

Single Case

Traditional Serrated Adenoma of the Ileum with Intussusception Successfully Treated with Laparoscopic Bowel Resection

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Keywords

Traditional serrated adenoma · Ileum · Intussusception

Abstract

The most common site of traditional serrated adenomas (TSA) is the area from the left colon to the rectum; however, there are few reports on TSA in the small intestine. Herein, we report a case of TSA of the ileum with intussusception that was diagnosed and successfully treated with laparoscopic bowel resection. The patient was a 29-year-old female with the chief complaint of recurrent abdominal pain and vomiting. Contrast-enhanced computed tomography showed a mass in the ileum and intussusception with the mass as the lead point. The patient was diagnosed with intussusception secondary to a small intestinal tumor. Due to the difficulty in endoscopic treatment resulting from the localization of the lesion, elective laparoscopic surgery was planned. Intra-abdominal examination revealed intussusception of the small intestine in the pelvic ileum, and an elastic soft mass 400 cm from the ligament of Treitz was identified at the lead point of intussusception. Partial laparoscopic resection of the small intestine was performed, with an operation time of 81 min, and a small amount of bleeding. The pathological diagnosis was TSA of the ileum, and the patient's postoperative course was good, with no complications. Seven months after the surgery, no recurrence of symptoms was observed. Therefore, from our case of TSA of the ileum with intussusception that was successfully treated with laparoscopic bowel resection, we conclude that when intussusception of the small intestine occurs, TSA of the ileum with malignant potential is possible, and early diagnosis by resection should be considered.

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Introduction

Serrated adenoma, a histological subtype of colorectal adenoma, is characterized by a serrated duct structure resembling hyperplastic polyps and neoplastic cell malformations resembling normal adenomas. These are relatively rare diseases, accounting for <1% of all colorectal polyps [1]. Serrated adenomas most often occur in the distal colon and are often treated endoscopically [2].

On the other hand, intussusception of the small intestine in adults is rare compared to that in children, and malignant tumors, hamartomas and lipomas, leiomyomas, schwannomas, adenomas, Peutz-Jeghers syndrome, and inflammatory fibroid polyps are considered differential diagnoses [3, 4]. The cause of small intestine intussusception is often difficult to diagnose due to its localization, and if symptoms persist, surgical resection is performed for symptom improvement and diagnosis.

To the best of our knowledge, serrated adenomas originating from the ileum are extremely rare, and there have been no reports of serrated adenoma diagnosed by intussusception. Herein, we report a case of traditional serrated adenoma (TSA) of the ileum with intussusception that was diagnosed and successfully treated with laparoscopic bowel resection.

Case Presentation

A 29-year-old female was admitted to our hospital with recurrent abdominal pain and vomiting. The patient had similar symptoms 2 weeks prior to presentation. After a thorough examination, she was diagnosed with intussusception of the small intestine and was hospitalized urgently. There was no medical, family, or dietary history. On physical examination, the abdomen was flat and soft, with tenderness in the lower abdomen and no peritoneal irritation symptoms. Additionally, laboratory data showed no abnormal values, and the tumor marker levels were within the normal range. Abdominal contrast-enhanced computed tomography showed a mass in the small intestine in the pelvic area and intussusception with the tumor as the lead point (Fig. 1a, b). The patient was diagnosed with intussusception secondary to a small intestinal tumor and was hospitalized for 5 days, with fasting and intestinal rest. However, there was no improvement in abdominal symptoms. Due to the difficulty in endoscopic treatment resulting from the localization of the lesion, elective laparoscopic surgery was planned. The operation was performed under general anesthesia in the supine position using three ports (Fig. 2). Intra-abdominal examination revealed intussusception of the small intestine in the pelvic ileum, with no other abnormal findings (Fig. 3a). After extracting the lesion from the body through a small laparotomy, the intussusception was removed manually, and an elastic soft mass, 3 cm in diameter, was identified at the lead point of the intussusception (Fig. 3b). The tumor was located approximately 400 cm from the ligament of Treitz. Partial laparoscopic resection of the small intestine was performed, with an operation time of 81 min and a small amount of bleeding. Macroscopic inspection of the resected ileum showed a pedunculated tumor measuring 29 × 27 × 24 mm in size (Fig. 4a).

