Total Obliteration of Colonic Lumen by Localized Giant Inflammatory Polyposis in Ulcerative Colitis: Report of a Japanese Case

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Although inflammatory polyposis of the colon is a recognized local complication of ulcerative colitis, giant inflammatory polyposis that totally obliterates the colonic lumen is uncommon, and most of the patients have been from Western countries. We report a Japanese man who had localized giant inflammatory polyposis that obstructed the retrograde flow of barium in a double-contrast barium enema study at the splenic flexure in ulcerative colitis. After resection by total proctocolectomy and ileostomy, pathological examination of the specimen confirmed localized giant inflammatory polyposis in the descending colon and splenic flexure without malignancy.

(Key words: descending colon, splenic flexure, colonic obstruction)

Introduction

Inflammatory polyposis of the colon is a recognized common complication of ulcerative colitis, reportedly occurring in 12–18% of the cases (1–3). However, giant inflammatory polyposis with a diameter exceeding 15 cm (4) is rare resultant with only seven cases of complete colonic obstruction reported in the literature (5–11); 6 of the 7 cases were from the United States (5–9, 11) and one was from Israel (10). Furthermore, seven other cases of incomplete colonic obstruction due to inflammatory polyposis were reported from Western countries (12–18), but only a few Japanese cases have been reported (19–21).

Hence, we report a Japanese patient with colonic obstruction due to localized giant inflammatory polyposis in ulcerative colitis and discuss the possible reason for the more frequent occurrence of this disorder in Western countries compared to Japan.

Case Report

A 54-year-old Japanese man first noticed a bloody stool in April, 1992. He was diagnosed four months later at our hospital as having ulcerative colitis. He responded well to administered salazosulfapyridine (6 g/day) and betamethasone suppositories (1 mg/day) for about 2 years. A double-contrast barium enema study in March 1993, revealed only a lack of haustral markings in the left colon without ulcerations or polyps.

Beginning in March 1994, he noticed a gradual increase in the frequency of bloody stools with a dull pain in the lower right quadrant of the abdomen. He was then administered salazosulfapyridine (6 g/day) and betamethasone enemas (6 mg/day) for 6 months. A barium enema study in May 1994, revealed fungating lesions with a nodular surface in the descending colon (Fig. 1A) and the splenic flexure (Fig. 1B). There was no obstruction of the colon. Colonoscopy at that time revealed severe ulcerations of the rectum (Fig. 2A) and the sigmoid colon. There was a polyoid mass in the descending colon at 40 cm which appeared to be benign (Fig. 2B) as the pit pattern of the surface seemed normal. Biopsy of the rectum and the polyoid mass showed acute inflammation but there was no evidence of malignancy. The diagnosis was benign inflammatory polyposis.

The patient’s abdominal symptoms did not improve with salazosulfapyridine and betamethasone administration, and he developed a low-grade fever and anorexia with a 10 kg weight loss. A repeat barium enema study in September 1994, demonstrated enlargement of the polyoid masses in the descending colon (Fig. 3A) and a lesion in the splenic flexure that partially obstructed the retrograde flow of barium (Fig. 3B). Colonoscopy
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Figure 1. Double-contrast barium enema of the descending colon (A) and splenic flexure (B) in May 1994. Two nodular polypoid lesions which did not obstruct the colon were found in the area. The length of the lesions in the descending colon and splenic flexure was 6 and 21 cm, respectively.

Figure 2. Colonoscopy of the rectum at 10 cm (A) and descending colon at 40 cm (B) in May 1994. Irregular ulcerations and edematous mucosa in the rectum indicate acute ulcerative colitis. A nodular polypoid mass in the descending colon appears non-neoplastic.
at that time showed that the polypoid mass in the descending colon obliterated the lumen. However, the mucosa of the rectum and sigmoid colon appeared regenerative. Since the colonoscope could not be advanced beyond the mass, the patient was admitted to our hospital in October 1994 for further study and treatment.

Examination revealed a well nourished man with normal physical findings except for a low-grade fever (37.1°C) and tenderness in the right lower quadrant of the abdomen on palpation. No mass was found on palpation of the abdomen. Bowel sounds were present and there was no organomegaly. The following laboratory tests were normal: serum levels of sodium, potassium, chloride, phosphorus, blood urea nitrogen, creatinine, bilirubin, alkaline phosphatase, glutamic oxaloacetic transaminase, glutamic pyruvic transaminase, iron, carcinoembryonic antigen; prothrombin time, partial thromboplastin time. Urinalysis was also normal. The serum total protein and albumin were decreased to 4.7, and 2.4 g/dl, respectively, and the glucose was elevated to 169 mg/dl. The hematocrit was 37% with adequate platelets. The leukocyte count was 9,700 with a marked shift to the left. X-rays of the chest and abdomen and the electrocardiogram were all normal.

On admission, the patient was placed on intravenous hyperalimentation for 2 weeks and on steroid pulse therapy for 3 days but without improvement in symptoms. Finally, in October, 1994, a barium enema study demonstrated the polypoid lesion in the splenic flexure had totally obliterated the colonic lumen (Fig. 4). Total proctocolectomy and ileostomy were then performed. At the time of surgery, a 9-cm mass was palpated in the descending colon and a 25-cm mass was palpated in the splenic flexure. The patient recovered uneventfully and was discharged on the 32nd postoperative day.

