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Case Report

A Case of Ulcerative Colitis with Stenosing Symptoms Caused by Localized Giant Inflammatory Polyposis

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SUMMARY

A 40-year-old man with relapsed ulcerative colitis (UC) underwent colonoscopy. Colonoscopy revealed a dense cluster of elongated polyps with surrounding ulcerative lesions in the ascending colon and intestinal stricture at the same site. Total colectomy was performed, and postoperative pathological findings showed localized giant inflammatory polyposis (LGIP) associated with UC.

While obstructive giant inflammatory polyposis associated with UC is considered uncommon, inflammatory polyposis is a recognized local complication.

Keywords : Ulcerative colitis, Localized giant inflammatory polyposis, total colectomy

INTRODUCTION

Inflammatory polyps are often coexistent with ulcerative colitis (UC). Inflammatory polyps do not cause bowel obstruction and are often followed up. In contrast, there is a morphological abnormality known as localized giant inflammatory polyposis (LGIP). This is a condition in which polyps of various sizes, including elongated polyps, are clustered together. It is rare for regional densification of LGIP to cause intestinal obstruction. Herein, we report a rare case of UC with intestinal stenosis caused by LGIP that was successfully treated by colectomy.

CASE REPORT

A 40-year-old man suffering from UC visited our hospital with a chief complaint of bloody stools (defecation frequency : 15 times/day) that had been persistent for a month. He had total colitis type UC with a disease duration of 16 years and was maintained in remission with oral mesalamine 4000 mg/day. His initial physical examination revealed tachycardia with a heart rate of 101 beats per minute, and abdominal examination revealed tenderness in the right lower abdomen. Severe clinical activity of UC was observed,

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Table 1 Laboratory data on admission.

AST	19 IU/L	WBC	6800 /μl
ALT	16 IU/L	Neutro.	67.9 %
T-Bil	0.3 mg/dl	Lymph.	24.1 %
ALP	352 IU/L	Mono.	7.3 %
LDH	146 IU/L	Eos.	0.3 %
γ-GTP	151 IU/L	Baso.	3.2 %
BUN	6.0 mg/dl	RBC	345×10^4 /μl
Cre	0.74 mg/dl	Hb	9.3 g/dl
Na	136 mEq/l	Ht	30.0 %
K	4.0 mEq/l	MCV	87.0 fL
Ca	8.7 mEq/l	Plt	51.8×10^4 /μl
TP	6.4 g/dl		
Alb	2.6 g/dl	CEA	0.71 ng/ml
CRP	14.5 mg/dl	CA19-9	1.20 U/ml
ESR (1 hr)	118 mm		
		CMV C7-HRP	—
		T-SPOT®	—

Abbreviations : CEA : carcinoembryonic antigen, CA19-9 : carbohydrate antigen 19-9, CMV C7-HRP : cytomegalovirus antigenemia assay, T-SPOT : T lymphocyte spot test for tuberculosis infection.

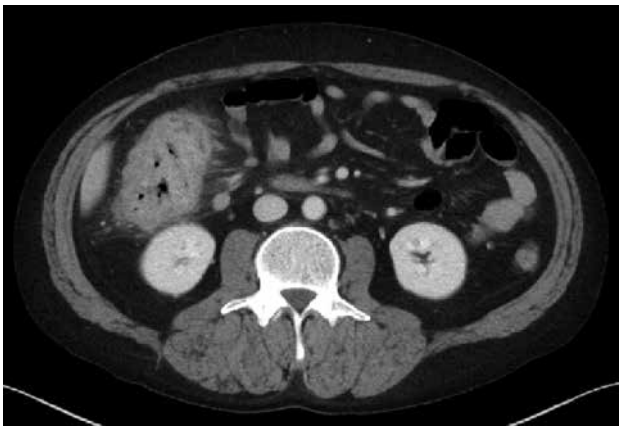

Figure 1
Contrast-enhanced computed tomography of the abdomen, showing marked wall thickening and a strong enhanced effect in the ascending colon.