Histopathological examination revealed typical serrated projections and extensive proliferation of ductal structures. In addition, a flat arrangement of eosinophilic cells and ectopic crypts were observed in some areas (Fig. 4b, c). Therefore, the diagnosis was TSA with negative KRAS and BRAF mutations. The patient's postoperative course was good, with no complications, and the patient was discharged on postoperative day 5. Seven months after the surgery, no recurrence of symptoms was observed. The CARE Checklist has been completed by the authors for this case report, attached as online supplementary material (for all online suppl. material, see www.karger.com/doi/10.1159/000529093).

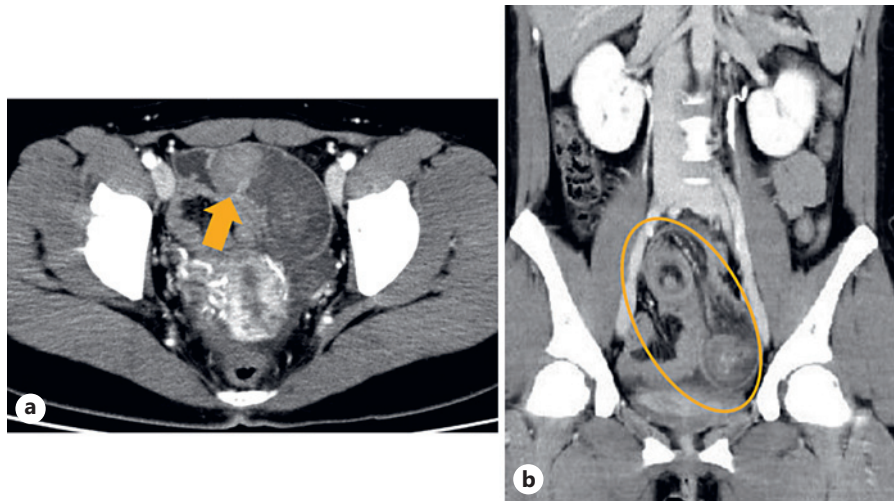


Fig. 1. Abdominal enhanced computed tomography imaging. **a** Tumor in the small intestine (pelvic area) (arrow). **b** Intussusception with the tumor as the lead point (circle).

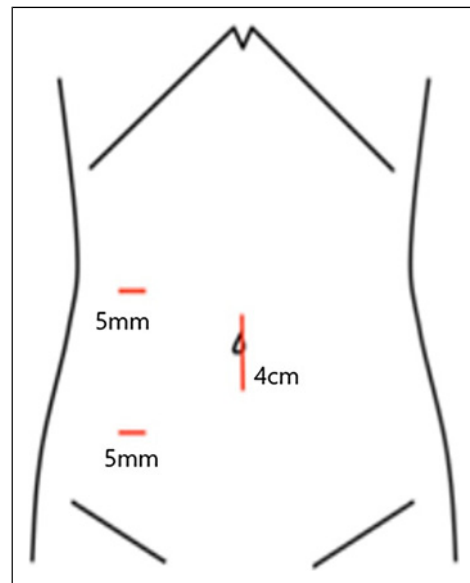


Fig. 2. Schema of the port position and skin incision.

Discussion

We encountered a case of serrated adenoma of the ileum, diagnosed as repeated intussusception. Currently, serrated lesions of the colon are classified into three categories according to the World Health Organization classification: hyperplastic polyposis, traditional serrated adenoma, and sessile serrated adenoma/polyp. Carcinogenesis also occurs in serrated lesions (serrated-neoplasia sequence), and the importance of diagnosing and treating these lesions has been recognized [5].

