Cap Polyposis (CP) Which Relapsed after Remission by Avoiding Straining at Defecation, and was Cured by *Helicobacter pylori* Eradication Therapy

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**Abstract**

A 52-year-old woman was diagnosed with cap polyposis (CP) with characteristic clinical, endoscopic, and histological features. By avoiding straining at defecation, her symptoms improved temporarily, however recrudesced. She was diagnosed with *Helicobacter pylori* (*H. pylori*) infection, and received eradication therapy successfully. After this eradication therapy, her symptoms and colonoscopic findings recovered completely. Only two reports in the English language literature have discussed the relationship between CP and eradication therapy for *H. pylori*, all patients achieved complete recovery. We recommend *H. pylori* testing for all cases of CP and *H. pylori* eradication therapy if necessary.

**Key words:** cap polyposis, *Helicobacter pylori*, eradication therapy


**Introduction**

Cap polyposis (CP) is an inflammatory bowel disease first reported in 1985 by Williams et al (1). It is characterized by the presence of inflammatory multiple polyps consisting of elongated tortuous crypts covered by a “cap” of granulation tissue. The symptoms of CP are mucoid stool, diarrhea, bleeding on defecation, tenesmus, abdominal pain and weight loss. Laboratory data usually reveal hypoproteinemia as a result of protein-losing enteropathy (2-6). Lesions are mainly located in the rectum and sigmoid colon, but some cases extend to the oral side of the colon (2, 4, 5, 7-9). The pathogenesis of this disease remains unknown and no specific treatment has been established. Since Oiya et al reported a case of CP cured by *Helicobacter pylori* (*H. pylori*) eradication therapy, a relationship between *H. pylori* infection and CP of the colon has been suggested; however, there have been only two reports in the English language literature discussing this relationship (7, 10). Here, we report a case of CP that relapsed after remission by avoiding straining, and was cured following eradication therapy for *H. pylori* infection.

**Case Report**

In August 2003, a 52-year-old woman visited another hospital complaining of diarrhea, mucous bloody stools and 5 kg weight loss over 6 months. Her past medical history included cholecystectomy for cholelithiasis. Physical examination and laboratory data were normal. Colonoscopy revealed multiple reddened and eroded lesions from the rectum to the transverse colon. She was diagnosed with nonspecific colitis or ulcerative colitis, and treated with lactomyn (3.0 g/day) and 5-aminosalicylic acid (1.5 g/day) without improvement. In September 2003, she was referred to our hospital.

Laboratory data at the first medical examination at our...
hospital revealed decreased total serum protein (5.9 g/dL) and other normal data. Fecal culture revealed no pathogenic organisms and serum anti-amebic antibody was negative. Diarrhea (about four stools per day) and mucous bloody stools had continued. On September 12, colonoscopy revealed multiple sessile polyps on the apices of the mucosal folds from the rectum to the sigmoid colon. The surface of these lesions was reddish and adhered with white mucus. The intervening mucosa was normal, and no diverticular disease was present around these lesions (Fig. 1a). Endoscopic ultrasonography with a 15-MHz miniature probe disclosed significant thickening of the mucosal layer. Histopathological examination of polypectomy specimens obtained from multiple inflammatory polyps revealed inflamed mucosa with elongated tortuous crypts and a granulation tissue so-called “cap” on the mucosa. Fibromuscular obliteration was evident in the lamina propria (Fig. 2a, b). *H. pylori* was not detected in these specimens. Given these characteristic endoscopic and histologic features, a diagnosis of CP was made. Because she tended to strain at defecation, we educated her to avoid this habit. By avoiding straining, her diarrhea and mucous bloody stools improved within a week, and colonoscopy showed improvement temporarily (Fig. 1b); however, colonoscopic findings deteriorated gradually each year despite avoiding straining, and diarrhea and mucous bloody stools had recurred by December 2006. In mid-December 2006, the colonoscopic findings were worse than the findings on admission (Fig. 1c). At this time, gastroscopy was performed, revealing atrophic gastritis, and she was diagnosed with *H. pylori* infection by the rapid urease test and the histology of gastric biopsy specimens. She received eradication therapy with 60 mg lansoprazole, 1,500 mg amoxicillin and 400 mg clarithromycin, all taken daily for 7 days. Eradication therapy was successful, and her symptoms were markedly improved. Colonoscopy revealed marked improvement about one month later, and normal findings 14 months later (Fig. 1d). She has had no recurrence in the 48 months since *H. pylori* eradication therapy.