with a Lichtiger index of 15 points[1], and the patient was hospitalized. Table 1 shows the blood examination findings at the time of admission. Contrast-enhanced computed tomography (CT) of the abdomen showed marked wall thickening of the ascending colon and a strong contrast effect in the same area (Figure 1). Colonoscopy showed inflammatory UC mucosa in the ascending colon, surrounded by clusters of small and large elongated polyps with ulcerative lesions, and intestinal stenosis at the same site (Figure 2, a-d). LGIP was suspected based on morphology. The biopsy showed a high degree of inflammatory cell infiltration, but no evidence of dysplasia or carcinoma. The colonoscopy one year ago showed no evidence of these findings, and since the patient presented with LGIP with intestinal stenosis, we also considered the possibility of future intestinal obstruction. Although biopsy did not indicate malignancy, UC-associated neoplasia could not be completely excluded. Furthermore, the clinical activity of UC was so severe that remission induction therapy by drug administration was considered difficult. Therefore, he underwent a total colostomy.

In the surgical specimens, the mucosal folds were preserved in the left colon, while the ascending colon showed subrounded ulceration and polyposis (Figure 3). Histologically, there was lamina muscularis mucosae thickening, abnormal gland arrangement, atrophy and partial crypt abscess in the entire intestinal tract, and especially strong inflammation and marked fibrosis in the LGIP area in the ascending colon. In the ulcer area, there was strong inflammation extending to the muscularis propria and serosa, but no evidence of malignancy. The LGIP findings by endoscopy were similar to those of pathology, and the final diagnosis was LGIP occurring in UC (Figure 4, a-d).