The most common site of TSA is the area from the left colon to the rectum, and it has been reported that the rate of malignant progression of colonic TSA ranges from 6.3 to 10% [6]. The mechanisms of malignant progression of TSA include TSA with APC mutations (20–36%), BRAF mutations (33–36%), and KRAS mutations (19–42%); additionally, p53 mutations are involved, which ultimately cause carcinogenesis [7]. Microsatellite stable colorectal cancer

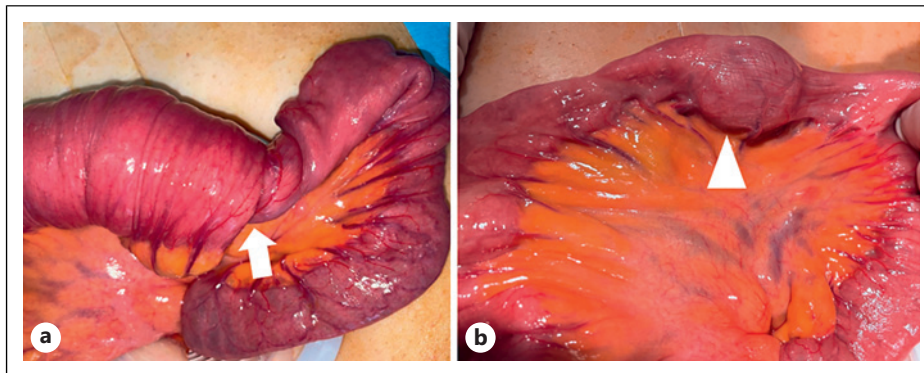


Fig. 3. Intraoperative findings. **a** Intussusception of the small intestine in the pelvic ileum (arrow). **b** An approximately 3-cm elastic soft mass 400 cm from the ligament of Treitz at the lead point of intussusception (arrowhead).

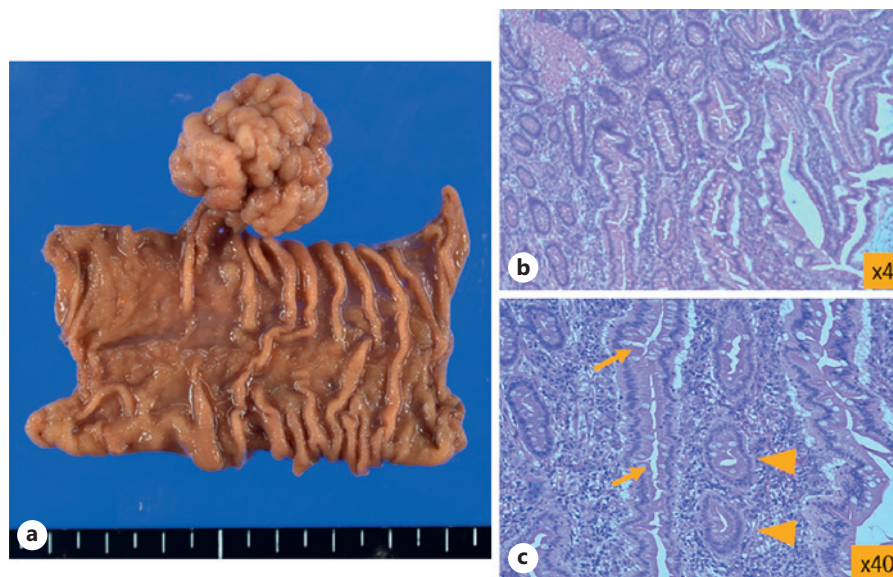


Fig. 4. **a** Macroscopic imaging of the ileum showing a pedunculated tumor ($\times 292 \times 7 \times 24$ mm). **b** Histopathological examination (HE $\times 4$) showing typical villiform projections. **c** Histopathological examination (HE $\times 40$) showing cells containing abundant brightly eosinophilic cytoplasm with centrally placed, slit-like serrations (arrow), and ectopic crypt formations (arrowhead).

with BRAF or KRAS mutation-positive TSA is known to have a poorer prognosis than sessile serrated adenoma/polyp-derived microsatellite stable or microsatellite instability colorectal cancer. In addition, recent studies have speculated that activation of the Wnt pathway and inactivation of p16 are involved; however, the details remain unknown [8].

