—Case Reports—

A Case of an Inflammatory Fibroid Polyp of the Cecum

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Abstract

An inflammatory fibroid polyp (IFP) is a rare benign lesion, originating in the submucosa of the gastrointestinal tract. It is histopathologically characterized by distinctively arranged fibrous connective tissue and blood vessels with inflammatory cell infiltration. It typically arises in the stomach and small intestine but also arises infrequently in the colon. This report describes a case of IFP of the cecum. A 63-year-old woman presented with persistent bloody stool for more than 1 month. Colonoscopy revealed a polypoid lesion, measuring 2.5 cm in diameter and 4 cm in length, with a thick pedicle in the cecum. Histopathological examination of the biopsy specimen showed hyperplastic changes of the mucosa. The lesion was diagnosed to be a submucosal tumor. We concluded that endoscopic mucosal resection would be difficult because the polyp showed signs of infiltration into the submucosa. Furthermore, the possibility of malignancy could not be ruled out. Laparoscopy-assisted ileocecal resection with lymphnode dissection was performed after the patient's informed consent was obtained. The lesion was finally diagnosed to be IFP on the basis of histopathological examination of the resected specimen. Immunohistochemical staining of the spindle-shaped cells, which were present around the small vessels in the stroma of the tumor, showed that the tissue expressed vimentin but not α-smooth muscle actin, desmin, S-100, c-kit or CD 34. IFP is difficult to diagnose without the recognition of its clinical and pathological characteristics. It is also important to determinate the depth of the lesion before selecting the therapeutic method.
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Key words: inflammatory fibroid polyp, submucosal tumor of the colon, laparoscopy-assisted surgery, immunohistochemical staining

Introduction

Inflammatory fibroid polyp (IFP) is a rare lesion that arises in the submucosa of the gastrointestinal tract[1–6]. It is composed of fibrous tissue, a network of blood vessels, and inflammatory cells[1–6]. The lesion occurs most commonly in the stomach, generally in the antrum or prepyloric region, less frequently in the ileum, and only occasionally in the colon, jejunum, duodenum and esophagus[1–6]. Although IFP is benign, laparotomy is necessary for resection in

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most cases because of the size of the lesion[5] and the difficulty of establishing a definitive diagnosis on the bases of biopsy specimens[6]. This report describes a case of an IFP in the cecum which was removed with laparoscopy-assisted ileocecal resection and finally diagnosed with a histopathological and immunohistochemical examination of the resected specimen.

Case Report

A 63-year-old woman complaining of hematochezia of 1 month's duration consulted this hospital. She had previously undergone resection of a histeomyoma at 45 years of age and an endoscopic polypectomy of a benign sigmoid colon polyp at 59 years of age. The patient's father had died of bladder cancer. Physical examination revealed no abnormalities. A pedunculated polypoid tumor in the cecum was found with a colonoscopy. Pedunculated early-stage colon cancer with stem invasion or a submucosal tumor was the initial diagnosis based on endoscopic findings, but hyperplastic mucosa with mature granulocytes and lymphocyte infiltration was recognized on histopathological examination of the biopsy specimens, and the lesion was diagnosed as a hyperplastic polyp. A second colonoscopic examination was performed (**Fig. 1**), and histopathological examination of biopsy specimens was performed again, but there were no malignant findings. In addition, the lesion was accompanied by edema and an inflammatory cell infiltration in the submucosa, which was diagnosed as normal mucosa. On the basis of its invasion of the submucosal layer, the lesion was judged to be difficult to remove completely with endoscopic mucosal resection. Because the patient frequently complained of hematochezia, resection of the lesion was deemed necessary. Therefore, laparoscopy-assisted ileocecal resection was planned after informed consent had been obtained from the patient and her family.