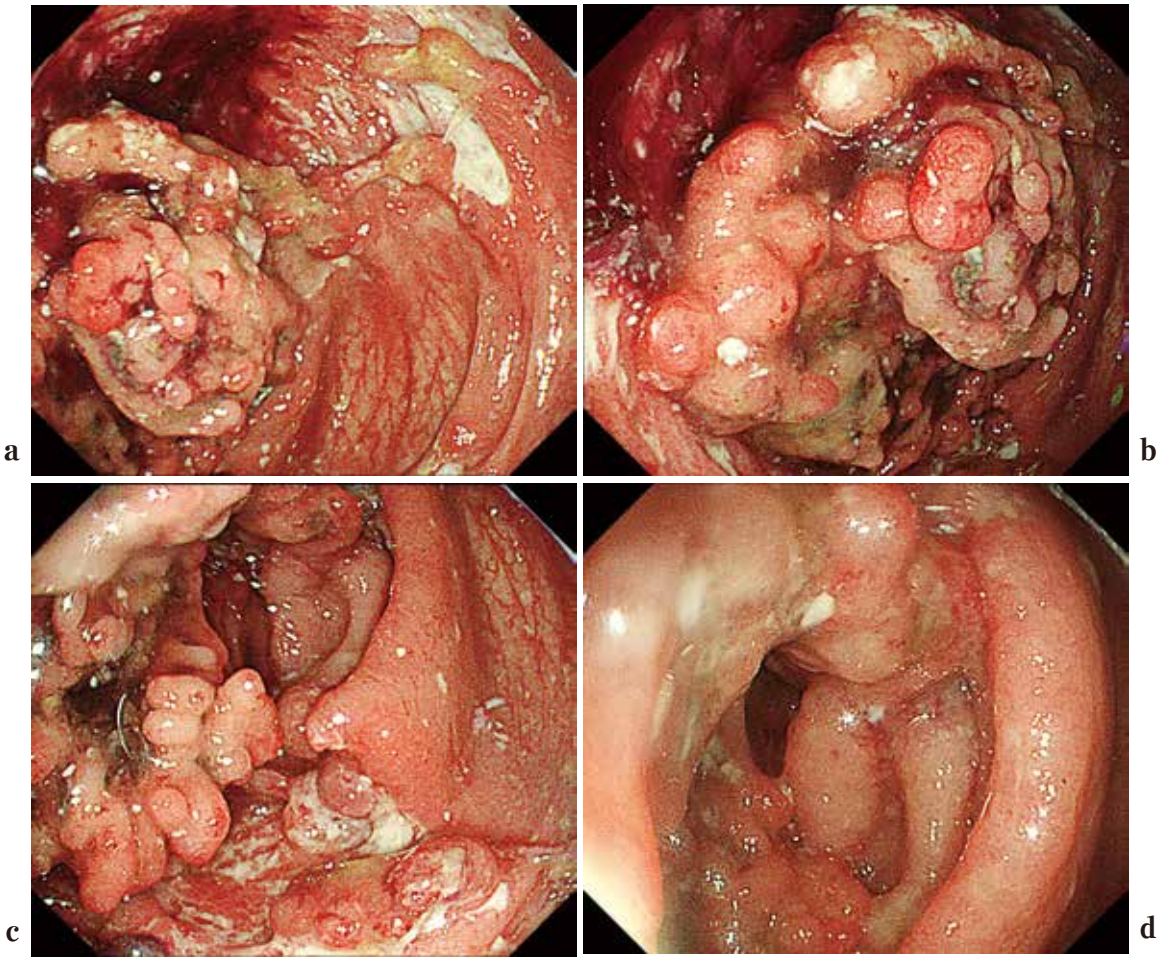

Figure 2 Colonoscopic findings.
a and **b** : Elongated polyps clustered together.
c : Deep ulcer surrounding polyps.
d : Stenosis of the colonic lumen.

Figure 3 Macroscopic findings of the resected right colon.
Long polyps were clustered in the ascending colon, forming a nodular lesion.