Additionally, there are few reports of serrated lesions in areas of the gastrointestinal tract other than the colon. Primary lesions in the esophagus, stomach, duodenum, pancreas, and gallbladder have been reported, while 20 cases in the small intestine have been reported [9–11], of which 16 were in the duodenum, 2 in the jejunum, and only 2 in the ileum. Of the two cases in the ileum, both patients were male (aged 39 and 66 years), and both lesions were located in the terminal ileum and were diagnosed by biopsy. The maximum tumor diameters were 20 and 8 mm, respectively (Table 1).

Table 1. Characteristics of three reported cases of patients with TSA of ileum

Case	Age, years	Sex	Polyp location	Size, mm	Specimen type	Malignant progression within TSA	KRAS	BRAF
#1 (this case)	29	F	Ileum	30	Surgical specimen	None	Wild-type	Wild-type
#2	39	M	Terminal ileum	20	Forceps biopsy	None	c.35G>A mutation	Wild-type
#3	66	M	Terminal ileum	8	Forceps biopsy	None	None	None

TSA, traditional serrated adenomas; M, male; F, female.

Unlike the previous reports, in this case, the tumor was a pedunculated polyp with intussusception in a young woman. In addition, the symptoms were repeated, and the tumor was located in the pelvic ileum and was difficult to reach with an endoscope; therefore, early operation was required. Laparoscopic surgery, which is minimally invasive, was effective because the intussusception could be identified by laparoscopic observation and the lesion in the pelvic small intestine could be easily pulled from the umbilical incision. Furthermore, laparoscopic surgery has the advantage of an esthetic outcome in young women. In this case, the pathological diagnosis showed no KRAS or BRAF mutations, and no malignant findings were observed.

Several reports have suggested that, compared to colorectal TSA, TSA in the upper gastrointestinal tract has a higher malignant potential [9]. Moreover, of the 73 reported cases of TSA in the upper digestive tract, 53.4% ($n = 39$) showed simultaneously growing invasive carcinomas. TSA lesions originating in the upper gastrointestinal tract progress rapidly; therefore, early diagnosis and resection by endoscopic or surgical resection are desirable. However, the early diagnosis of tumors originating from areas of the small intestine other than the duodenum or terminal ileum is difficult; therefore, early surgical resection should be considered when symptoms appear or when abnormal radiographic findings are obtained. In addition, TSA is usually pedunculated, and the maximum tumor size often exceeds 50 mm, which easily causes intussusception; therefore, TSA should be considered differentially when intussusception occurs [12].

TSA of the ileum is extremely rare, and there are many unclear aspects, including the degree of malignancy and treatment. However, when intussusception of the small intestine occurs, TSA of the ileum with malignant potential is possible; therefore, early diagnosis by resection is recommended in these cases. We encountered a case of serrated adenoma of the ileum with intussusception that was successfully treated with laparoscopic bowel resection.

Statement of Ethics

Written informed consent was obtained from the patient for the publication of this case report and any accompanying images. This study protocol was reviewed, and the need for approval was waived by Ethics committee of International University of Health and Welfare Hospital.

Conflict of Interest Statement

The authors declare that they have no competing interests.

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Author Contributions

Ryo Nishide: study design, data collection, data analysis, and writing. Teppei Kamada: critical revision. Yutaka Suzuki: final approval of the article. Junji Takahashi, Keigo Nakashima, Eisaku Ito, Yuichi Nakaseko, Norihiko Suzuki, Masashi Yoshida, and Hironori Ohdaira: data collection. All authors read and approved the final manuscript.

Data Availability Statement

All data generated or analyzed during this study are included in this article and its online supplementary material. Further inquiries can be directed to the corresponding author.

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