has been established. We report a case of cap polyposis that improved remarkably after infliximab infusion and had no recurrence for 3 years. **(Gut and Liver 2009;3: 325-328)**

Key Words: Cap polyposis; Infliximab; Therapeutics; Long term follow up

INTRODUCTION

Cap polyposis is a rare but distinct disorder with characteristic endoscopic and histological features.[1,2] It is characterized by multiple distinctive erythematous, inflammatory colonic polyps located from the rectum to the distal colon. And the polyps are covered with fibrinopurulent mucus which appears like a 'cap.' The common symptoms are mucous and bloody diarrhea with abdominal pain and tenesmus. The etiology of this disease is still unknown, and no specific treatment has been established. There have been a few reports about the cases of cap polyposis responsive to infliximab.[3] Herein we report a cap polyposis that was remarkably improved after a single infliximab infusion and had no recurrence for 3 years.

CASE REPORT

A 58-year-old woman was admitted to our hospital because of mucous bloody stools, frequent defecation and tenesmus for 2 weeks. One month ago, the patient had been managed in other hospital with 2nd generation cephalosporin antibiotics because of community acquired pneumonia. On physical examination, abdomen was soft and there was no tenderness or palpable mass. Hemoglobin was 14.5 g/dL, white blood cell count was 6,380/mm^3, platelet count was 319,000/mm^3 and data of C-reactive protein or erythrocyte sedimentation rate were not increased. Stool occult blood test was positive, but, *Clostridium difficile* antigen assay of stool was negative.

Colonoscopy showed about 20 reddish sessile polyps covered with white purulent exudates, and scattered hyperemia on rectum and sigmoid colon. The polyps were located on apices of mucosal fold (Fig. 1). The histological finding of sessile polyp indicated chronic and acute inflammations with acute cryptitis. We first diagnosed pseudomembranous colitis based on patient's history of antibiotics administration and colonoscopic finding. However, there was no clinical symptom improvement after oral administration of 250 mg metronidazol qid for 3 weeks.

The colonoscopic finding for follow up showed no improvement, and additional biopsy was performed. Histological finding showed that the polyps were consisted of elongated, tortuous, and hyperplastic crypts that attenuated toward the surface (Fig. 2). Heavy infiltration of inflammatory cells, ulcerated mucosal surface and fibrinopurulent exudates are characteristic of the so-called "cap polyp." On the basis of these characteristic colonoscopic and histologic findings, therefore, the patient was diagnosed with cap polyposis.

We considered conservative management and bowel ha-

Correspondence to: Yoon Tae Jeen
Department of Internal Medicine, Korea University College of Medicine, 126-1, Anam-dong 5-ga, Seongbuk-gu, Seoul 136-705, Korea
Tel: +82-2-920-6555, Fax: +82-2-953-1943, E-mail: ytjeen@korea.ac.kr
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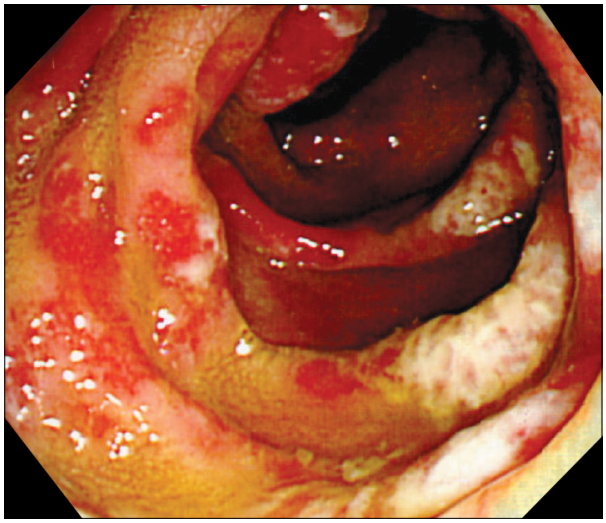

Fig. 1. Sessile polyps with exudates and hyperemia in the sigmoid colon.

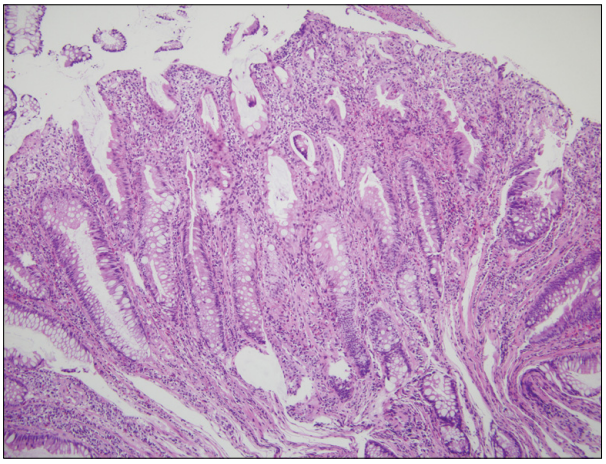

Fig. 2. Histological findings. The polyps comprise elongated, tortuous, and hyperplastic crypts that attenuate toward the surface. Heavy infiltration of inflammatory cells, an ulcerated mucosal surface, and fibrinopurulent exudates are characteristic of the so-called "cap polyp" (H&E stain, ×100).

