Adult Ileo-ileal Intussusception Due to Inflammatory Fibroid Polyp: A Case Report

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Introduction: Intussusception occurs when one part of the intestines slides into the adjacent intestine resulting in bowel obstruction. It is a rare condition in adults, accounting for only 5% of all intussusceptions. It has multiple causes, with inflammatory fibroid polyps (IFPs) very infrequently being the cause. We present a rare case of intussusception in an adult due to an IFP.
Case presentation: A 72-year-old woman visited our hospital complaining of abdominal pain. Abdominal contrast-enhanced computed tomography (CT) demonstrated an ileo-ileal intussusception due to a round mass. An emergency surgery involving a partial ileal resection with laparoscopic assistance was performed. Pathological findings of the tumor showed proliferation of spindle-shaped cells, edematous stroma, dilation of lymphatic vessels, and infiltration of inflammatory cells, which were mainly eosinophils. Immunohistochemistry was positive for vimentin and SMA and negative for CD117, CD34, S-100, and desmin. Based on these findings, the tumor was diagnosed as an IFP.
Conclusion: Bowel obstruction in adults due to intussusception is rare, and those due to IFPs are even more rare. Preoperative diagnosis of IFP is difficult, but surgeons must keep in mind that it can be a cause of adult intussusception.

Key words: inflammatory fibroid polyp, adult intussusception

INTRODUCTION

Intussusception occurs when one part of the intestine slides into the adjacent intestine, causing bowel obstruction. It is common in children, but adult intussusception is a rare condition, accounting for 5% of all cases. Furthermore, it is a rare cause of intestinal obstruction in adults, occurring in < 1% of obstructions [1, 2]. Intussusception caused by inflammatory fibroid polyps (IFPs), which are relatively rare benign lesions of the gastrointestinal tract, is uncommon. In this case report, we discuss a case of adult ileo-ileal intussusception due to an IFP.

CASE REPORT

A 72-year-old woman visited our hospital complaining of abdominal pain for 1 month. She had no significant medical or family history. An upper gastrointestinal endoscopy was performed and no abnormalities were noted. However, she continued to have abdominal pain and vomiting, and was referred to our hospital. Her body temperature was 36.9˚C, pulse rate 76 beats per minute, and her blood pressure was 111/76 mmHg. Blood biochemistry showed no abnormal findings except for an increase in leukocytes at 12400/μl. An abdominal X-ray showed gas with mirror surface formation in the small intestine (Fig. 1). Abdominal contrast-enhanced computed tomography (CT) demonstrated an ileo-ileal intussusception with dilated proximal small bowel loops due to a well-defined rounded enhancing endoluminal mass (Fig. 2). From the above findings, intussusception with intestinal obstruction due to a small intestinal tumor was diagnosed, and an ileus tube was inserted for the purpose of intestinal decompression. We decided to perform emergency surgery on the same day with laparoscopic assistance. One port was placed into the navel via a 12-mm trocar and another 2 into the lower left and lower right abdomen via 5-mm trocars. Intraoperative laparoscopy showed intussusception in the ileum 120 cm proximal to the ileocecal valve. That section of the ileum was pulled out of the body through a small incision without releasing intussusception (Fig. 3). Partial ileal resection and functional end-to-end anastomosis were performed. The surgical specimen showed a 28 × 32 × 42-mm polypoid tumor covered with normal mucous membrane and the surface layer formed erosion, we suspected submucosal tumor such as gastrointestinal stromal tumor (GIST) from the macroscopic findings (Fig. 4). Pathological findings of the tumor showed proliferation of spindle-shaped cells, edematous stroma, dilation of lymphatic vessels, and Infiltration of inflammatory cells, which were mainly eosinophils in the submucosal layer (Fig. 5a, b).