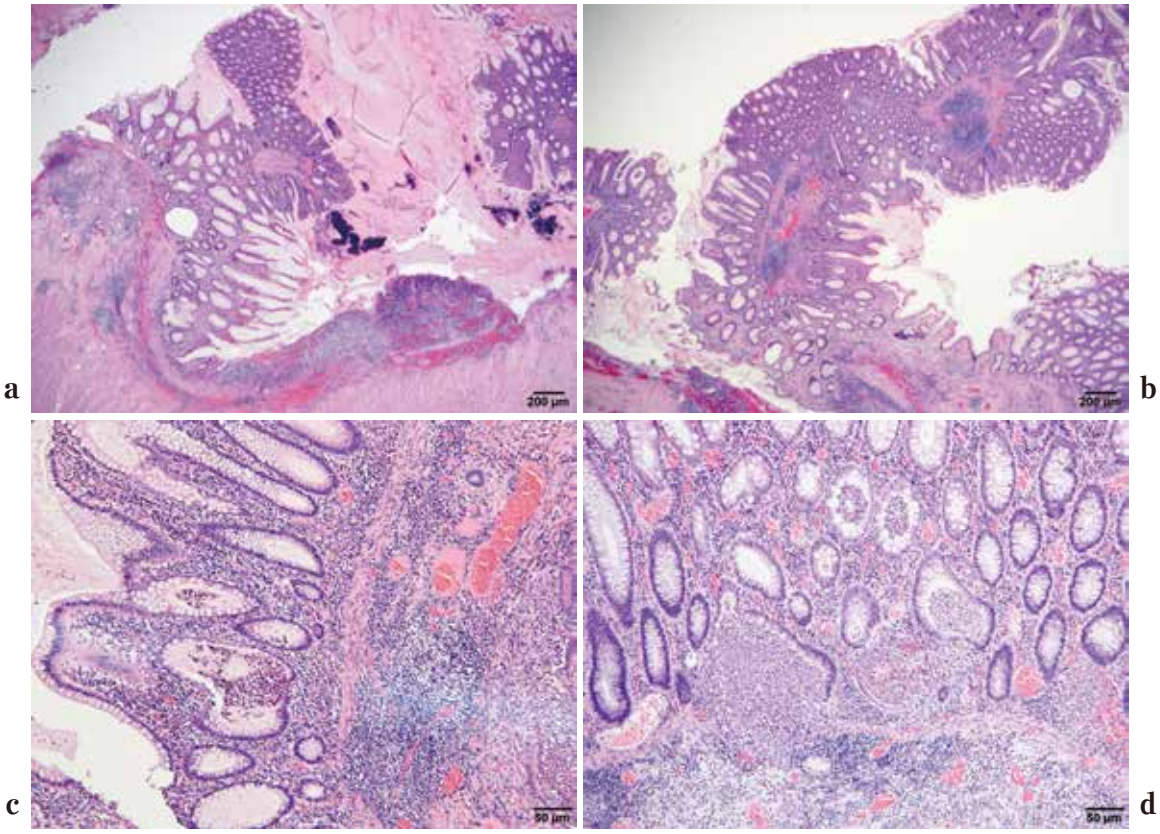

Figure 4 Microscopic pathological findings of the tissue preparations stained with hematoxylin and eosin.
a：(×2). Strong inflammation extending to the muscularis propria and serosa in the ascending colon.
b：(×2). and **c**：(×10). Submucosal structures including vascular and smooth muscle tissue were observed in the center of the polyp in the ascending colon.
d：(×10). Crypt abscess in the sigmoid colon.

DISCUSSIONS

UC is a chronic inflammatory bowel disease of unknown etiology characterized by repeated relapse and remission[2]. In UC, the process of tissue destruction and repair based on chronic mucosal inflammation often results in the formation of elevated lesions, or inflammatory polyps. However, as in this case, it is rare for inflammatory polyps to be localized and concentrated, resulting in LGIP and intestinal stenosis with stenotic symptoms. LGIP was first reported by Goldgraber in 1965 using the term localized giant pseudopolyposis, and since then, there have been several similar reports on LGIP[3]. Filiform polyposis, a pathological condition similar to LGIP, was described as a polyp with a characteristic morphology reported by Goldgraber[3], and in 1974 Appleman et al. defined filiform polyposis as a polyp with a filament to elongated shape[4]. In 1985, Brozna et al. defined filiform polyposis as a pathological condition in which the central submucosal layer of the polyp has a fibrovascular core that contains vascular and smooth muscle fiber growth, distinguishing it from typical inflammatory polyps[5]. Therefore, there is currently no consensus as to whether LGIP and filiform polyposis are the same disease.

Although etiology of LGIP development is unclear, Kelly et al. explained that large mucosal tags are initially formed when they are stretched, twisted, or wounded by fecal flow[6]. However, LGIP has been found in the ascending colon where stool is not solidified, as in this case. Thus it is likely that LGIP is caused by multiple factors, including traction by stool, peristalsis, physical stimulation by intestinal bacteria, or even factors in ulcer healing. In contrast, hyperplasia and disorganization of the neuromuscular layer and vascular connective tissue are observed in filiform polyposis, suggesting that filiform polyposis occurs in

the process of hamartoma formation[7].

Yada et al. reviewed 42 cases of LGIP reported in the literature[8]. According to the report, 37 of the 40 patients underwent colectomy, of which 25 underwent total colectomy. Total colectomy is more frequently performed in total colitis type cases. In addition, there are some reports of LGIP developing in patients with a short duration of UC, but in this case, the onset of LGIP was observed after 17 years of UC[9,10]. We chose to treat the patient with total colectomy rather than medical therapy due to disease severity, the high possibility of developing intestinal obstruction, and the possibility of UC-associated dysplasia and carcinoma. However, as according to Esaki et al. , internal medical therapy should first be given in a situation where surgery can be delayed, rather than invasive surgery being the first choice[11]. For internal medical therapy, the efficacy of anti-tumor necrosis factor (TNF)-α inhibitors for UC with LGIP has been reported in a case report[12]. However, prospective studies including anti-TNF-α inhibitors, Janus kinase inhibitors, and integrin inhibitors are expected in the future.

CONCLUSIONS

We report a case of UC with stenosing symptoms caused by LGIP. Although LGIP is a rare phenomenon, it is necessary to understand the clinical features of LGIP in the follow-up of UC, because intestinal tract passage obstruction may require surgery.

Disclosure Statement

None of the authors reports any conflict of interest in this work.

Author Contributions

Yamaguchi S, Abe K, Tominaga K, and Irisawa A wrote the manuscript ; Tanaka T, Kanazawa M, Watanabe S, Ishikawa M, Teratani N, Kanamori A, and Goda K, contributed to the manuscript discussion and reviewed the manuscript ; Ihara K and Nakamura T performed the operation. Ishida K established pathological diagnosis.

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