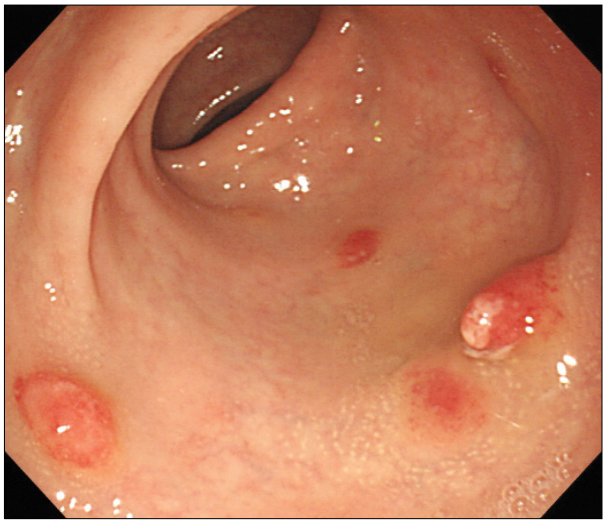

Fig. 3. Colonoscopy conducted 4 weeks after infliximab infusion revealed reductions in the size and number of the sessile polyps.

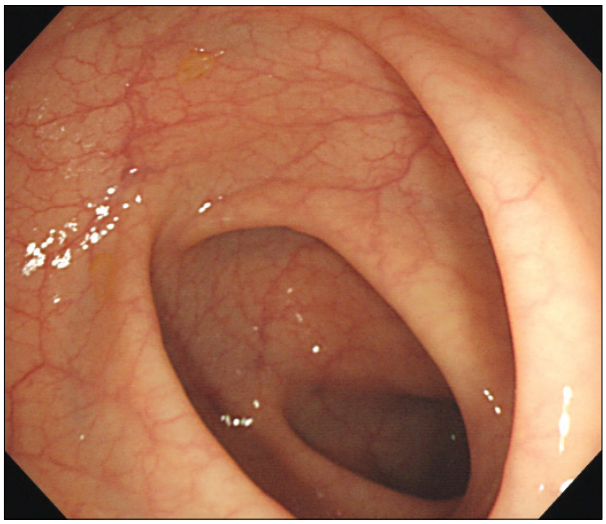

Fig. 4. Follow-up colonoscopy conducted 36 months after the single infusion of infliximab, revealing maintenance of the 4-week postinfusion state (i.e., no recurrence of cap polyposis).

bitual correction to avoid straining at defecation. After 12 months of these managements, there was no improvement of clinical symptom and colonoscopic finding. Then, we considered surgical management or infliximab infusion. Therefore, we first administered 5 mg/kg dose of infliximab. At 7th days, following the infliximab infusion, clinical improvement occurred. After 4 weeks of infliximab infusion, colonoscopy revealed that the multiple sessile polyps decreased in size and numbers (Fig. 3), and no side effect of infliximab was observed. Therefore, we decided to follow up the patient with no additional administration of infliximab. For 3 years, the patient experienced no clinical symptom recurrence, and the last colonoscopy revealed almost complete mucosal recovery except for tiny scant scars (Fig. 4).

DISCUSSION

Common clinical manifestation of cap polyposis is mucous bloody diarrhea lasting for weeks to months, and women are mostly afflicted. Tenesmus, rectal bleeding, abdominal pain, constipation, weight loss, and hypoproteinemia have also been reported.[3,4] Epidemiology and etiology of cap polyposis have not been well known.

Several suggestions have been made on its pathogenesis, including a form of inflammatory bowel disease, an infectious origin such as *Helicobacter pylori* or *Escherica coli* 018, improvement after antibiotics treatment,[5,6] whereas other suggested on association with mucosal prolapse syndrome or abnormal colonic motility resulting in local