Pathology

Gross appearance of lesion

We removed 9 cm from the opened rectum and 131 cm of the opened colon. A polypoid mass (8.5 cm long) was present in the descending colon and another (23 cm long) in the splenic flexure (Fig. 5A, B), and each was 2 cm in height. The splenic flexure was dilated to a maximum diameter of 18 cm. Both masses consisted of innumerable, conglomerated, clustered polyps with frequent branches. While there was a loss of the normal colonic haustra in the left colon, no mucosal ulceration was observed in the rectum or the entire colon. The cut surface of the mass in the splenic flexure was composed of firm, brownish-red tissue that did not appear to penetrate into the muscularis propria.
Figure 4. Double-contrast barium enema of the splenic flexure in October 1994. The polypoid lesion totally obliterates the colonic lumen.

Microscopic appearance

Sections through the main polypoid mass revealed that it composed confluent, branching, inflammatory polyps that consisted of regenerating, non-neoplastic glands with numerous crypt abscesses separated by a severely inflammed lamina propria that was infiltrated by neutrophils, lymphocytes, and plasma cells (Fig. 6A, B). In the rectum and the flat areas of the colon, the mucosa was regenerating, resulting in chronic inflammation, but it was partially inflammed with depletion of goblet cells and deep ulcer scars. The inflammation of the lamina propria in these areas was weaker than in the polypoid lesion, suggesting that the polypoid masses had grown on the severely inflammed mucosa. No granulomatous, adenomatous, or carcinomatous proliferation was observed in the sections of the resected specimen or within the nine regional lymph nodes observed. The final pathologic diagnosis was chronic active ulcerative colitis with giant inflammatory polyposis.

Discussion

Giant inflammatory polyposis totally obliterating the colonic lumen in a patient with ulcerative colitis was first reported by Fitterer et al (5). Six other cases were subsequently published (6–11). These patients had a 2- to 10-year history of ulcerative colitis before the inflammatory polyposis was diagnosed. Microscopic examination of the resected specimens demonstrated that the circumferential mucosa of the inflammatory polyp exhibited regenerative changes compatible with chronic active ulcerative colitis, as seen in most of the cases, including the present case. Kelly and Gabos (22) divided inflammatory polyps into two major groups; 1) polypoid mucosal tags due to active undermining ulceration, and 2) mature inflammatory polyps composed of mucosa, muscularis mucosa, and a submucosal core. The mature inflammatory polyps are derived from the polypoid mucosal tags after regeneration and the adjacent mucosa shows regenerative changes (22). According to this concept of the pathogenesis of inflammatory polyps, most of the giant inflammatory polyposis with colonic obstruction were of the mature type.

The question arises as to why more Westerners develop this disorder than Japanese? In the seventeen reported cases (fourteen Western cases and three Japanese cases) (5–21) of giant inflammatory polyposis in ulcerative colitis, the main occup-
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Figure 6. Microscopic findings of a section made through the polypoid mass in the splenic flexure. Individual polyps of the mass consist of regenerating non-neoplastic glands with numerous crypt abscesses (B) separated by an inflammed lamina propria (A). HE stain. Calibration bar (A) indicates 2 mm.

ing portion of the polypoid lesion did not differ much between Western and Japanese cases. The transverse colon was the main portion in seven of the seventeen cases, the descending colon in four, the sigmoid colon in two, the splenic flexure in one, and the whole colon in one. It is suggested that the location may be unrelated to the growing of giant inflammatory polyposis. Histological findings of the polypoid lesion were also similar among the cases. The incidence of complicating inflammatory polyps in Japanese with ulcerative colitis is about 30%, which is similar to that reported in Westerners (23). However, it is generally accepted that among Westerners the incidence of colon cancer is greater than among Japanese. We suggest one of the causes of more frequent complications with giant inflammatory polyposis seen in Westerners may be related to a high fat, low fiber diet, which is recognized to be one of the risk factors of colon cancer.

In a detailed analysis by Kelly et al (24), inflammatory polyps were associated with Crohn’s disease in approximately two-thirds of the cases, and with ulcerative colitis in one-third of the cases. The transverse colon is the most common site (40%), followed by the sigmoid colon (15%), the descending colon (15%), the cecum (14%), the splenic flexure (7%), and the hepatic flexure (7%), similar to our review of the literature. More than 50% of these cases mimicked neoplasm on barium enema (24). De Dombal et al (1), however, reported that only one of 58 patients (2%) with pseudopolyposis in ulcerative colitis had the true adenomatous polyps. The estimated incidence of a complicating malignancy in a patient with ulcerative colitis is 3–5% (25), suggesting that the risk of malignancy in a patient with ulcerative colitis and inflammatory polyposis is about equal to that in a patient without inflammatory polyposis. This consideration was confirmed by the review of Goldenberg et al (26) that inflammatory polyps with ulcerative colitis are not considered precancerous. However, one case of localized giant pseudopolypsis in ulcerative colitis with infiltrating adenocarcinoma has been reported (27), which indicates that giant inflammatory polyposis may be an indication for surgery when it mimicks a neoplasm on examination of the colon.

In conclusion, we have reported a Japanese case of localized giant inflammatory polyposis that totally obliterated the colonic lumen in ulcerative colitis. The incidence of complicating inflammatory polyposis in a patient with ulcerative colitis may increase with a high fat, low fiber diet.

References

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