**Discussion**

This study describes a strong relationship between CP and *H. pylori* eradication therapy. We performed a search of the MEDLINE database from 1985 to 2009 regarding CP, and the treatments, outcomes of treatments and clinical courses were analyzed. Nineteen reports in English, including 26 cases, were evaluated (Table 1). We classified treatment outcomes into three groups, effective, temporarily effective and ineffective. Even if symptoms of CP improved, recurrences after remission were reported in many cases (2, 7, 10-15), as in the present case. So, not only symptomatic improvement but also the disappearance of lesions is thought to be required to evaluate treatment effectiveness.

Surgical resections of the affected colon were performed in nine of 26 cases, but recurrence of symptoms and endoscopic findings were recognized in four cases (10-12, 14).
Figure 2.  a. Histopathological examination of biopsy specimens revealed inflamed mucosa with elongated crypts and granulation tissue, a so-called "cap", on the mucosa (Hematoxylin and Eosin stain (HE) x4). b. The fibromuscular obliteration was evident in the lamina propria (Hematoxylin and Eosin staining x20).

Table 1. Outcome of Each Treatment for Cap Polypsis from Past English Literatures

<table>
<thead>
<tr>
<th>Treatment</th>
<th>Effective</th>
<th>Temporarily Effective</th>
<th>Ineffective</th>
<th>Rate of Effectiveness (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>H. pylori eradication</td>
<td>5</td>
<td></td>
<td></td>
<td>100</td>
</tr>
<tr>
<td>Colostomy</td>
<td>1</td>
<td></td>
<td></td>
<td>100</td>
</tr>
<tr>
<td>Surgical resection</td>
<td>5</td>
<td>4</td>
<td>0</td>
<td>56</td>
</tr>
<tr>
<td>Infliximab</td>
<td>1</td>
<td></td>
<td>1</td>
<td>50</td>
</tr>
<tr>
<td>No treatment</td>
<td>1</td>
<td>2</td>
<td>1</td>
<td>25</td>
</tr>
<tr>
<td>MNZ</td>
<td>2</td>
<td>2</td>
<td>7</td>
<td>18</td>
</tr>
<tr>
<td>Avoidance of straining</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>17</td>
</tr>
<tr>
<td>Steroid therapy</td>
<td>2</td>
<td>9</td>
<td>0</td>
<td></td>
</tr>
<tr>
<td>APC, EMR</td>
<td>1</td>
<td>2</td>
<td>0</td>
<td></td>
</tr>
<tr>
<td>TPN</td>
<td>1</td>
<td>1</td>
<td>0</td>
<td></td>
</tr>
<tr>
<td>Other antibiotics</td>
<td>2</td>
<td>0</td>
<td>0</td>
<td></td>
</tr>
<tr>
<td>5-ASA, sulfasalazine</td>
<td>13</td>
<td>0</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

