CASE REPORT

Nine-year Follow-up from Onset to Spontaneous Complete Remission of Cap Polyposis

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Abstract

A 58-year-old woman with severe constipation and a habit of straining at defecation was diagnosed to have mucosal prolapse syndrome. One year later, her primary symptom changed to bloody diarrhea. The colonoscopic and histological findings were consistent with the characteristics of cap polyposis. After nine years, her symptoms and colonoscopic abnormalities disappeared completely without treatment. For two years since that time, the patient has remained well with normal endoscopy findings and a high value of anti-\textit{Helicobacter pylori} immunoglobulin G. In this case, cap polyposis might have developed via mucosal prolapse syndrome and then regressed completely, irrespective of the \textit{Helicobacter pylori} infection.

Key words: mucosal prolapse syndrome, spontaneous remission, \textit{Helicobacter pylori}

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Introduction

Cap polyposis is a rare disorder associated with bloody diarrhea and abdominal pain that is characterized by rectosigmoid polyps covered by caps of fibrous purulent exudate; it was first described by Williams et al. in 1985 (1). The etiology of cap polyposis is unknown; however, rectal prolapse and \textit{Helicobacter pylori} (\textit{H. pylori}) infection have been implicated (2-6). It has been reported that some cases have been treated successfully by the avoidance of straining at defecation (2), metronidazole (7), steroids (8), \textit{H. pylori} eradication therapy (5, 6), infliximab (9) and surgical resection (3). However, no curative standard treatments for this condition have been established.

We herein report the first case of cap polyposis considered to have transformed from mucosal prolapse syndrome and for which spontaneous complete remission occurred nine years after diagnosis against a constant background of \textit{H. pylori} infection.

Case Report

A 58-year-old woman presented with severe constipation and a positive test result for fecal occult blood at a health check-up. The patient had no remarkable medical or family history, although she had a habit of straining at defecation. Total colonoscopy demonstrated shallow ulcers on the anterior wall of the rectum (Fig. 1A). A histologic examination of a biopsied ulcer showed severe infiltration of inflammatory cells with elongated stroma on the surface of the mucosa and fibromuscular obliteration of the lamina propria (Fig. 1B). These findings made distinguishing between a differential diagnosis of initial cap polyposis and mucosal prolapse syndrome difficult, although the fibromuscular obliteration of the lamina propria seemed to be too marked for cap polyposis. Given these clinical and histological findings, a diagnosis of mucosal prolapse syndrome was made, and we instructed the patient to avoid straining at defecation.

One year after diagnosis, however, the patient developed bloody diarrhea (seven to eight bowel movements/day).
Laboratory studies showed normal hemoglobin levels (14.3 g/dL), white blood cell counts (5,910/μL), liver enzyme levels and albumin levels (3.8 g/dL). The level of C-reactive protein was 0.2 mg/dL (reference range: 0-0.24 mg/dL). Stool examinations for enteric pathogens showed negative results. Colonoscopy revealed multiple reddish sessile polyps covered with a layer of fibrinopurulent exudate extending from the rectum to the distal sigmoid colon (Fig. 1C). A histopathological examination of the polypectomy specimens revealed inflamed mucosa with elongated tortuous crypts and a so-called “cap” of granulation tissue on the mucosa (Fig. 1D, E). Fibromuscular obliteration was evident in the lamina propria. These histological findings, in combination with the clinical and endoscopic features, were consistent with a diagnosis of cap polyposis.

The bloody diarrhea decreased without treatment (three to four bowel movements/day), and the patient was followed after diagnosis. Although the rapid urease test for detection of *H. pylori* using a gastric biopsy specimen gave a positive result, the patient declined eradication therapy. Although her symptoms had improved (diarrhea two to three times/day, often bloody), she requested further observation, and we therefore performed colonoscopic examinations every year (Fig. 2A-C), which continued to demonstrate the features of cap polyposis and mild symptoms.

Nine years after the diagnosis of cap polyposis, the patient’s bloody diarrhea stopped completely without medication. Surprisingly, colonoscopy revealed no signs of any polypoid lesions (Fig. 2D). The serum level of anti-*H. pylori* immunoglobulin G (anti-*HP* IgG) was high (131 U/mL; ref-
Figure 2. Colonoscopic views obtained (A) three, (B) five and (C) seven years after diagnosis of cap polyposis. (D) Disappearance of cap polyposis nine years after diagnosis and (E) two years later, showing no evidence of recurrence.