The laboratory studies at admission showed no signs of inflammation; no abnormalities were indicated, except anemia (hemoglobin, 10.2 mg/dL). No abnormal findings were detected on radiography of the chest and abdomen. A barium enema study revealed a pedunculated lesion with a thick stalk in the cecum (**Fig. 2**). The cecal lesion was not identified with abdominal computed tomography (CT), and no lymph-adenopathy of the cecal circumference was observed. During the operation the cecal circumferential lymph nodes were dissected because the possibility of malignancy could not be ruled out. The resected specimen had a diameter of 2.5 cm and included a 4.0-cm-long pedunculated tumor in the cecum (**Fig. 3**). Histopathological examination showed that the tumor arose from the submucosal layer and included inflammatory cell infiltration. Collagen fibers and a convoluted arrangement of spindle-shaped cells seemed to surround the small vessels; this finding was characteristic of an IFP (**Fig. 4**). No malignant cells or signs of an infiltration into the muscularis propria were observed. An immunohistochemical examination showed that the spindle-shaped cells in the circumference of the small blood vessels expressed vimentin but not desmin, α-smooth muscle actin (α-SMA), S-100, c-kit, or CD 34 (**Fig. 5**). The lesion was finally diagnosed as an IFP that had arisen from the cecum.

The postoperative course was good, and the patient was discharged after 15 days. There were no signs of the recurrences based on the colonoscopy and CT images obtained 1 year after the operation.

Discussion

IFP was first described as a "gastric fibroma" by Konjetzny[7] in 1920. In 1949, Vanek[8] described it as "gastric submucosal granuloma with eosinophilic infiltration." In 1953, Helwig and Ranier[9] proposed the term "IFP" for a gastric polypoid lesion that had three histological characteristics: various-sized blood vessels in the stroma of the lesion, fibrous connective tissue consisting of spindle-shaped cells around the blood vessels (onion skin appearance), and nonspecific inflammatory cell infiltration, especially eosinophils[9]. However, at present eosinophilic infiltration may not be necessary for the definitive diagnosis of an IFP[4]. IFPs arise in the submucosa of the gastrointestinal tract, most often in the stomach and small intestine but rarely in the

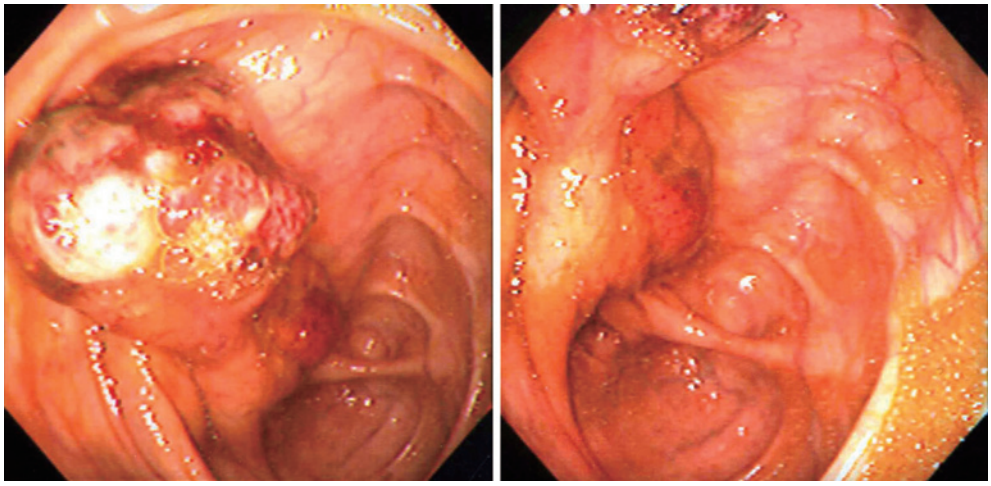

Fig. 1 Colonoscopy revealed a large pedunculated polyp, without a tumor-free stalk, in the cecum.

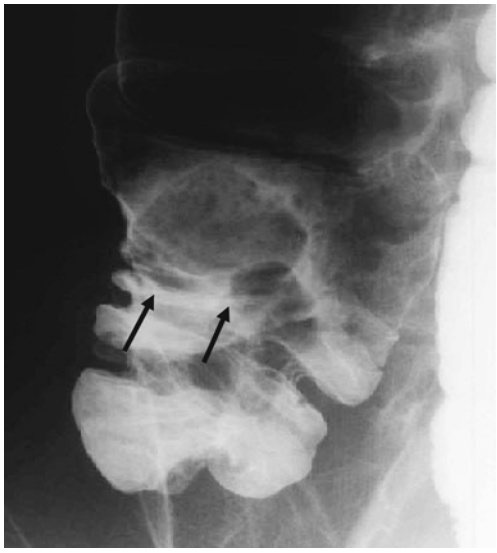

Fig. 2 A barium enema examination showed a large pedunculated polyp with a thick stalk in the cecum.