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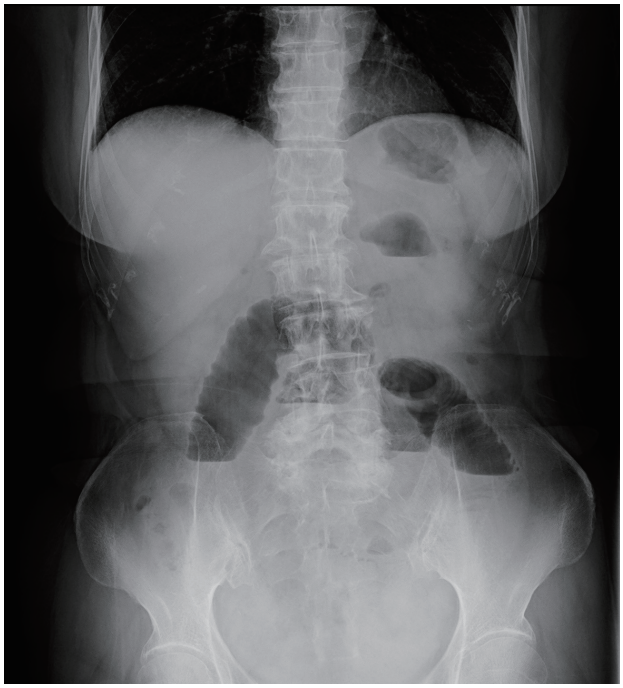

Fig. 1 X-ray of the abdomen: X ray showed small intestinal gases with mirror surface formation.

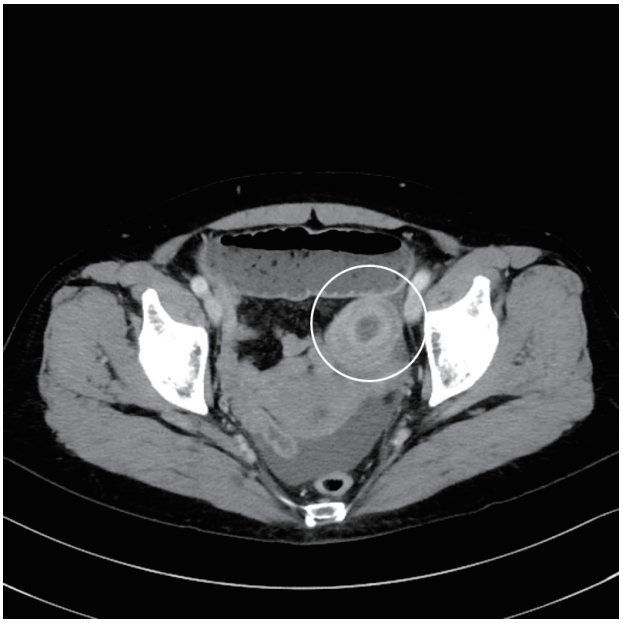

Fig. 2 Abdominal contrast-enhanced computed tomography (CT): Abdominal contrast-enhanced CT demonstrated an ileo-ileal intussusception due to a well-defined rounded enhancing endoluminal mass (white circle).

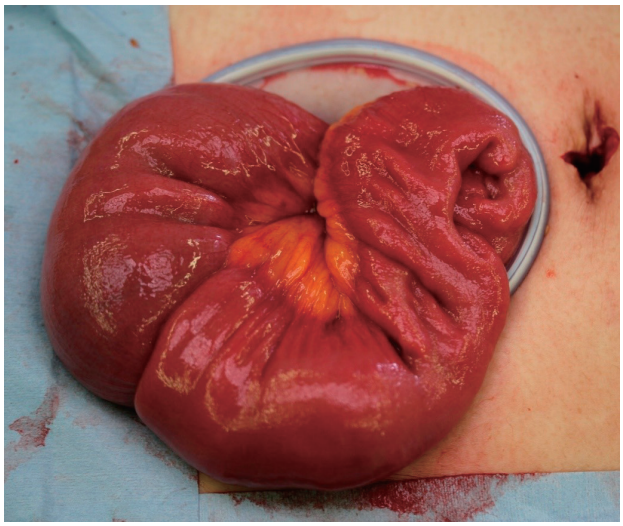

Fig. 3 Intraoperative findings: The ileum was the site of intussusception confirmed during operation.

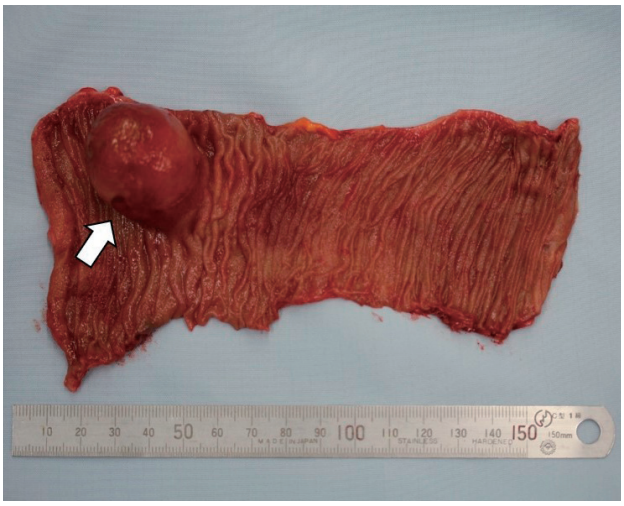

Fig. 4 Surgical specimen: Polypoid tumor with erosion was found in the ileum (white arrow).