Table. Clinical Characteristics of Two Previously Reported Patients Showing Spontaneous Complete Remission of Cap Polyposis.

<table>
<thead>
<tr>
<th>Age (years)</th>
<th>Gender</th>
<th>Symptoms</th>
<th>Location of polyp</th>
<th>Remission after diagnosis</th>
<th>Reference</th>
</tr>
</thead>
<tbody>
<tr>
<td>67</td>
<td>Female</td>
<td>Mucous and bloody diarrhea</td>
<td>Rectum to sigmoid colon</td>
<td>2 months</td>
<td>[10]</td>
</tr>
<tr>
<td>42</td>
<td>Female</td>
<td>Abdominal pain and bloody diarrhea</td>
<td>Rectum to transverse colon</td>
<td>18 months</td>
<td>[8]</td>
</tr>
</tbody>
</table>

Discussion

Cap polyposis is rare and has unique clinical, endoscopic and histological characteristics. Its clinical course has been reported to vary from spontaneous remission to persistence requiring surgical resection of the lesions (3, 8, 10, 11), while little is known about its long-term course. To our knowledge, only two previously untreated patients have been reported to show spontaneous and complete clinical and endoscopically confirmed remission (Table). Both patients were women, as in the present case; however, their remissions lasted for relatively short durations.

The pathogenesis of cap polyposis remains controversial. In particular, there has been debate about whether cap polyposis is a specific form of inflammatory disorder or part of a spectrum of mucosal prolapse syndrome (12). Mucosal prolapse syndrome is caused by abnormal colonic motility with subsequent local ischemia and repeated mucosal trauma, mimicking the pathological features of cap polyposis. In the present case, mucosal prolapse syndrome had transformed into typical cap polyposis, even though the patient had avoided straining at defecation. It has been suggested that mucosal prolapse syndrome may participate in the etiology of cap polyposis. In addition, if progression to cap polyposis occurs despite effective treatment for mucosal prolapse syndrome, then other factors may be involved.

It has been reported that some cases of cap polyposis have been cured after the administration of H. pylori eradication therapy (4-6). Although some cases have been treated effectively with infliximab or steroids (8, 9), inflammation due to bacteria other than H. pylori might play a role in the development of cap polyposis. The high value of anti-HP IgG observed in our patient after spontaneous complete remission lends some weight to this hypothesis. It has been shown that the anti-HP IgG titer decreases by more than 40% until two to six months after eradication of H. pylori (13, 14), and a reduction of the titer by more than 30% within 12 months is taken as an indicator of eradication.

ference range, <10 U/mL). After an additional two years of follow-up, the patient remained asymptomatic and colonoscopy revealed no evidence of cap polyposis (Fig. 2E), while the high value of anti-HP-IgG (100 U/mL) was retained.

Age (years) Gender Symptoms Location of polyp Remission after diagnosis Reference
| 67  | Female  | Mucous and bloody diarrhea | Rectum to sigmoid colon | 2 months | [10]   |
| 42  | Female  | Abdominal pain and bloody diarrhea | Rectum to transverse colon | 18 months | [8]    |
with a specificity of 95.4% and a sensitivity of 100% (15). Therefore, in the present case, the \textit{H. pylori} infection appeared to be persistent for at least two years after spontaneous remission. It has been reported that the aging process profoundly affects the structure of the human gut microbiota (16). Therefore, it is speculated that changes in the gut microbiota of our patient during the 9-year period might have played a role in the spontaneous remission we observed.

In conclusion, we herein described the long-term course of a case of cap polyposis, which developed from mucosal prolapse syndrome and showed a complete remission despite a persistently high level of anti-\textit{HP} IgG. This case suggests that mucosal prolapse syndrome may be an important causative factor for cap polyposis and that the remission we observed was unlikely to have been associated with \textit{H. pylori} infection. The present findings may offer some valuable insight into the clinical course and pathogenesis of cap polyposis. Further studies to elucidate its etiology are therefore warranted.

The authors state that they have no Conflict of Interest (COI).

References


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