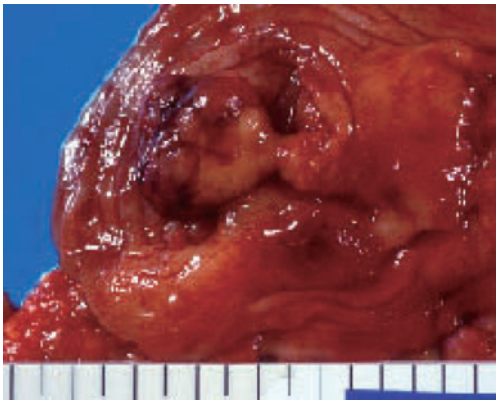

Fig. 3 Macroscopic view of the resected specimen with a 2.5×1.5×4.0-cm polypoid mass.

large intestine[1–6]. The first case of IFP of the colon was reported by Kofler[10] in 1952. In Japan, Nishihara et al[11]. reported the first case of a colonic IFP in 1978. To date, 35 cases of colorectal IFP have been reported in Japan[5,11–13]. The details of the reported cases, including the present case, are summarized in **Table 1**. Women (25 of 36 patients) were affected more frequently than were men. The age at onset of the symptoms ranged from 1 year to 72 years (mean, 47.3 years). Most colorectal IFPs were located proximally, and the size of the lesions ranged from 0.5 to 14.3 cm in diameter. The macroscopic appearance was pedunculated in 53% of cases (19 of 36 cases), and sessile in 39% (14 of 36 cases). Iwamoto et al[12]. have recently reported an interesting case of a colonic depressed (IIa + IIc) IFP, which magnifying colonoscopy showed to have a type V_I pit pattern in the central depression. The chief clinical symptoms of colorectal IFPs are abdominal pain, occult blood in the stool, and hematochezia. There were 7 proximal cases which presented as intussusception, which is more frequently observed with IFPs of the small intestine[3].

The etiology of IFP remains unknown[1–6]. Some authors have proposed that IFP is caused by an allergic reaction to an inflammatory stimulus, such as bacteria, chemicals, and trauma[9], or that it is a reactive lesion of fibroblastic or myofibroblastic nature[5]. Electron microscopy shows that the predominant cells of an IFP are the mesenchymal