Immunohistochemistry was positive for vimentin and smooth muscle actin (SMA) and negative for CD117, CD34, S-100, and desmin (Fig. 6a–f). A diagnosis of IFP of the ileum was made. Cellular proliferation was studied using Ki-67, which showed rare mitotic figures. The patient had no specific post-operative complications. She was discharged on the 9th postoperative day. There was no recurrence at the 6-month follow-up.

DISCUSSION

Intussusception is common in children but is rare in adults, accounting for 5% of intussusceptions. It is a rare cause of intestinal obstruction in adults as < 1% of bowel obstruction cases are attributable to it [1, 2]. Adult intussusception is caused by the following: malignant tumors, benign tumors, and idiopathic causes with each accounting for 32.9%, 37.4%, and 15.1% respectively [3]. Causes of small bowel intussusception include hamartomas, lipomas, adhesions, Meckel's diverticulum, lymphoid hyperplasia, trauma, intestinal duplication, and tuberculosis. Colon adenocarcinoma is considered the most important cause in cases of large bowel intussusceptions [4].

IFPs are relatively rare benign mesenchymal neoplasms of the gastrointestinal tract, first described by Vanek in 1949 as an eosinophilic submucosal granuloma [5]. Helwig and Ranier proposed the term inflammatory fibroid polyp in 1953 for a gastric polyp, and this name has gained acceptance for similar lesions throughout the gastrointestinal tract [6]. IFPs have a slight female predominance and are seen more frequently in the sixth and seventh decades of life [7, 8]. IFPs can develop in many different locations in the gastrointestinal tract, with the most common being the