Table 1. Case Reports of Cap Polyposis and Treatments

Case	Gender	Age	Symptom	Location	Management	Follow up duration	Result
Campbell *et al.* (1993)[2]	Male	68	Weight loss, diarrhea	Sigmoid colon	Total colectomy	−	Improved
	Female	65	Diarreha	Rectum	Cleversal enema	−	Improved
Gehenot *et al.* (1994)[6]	Female	42	Bloody diarrhea	Rectosigmoid	Sigmoid colostomy	9 months	Resolved
Shiomi et al. (1998)[7]	Female	54	Hypoproteinemia, diarrhea	Descending colon	Left hemicolectomy	−	Resolved
Oriuchi et al. (2000)[8]	Female	20	Hypoproteinemia, mucous diarrhea	Rectosigmoid	Avoidance of straining	4 years	Impreved
	Female	52	Hypoproteinemia, mucous diarrhea	Rectosigmoid	Colostomy for avoid constipation	−	Improved
Kajihara et al. (2000)[9]	Female	38	Bloody diarrhea	Rectosigmoid	Metronidazole	2 months	Resolved
Isomoto *et al.* (2001)[10]	Female	51	Mucous, bloody diarrhea	Rectosigmoid	Sigmoid colostomy	1 year	Resolved
Esaki *et al.* (2001)[11]	Male	21	Weight loss, bloody diarrhea	Rectosigmoid	Metronidazole	6 months	Transiently improved
	Female	67	Abdominal pain, mucous diarrhea	Rectosigmoid	Proctosig-moidectomy	18 months	Resolved
	Male	21	Abdominal discomfort	Rectum	Refuse	-	Improved (only symptom)
	Female	76	Mucous diarrhea, tenesmus	Rectosigmoid	Refuse	22 months	Sustained tenesmus
Sadamoto *et al.* (2001)[12]	Male	73	No symptom	From sigmoid to cecum	No treatment	4 months	Resolved
Oiya *et al.* (2002)[13]	Male	63	Mucous diarrhea	From rectum to ascending colon	*Helicobacter* eradication	8 months	Resolved
Shimizu *et al.* (2002)[5]	Female	12	Mucous bloody diarrhea	Rectosigmoid	Metronidazole	12 months	Resolved
Park *et al.* (2002)[14]	Female	60	Abdominal pain, tenesmus	Rectum	Surgical resection	6 months	Resolved
Ohkawara *et al.* (2003)[15]	Female	67	Mucous bloody diarrhea	Rectosigmoid	No treatment	12 months	Resolved
Akamatsu *et al.* (2004)[16]	Female	33	Hypoproteinemia, mucous bloody stool	Rectum	*Helicobacter* eradication	18 months	Resolved
	Female	50	Weight loss, mucous bloody stool	Rectosigmoid	*Helicobacter* eradication	26 months	Resolved
	Female	53	Mucous bloody stool	Rectosigmoid	*Helicobacter* eradication	15 months	Resovled
Bookman *et al.* (2004)[3]	Female	36	Abdominal pain, mucous bloody diarrhea	Rectosigmoid	Infliximab infusion	38 months	Resolved
Konishi *et al.* (2005)[17]	Female	76	Hypoproteinemia, mucous bloody stool	Total colon	Sigmoid colostomy	3 months	Resolved
Maunoury *et al.* (2005)[18]	Female	52	Abdominal pain, mucous diarrhea	Rectum	Imfliximab infusion	−	Failed
Ryu *et al.* (2006)[19]	Male	64	Weight loss, diarrhea	Rectosigmoid	Conservative	1 year	Resolved

ischemia and recurrent mucosal trauma.[5,6] Diagnosis of this disease in the present case was established through colonoscopic finding, clinical manifestation and histological finding. The endoscopic finding showed erythematous polyps with adherent fibrinopurulent exudates like a cap, and this finding resembled inflammatory polyp or pseudomembranous colitis. The microscopic finding revealed elongated hyperplastic glands with inflammatory infiltrate in the lamina propria and fibromuscular obliteration of lamina propria. The cap of polyp is formed by mucus, fibrin, and inflammatory cells.[1,3]

Several case reports have suggested a few treatment modalities, based on etiological hypothesis; anti-inflammatory agent, antibiotics, immunomodulators, and endoscopic and surgical therapy (Table 1). However optimal treatment has not yet been established. The effectiveness and administration schedule of infliximab for cap polyposis also have not yet been established. One report described complete remission after four infusions of infliximab at 0, 8, 12, and 24 weeks, however another reports showed no benefit after a similar treatment.[3,9] In our present case, the patient fortunately achieved remarkable clinical, endoscopic and histological responses after single infusion of infliximab. The short term response of our patient was published in Korean.[20] Our present case is the long term follow up result after 3 years. Furthermore, resolution of the disease maintained for 36 months. Consequently, our present case might support the hypothesis that inflammation has some role in the pathogenesis of cap polyposis.[3] Of course, additional studies about cap polyposis treated with infliximab infusion, including its optimal dosage and administration schedule, are needed. Nevertheless we suggest that infliximab might be a good treatment modality for cap polyposis patients who are refractory to conservative management.

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