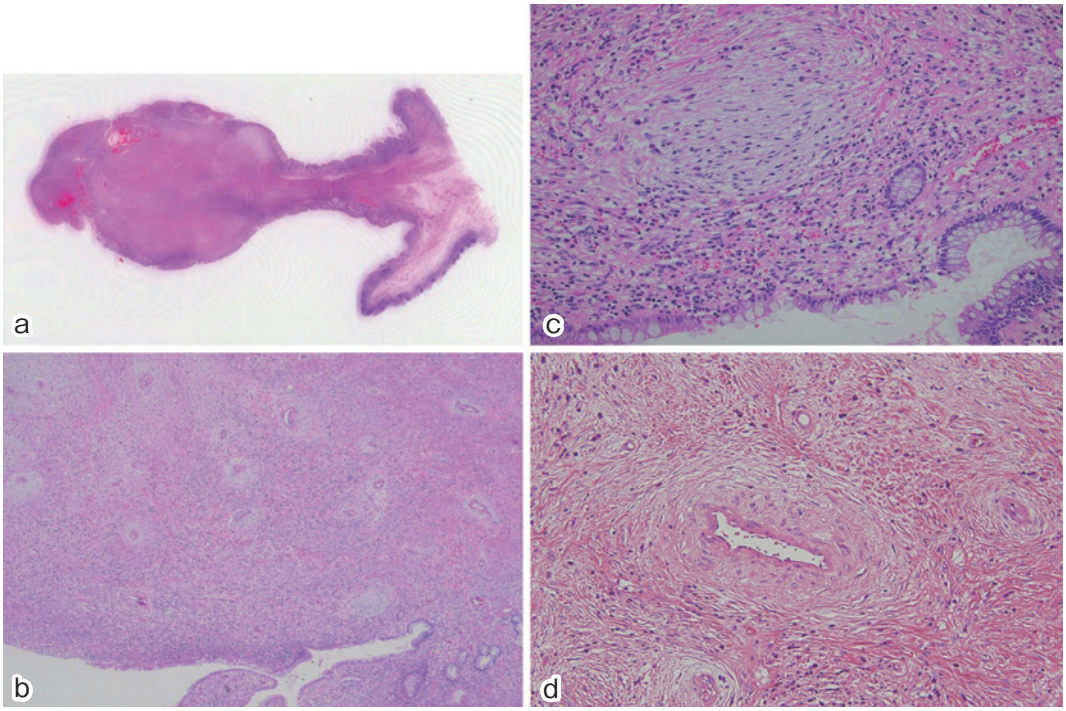

Fig. 4 Histological examination of the pedunculated polypoid lesion in the cecum established the diagnosis of IFP (hematoxylin and eosin staining). **a**: Scanning magnification **b**: Low-power magnification (×40) **c**: Middle-power magnification (×100) **d**: High-power magnification (×200) Microscopic view of the polypoid lesion showed the perivascular arrangement of the spindle-shaped cells and inflammatory cell infiltration.

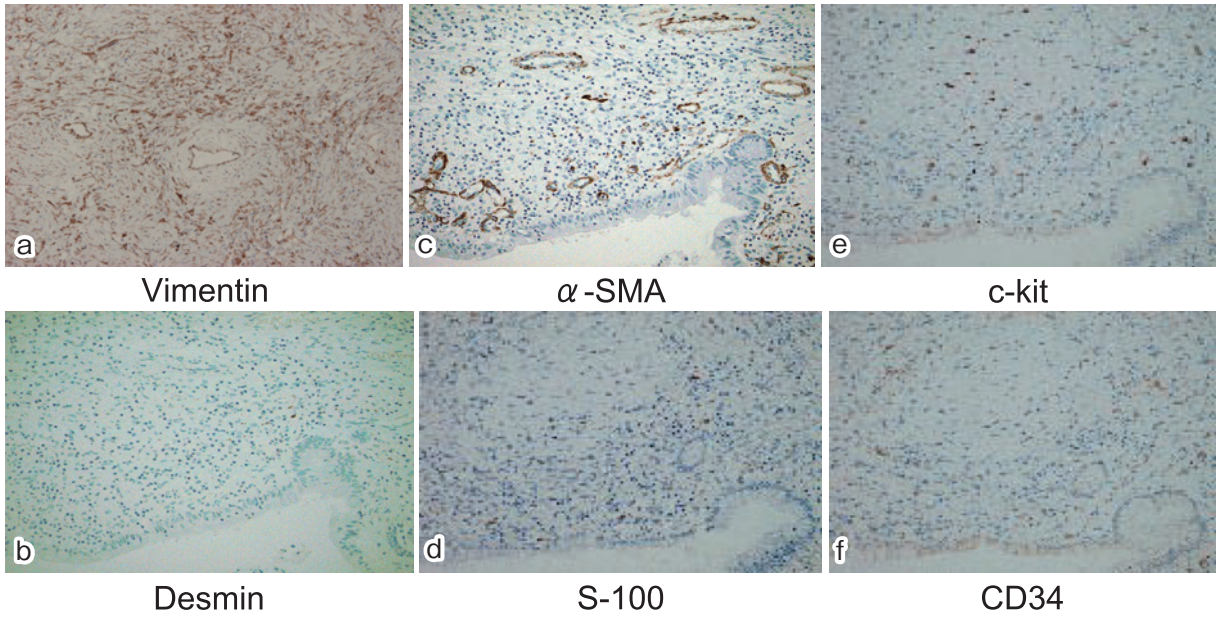

Fig. 5 Immunohistochemical staining

The lesion demonstrated defuse expression of vimentin **(a)**. Desmin, α-SMA, S-100, c-kit, CD 34 **(b – f)** were not expressed; however, cells in the endothelium of the small blood vessels expressed α-SMA **(c)**.