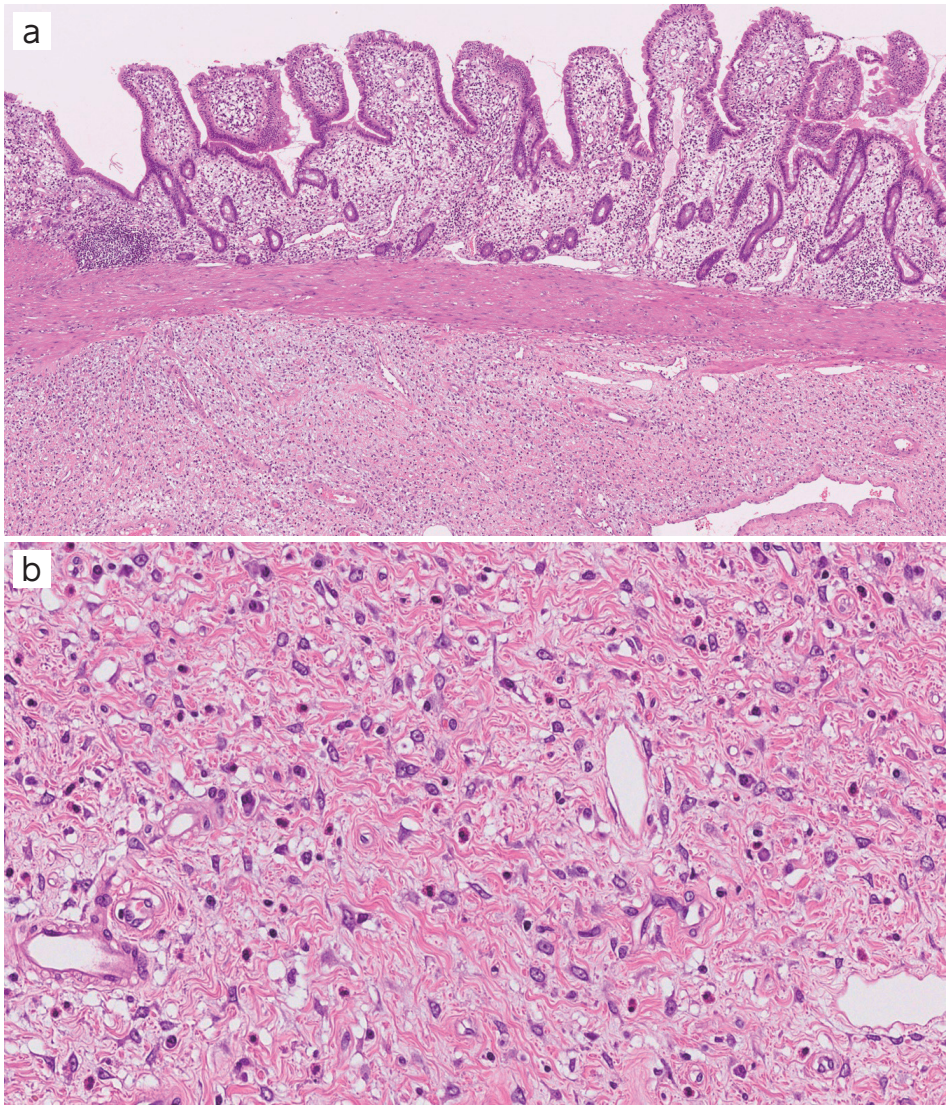

Fig. 5 Pathological findings: The tumor consisted of a proliferation of spindle-shaped cells, edematous stroma, dilation of lymphatic vessels and Infiltration of inflammatory cells, mainly eosinophils in the submucosal layer (hematoxylin and eosin staining; magnification, (a): × 4, (b): × 400).

gastric antrum (60–70%), followed by the small intestine (18–20%), colorectum (4–7%), and far less commonly (1%) in the esophagus, duodenum, gallbladder, and appendix. However, the ileal segment is the most common site where these polyps cause intussusception [9].

IFPs in the small intestine are usually asymptomatic and are typically discovered incidentally as occurred in our case. Gastric polyps may present with abdominal pain and may be diagnosed on upper gastrointestinal endoscopy. IFPs in the small intestine are unlikely to produce symptoms unless they lead to obstruction with or without intussusception [10]. Colicky abdominal pain, nausea, vomiting, constipation, and abdominal distention are the most common symptoms seen in these cases [11]. Although rare, gastrointestinal bleeding caused by giant gastric IFPs have been reported [12]. Most of these polyps usually measure between 2 and 5 cm in diameter, although giant IFPs up to 20 cm have also been reported [13, 14]. The pathogenesis of IFPs remains unclear, but trauma, allergy, genetics, as well as bacterial, physical, chemical, and metabolic stimuli have been suggested as initiators of the inflammatory process [9].

One of the characteristic histological features of IFPs is an onion skin appearance, which consists of fibroblast-like spindle cells arranged in a concentric formation around vessels [15]. Liu *et al.* reported 2 alternative findings, i.e., short fascicular growth pattern and the presence of sparse eosinophils with prominent hyalinization, in addition to the characteristic histological features of IFPS [8]. IFPs may have the same pathological features as a GIST. Immunohistochemistry is useful to differentiate them. Although both are positive for CD34 and vimentin, CD117 (c-kit) is specific for GIST. Pantanowitz *et al.* and Unal *et al.* reported CD34 was positive in all IFP cases and CD117 was negative in all IFP cases. Vimentin, SMA, Fascin, and Calponin are positive in IFPs, but S-100 and desmin, which are positive in GIST, are negative in IFPs [15, 16]. In this case, both CD34 and CD117 were negative, but vimentin and SMA were positive, with CD117, S-100, and desmin negative; therefore, a diagnosis of IFP of the ileum was made.

Abdominal CT is the most useful imaging modality for tumor localization and size, but due to a lack