MNZ: metronidazole, 5-ASA: 5-aminosalicylic acid, APC: argon plasma coagulation, EMR: endoscopic mucosal resection, TPN: total parenteral nutrition, *: including our case, **: eradication regimen: 60 mg lansoprazole or 20-40mg rebeprazole + 1500mg amoxicillin + 400mg or 800mg clarithromycin for 7-14 days. ***: levofloxacin, and cyplofloxacin hydrochloride

was recognized that 5-aminosalicylic acid, sulfasalazine (5, 6, 10-14, 16-18), steroid therapy (2, 4, 7, 10, 11, 13, 14, 16, 17), Argon plasma coagulation (APC) and endoscopic mucosal resection (EMR) (10, 13, 15) are ineffective for CP from these analyses of the past English language reports. Bookman et al reported the successful treatment of CP with infliximab, and suspected that TNFα plays a role in the pathogenesis of CP (17). On the other hand, Maunoury et al reported a case of CP which was unresponsive to infliximab (13). Four cases of spontaneous improvement of CP were reported (9, 16, 19), but complete recovery was achieved in only one case (19).

As for the pathogenesis of CP, chronic mechanical stimulation by abnormal colonic motility and repeated trauma to the colonic mucosa caused by straining at defecation have been suggested (1, 6, 14). Histological findings of CP have been described to be similar to those seen in mucosal prolapse syndrome (MPS) in the points of obliteration of the lamina propria by fibroblasts and smooth muscle fibers (fibromuscular obliteration), superficial erosion associated with granulation tissue, elongated tortuous crypts and intramucosal elastin (6, 14, 20). Oriuchi et al reported two cases of successful treatment of CP by avoidance of intraluminal trauma, and suggested that CP might be a subtype of MPS (6). But, avoidance of straining at defecation was not effective in the other five cases of CP including our case (10, 17), and some cases of CP had no evidence of abnormal colonic motility, fibromuscular obliteration or intramucosal elastin (7, 11, 17). The CP of our case relapsed after remission by avoiding straining at defecation, and fibromuscular...
obliteration was evident in the lamina propria. Mucosal prolapse and abnormal colonic motility may in part contribute to the pathogenesis of CP, but whether this pathogenesis is the principal cause of CP remains uncertain.

Although a causative organism of CP has not been identified to date, an infectious etiology of CP has been proposed. Metronidazole (MNZ) has been effective in only two cases identified to date, an infectious etiology of CP has been proposed. Metronidazole (MNZ) has been effective in only two cases identified to date, an infectious etiology of CP has been proposed. Metronidazole (MNZ) has been effective in only two cases identified to date, an infectious etiology of CP has been proposed. Metronidazole (MNZ) has been effective in only two cases identified to date, an infectious etiology of CP has been proposed.

The anti-inflammatory actions of MNZ may play a central role in healing CP rather than its antibiotic action (21). Broad-spectrum antibiotics, levofloxacin and ciprofloxacin hydrochloride, were ineffective for CP (17, 21). Only two reports in the English language literature, including four cases of *H. pylori* eradication therapy for CP, have been reported, but all cases achieved complete recovery (7, 10). These are notable outcomes in comparison with other treatments (Table 1). Although there is a possibility of other bacterial infections which are sensitive to the antibiotic combination used in *H. pylori* eradication therapy, fecal cultures revealed no pathogenic organisms in any *H. pylori* eradicated cases (7, 10). Akamatsu et al reported that *H. pylori* was not detected in the colonic mucosa obtained from multiple inflammatory polyps of CP (10). These results suggest that *H. pylori* infection plays a part in the etiology of CP, not directly but indirectly. Since the discovery of *H. pylori*, several studies have been published concerning the hypothetical role of this bacterium in different extragastric diseases, such as ischemic heart disease (23), idiopathic thrombocytopenic purpura (24), iron deficiency anemia (25), chronic urticaria (26) and other disorders. *H. pylori* infection may cause extragastric manifestations, directly or indirectly, by various mechanisms, including atrophic gastritis, the release of inflammatory mediators, molecular mimicry, and systemic immune response. In the etiology of CP, persistent *H. pylori* infection may contribute by such mechanisms.

In conclusion, we recommend testing for *H. pylori* in all cases of CP and administering *H. pylori* eradication therapy if necessary. Further examination of the role of *H. pylori* infection in CP is necessary.

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References


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