fibroblasts, and neurogenic or vasculogenic origin was denied[2]. In the present case, the results of immunohistological staining for markers, such as desmin, α-SMA, and S-100, ruled out a myogenic or neurogenic origin. In addition, c-kit and CD 34 are known to be expressed at high levels in gastrointestinal stromal tumors (GISTs), which consist of spindle-shaped cells, as do IFPs. However,

Table 1 Summary of reported cases of colonic IFP in Japan (n＝36)

Gender (male : female)	11 : 25	Chief Complaints	
Age	1－72 (mean 47.3)	abdominal pain	11 (31%)
Location		stool occult blood positive	7 (19%)
Appendix	3 (8%)	hematochezia	5 (14%)
Cecum	11 (31%)	anemia	3 (8%)
Ascending colon	3 (8%)	fever	2 (6%)
Transverse colon	11 (31%)	others	6 (17%)
Descending colon	2 (6%)	unknown	2 (6%)
Sigmoid colon	5 (14%)	Therapeutic method	
Rectum	1 (3%)	Surgical resection	25 (69%)
Size (cm)	0.5－14.3 (mean 3.2)	Open	20 (56%)
≧ 1.0	5 (14%)	Laparoscopy-assisted	5 (14%)
1.1－2.0	7 (19%)	Endoscopic resection	10 (28%)
2.1－3.0	10 (28%)	Biopsy only	1 (3%)
3.1－6.0	11 (31%)	Immunohistochemical findings	
6.1 ≦	3 (8%)		No. of the positive case/ No. of the case reported
Macroscopic findings		Vimentin	5/6
sessile	14 (39%)	Desmin	0/5
pedunculated	19 (53%)	S-100	0/8
tumor like	2 (6%)	α-SMA	0/2
depressed	1 (3%)	c-kit	0/1
		CD 34	1/2
		NSE	0/1
		Factor Ⅷ	0/2

because neither c-kit nor CD 34 was expressed in the present case, GIST was also ruled out. Kolodziejczyk et al[14]. have reported the immunohistochemical profiles of 46 gastric IFPs. They demonstrated diffuse expression of vimentin by spindle-shaped cells in an IFP, variable expression of SMA and CD 34, and no expression of desmin, S-100, c-kit, factor VIII, or neuron-specific enolase (NSE). These results are consistent with the reported cases of colonic IFPs in Japan (**Table 1**).

There are no distinctive radiologic features that can be used to differentiate IFPs from other mural or intramural lesions of the gastrointestinal tract[15]. The endoscopic findings tend to reveal solitary pedunculated or sessile polypoid lesions with surface erosion of the overlying mucosa. IFP is difficult to diagnose, and histological confirmation is necessary in almost all cases[4,6,15]. Because of the submucosal origin of IFPs, endoscopic biopsy may not always yield sufficient tissue for diagnosis[6], as in the present case. Although an IFP is a benign lesion, laparotomy is needed for resection of a colonic IFP in most cases because the polyp is usually large[5] and difficult to diagnose preoperatively. In the present case, the lesion was diagnosed before histological confirmation to be 2.5-cm-diameter pedunculated submucosal tumor. Furthermore, it was impossible to rule out malignancy in this case. Therefore, laparoscopy-assisted ileocecal resection was performed because it is less invasive than open laparotomy surgery.

Several cases of small IFPs that were resected endoscopically have been reported[5,13,16,17]. However, it is difficult to judge whether endoscopic resection or surgery is appropriate to treat colorectal IFPs. Recently, endoscopic ultrasonography (EUS) has been considered to be effective for the diagnosis of submucosal tumors of the gastrointestinal tract. The most frequent EUS features of IFPs are an indistinct margin and a hypoechoic, homogeneous echo pattern[18]. In addition EUS is useful for determining the depth of the lesions and for evaluating whether they can be resected endoscopically[5]. Ten cases of the 36 (28%) colorectal IFPs reported in Japan were removed through endoscopic resection (**Table 1**). In these cases, EUS should be performed to determine whether endoscopic resection can be safely

performed[5]. Fukada et al[13]. have reported a case of a sessile colonic IFP 1 cm in diameter which was persisted 6 months after endoscopic resection and required a laparoscopy-assisted colectomy for resection.