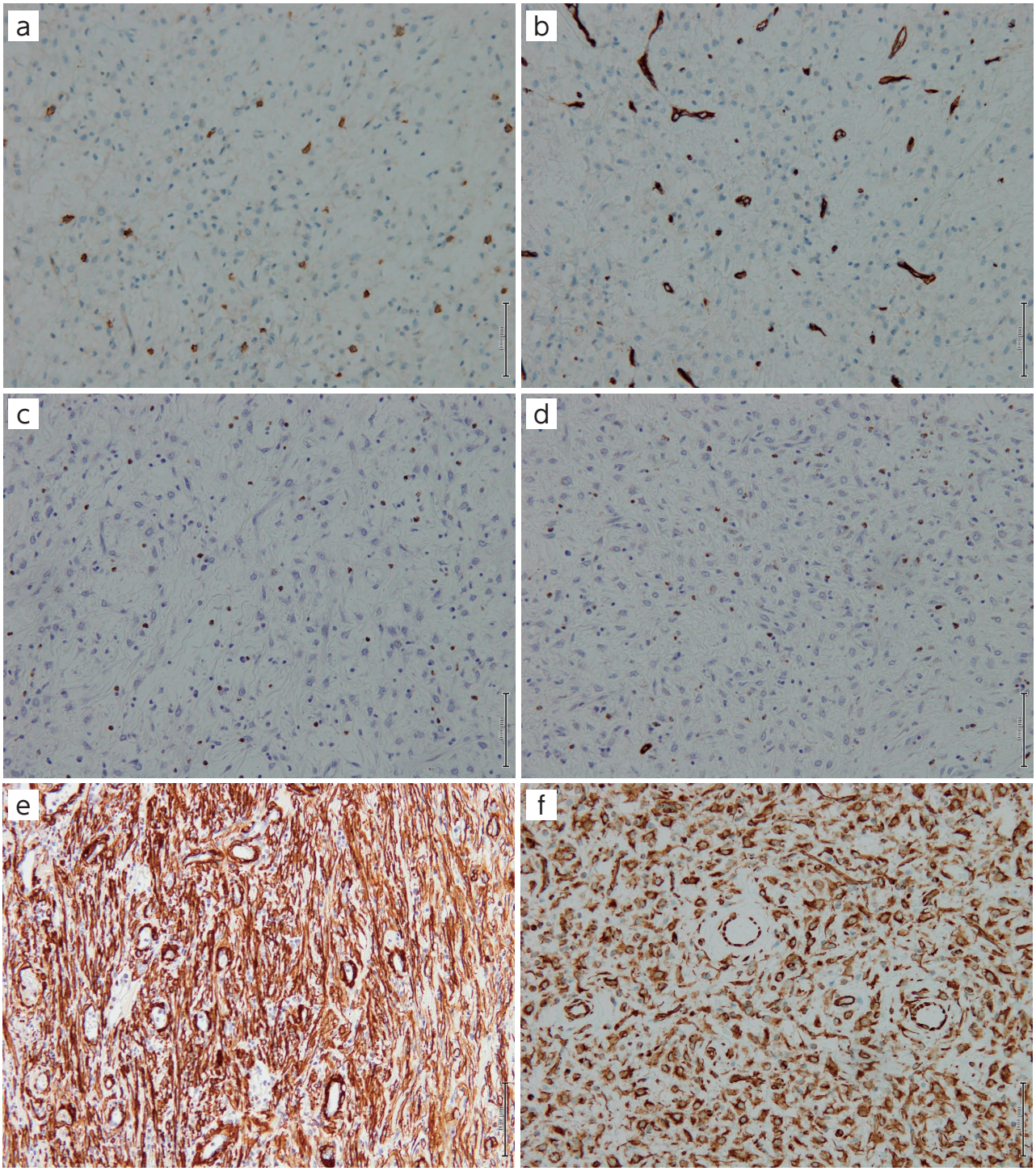

Fig. 6 Immunohistochemistry
Tumor cells were negative for CD117 (a), CD34 (b), Desmin (c), and S-100 (d) but positive for SMA (e) and vimentin (f).

of distinctive radiological features of IFPs, accurate preoperative diagnosis of IFPs on CT is difficult. The main differential diagnosis are GIST, inflammatory myofibroblastic tumor, and inflammatory polyps related to inflammatory bowel disease (Crohn disease and ulcerative colitis). Therefore, the diagnosis of IFPs is only possible with histological examination of endoscopic biopsy tissue or a surgically resected specimen. However, a retrospective study by Han *et al.*, which analyzed abdominal contrast-enhanced CT findings in 27 patients who were proven to have IFPs by histopathology, found common characteristic features. These include an endoluminal growth pattern, well-defined margins, a round or ovoid shape, lobulated contours, and various enhancement patterns [17]. In our case, a well-defined rounded enhancing endoluminal mass was noted. In recent years, double-balloon endoscopy and capsule endoscopy have been developed; therefore, preoperative diagnosis of IFPs of the small intestine may increase.

IFPs are usually treated with surgical resection, which is curative. However, 2 cases of polyp recurrence are found in the literature [18, 19]. In our case, wide resection of the intussuscepted segment was performed, as there was intraoperative concern for malignancy.

CONCLUSION

This case highlights the importance of considering intussusception as a differential diagnosis in the case of an adult presenting with abdominal pain. Adult intussusception is rare, and an intussusception due to IFPs is even rare. Preoperative diagnosis of IFP is difficult, but surgeons must keep in mind that it can cause adult intussusception.

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