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

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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. 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. 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/mm³, platelet count was 319,000/mm³ 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-
Colonoscopy conducted 4 weeks after infliximab infusion revealed reductions in the size and number of the sessile polyps.

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).

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. 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 *Escherichia 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 Polyps 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        |
| Gehon et al. (1994)\(^5\)     | Female | 65  | Diarrhea                   | Rectum                 | Cleversal enema                   | 9 months           | Improved        |
| Shiomot 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            | Improved        |
| Shiomi et al. (1998)          | 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 colectomy                 | 1 year             | Resolved        |
| Esaki et al. (2001)\(^11\)    | Male   | 21  | Weight loss, bloody diarrhea, abdominal pain, mucous diarrhea | Rectosigmoid         | Metronidazole                      | 6 months           | Transiently improved |
| Oiya et al. (2002)            | Male   | 73  | No symptom                 | From sigmoid to cecum | No treatment                       | 4 months           | Resolved        |
| Shimizu et al. (2002)\(^3\)   | Female | 12  | Mucous bloody diarrhea     | Rectosigmoid           | Metronidazole                      | 12 months          | Resolved        |
| Okawara 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 diarrhea | Rectum               | *Helicobacter* eradication         | 18 months          | Resolved        |
| Bookman et al. (2004)\(^3\)   | Female | 36  | Abdominal pain, mucous bloody stool diarrhea | Rectosigmoid         | *Helicobacter* eradication         | 26 months          | Resolved        |
| Konishi et al. (2005)\(^17\)  | Female | 76  | Hypoproteinemia, mucous bloody stool diarrhea | Total colon           | Sigmoid colectomy                 | 3 months           | Resolved        |
| Maunoury et al. (2005)\(^18\) | Female | 52  | Abdominal pain, mucous diarrhea | Rectum               | Infliximab infusion               |                    | Failed          |
| Ryu et al. (2006)\(^19\)      | Male   | 64  | Weight loss, diarrhea      | Rectosigmoid           | Conservative                       | 1 year             | Resolved        |
ischemia and recurrent mucosal trauma. 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.

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 report showed no benefit after a similar treatment. 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. 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. 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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