In summary, this case and those previously reported the literatures conform that an IFP is difficult to diagnose without recognition of its clinical and pathological characteristics. Furthermore, it is important to determine the depth of the lesion by EUS before the type of resection is selected.

References

1. Johnstone JM, Morson BC: Inflammatory fibroid polyp of the gastrointestinal tract. Histopathology 1978; 2: 349–361.
2. Shimer GR, Helwig EB: Inflammatory fibroid polyps of the intestine. Am J Clin Pathol 1984; 81: 708–714.
3. Fujiyama K, Katsuta T, Nakamura A, et al.: A case of inflammatory fibroid polyp of ileum—Review of inflammatory fibroid polyp of small intestine in Japan—. Nippon Syokakibyo Gakkai Zasshi 1991; 88: 1245–1249 (In Japanese).
4. Hayashi Y, Arita T, Morimatsu M: Inflammatory fibroid polyp of the intestine. Nippon Rinsho 1994; 6 (Suppl): 336–339 (In Japanese).
5. Nakase H, Mimura J, Kawasaki T, et al.: Endoscopic resection of small inflammatory fibroid polyp of the colon. Intern Med 2000; 39: 25–27.
6. Ozolek JA, Sasatomi E, Swalsky PA, Rao U, Krasinskas A, Finkelstein SD: Inflammatory fibroid polyps of the gastrointestinal tract: Clinical, pathologic, and molecular characteristics. Appl Immunohistochem Molecul Morphol 2004; 12: 59–66.
7. Konjetzny GE: Über Magenfibrome. Beitr Klin Chir 1920; 119: 53–61 (In Germany).
8. Vanek J: Gastrointestinal submucosal granuloma with eosinophilic infiltration. Am J Pathol 1949; 25: 397–411.
9. Helwig EB, Ranier A: Inflammatory fibroid polyps of the stomach. Surg Gynecol Obstet 1953; 96: 355–367.
10. Kofler E: Über die Granulome des Magen-Darmschlauches. Arch Pathol Anat 1952; 321: 121–133 (In Germany).
11. Nishihara K, Mizushima M, Ito I, Nagai U: A case of inflammatory fibroid polyp (Helwig) of the cecum. Gann no Rinsho 1978; 24: 1311–1316 (In Japanese).
12. Iwamoto K, Sakashita M, Takahashi T, et al.: Depressed type of inflammatory fibroid polyp of the colon. Int J Colorectal Dis 2006; Jun 28: Online First™ 107; Letter to the editor.
13. Fukada M, Sugisaka M, Kijima H, et al.: A case of inflammatory fibroid polyp of the transverse colon. Tama Sym J Gastroenterol 2003; 17: 33–37 (In Japanese).
14. Kolodziejczyk P, Yao T, Tsuneyoshi M: Inflammatory fibroid polyp of the stomach: A special reference to an immunohistochemical profile of 42 cases. Am J Surg Pathol 1993; 17: 1159–1168.
15. Harned RK, Buck JL, Shekitka KM: Inflammatory fibroid polyps of the gastrointestinal tract: Radiologic evaluation. Radiology 1992; 182: 863–866.
16. Wei SC, Wong JM, Shieh MJ, Sun CT, Wang CY, Wang TH: Endoscopic resection of gastrointestinal submucosal tumors. Hepato-gastroenterology 1998; 45: 114–118.
17. Tada S, Iida M, Yao T, et al.: Endoscopic removal of inflammatory fibroid polyps of the stomach. Am J Gastroenterol 1991; 86: 1247–1250.
18. Matsushita M, Hajiro K, Okazaki K, Takakuwa H: Gastric inflammatory polyps: endoscopic ultrasonographic analysis in comparison with the histology. Gastrointest Endosc 1997; 46: 53–57.

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