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Case Report

Case Report: Repetitive Surgical Resections for Intestinal Intussusception due to Multiple Ileal Lipomatosis

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Adult intestinal intussusception is a rare disease known to be associated with intestinal tumors. We describe a case requiring partial ileal resection in 2 occasions due to intussusception from multiple lipomatosis. A 45-year-old Japanese man was referred to our hospital for detailed examination after positive fecal blood test results and intermittent abdominal pain. He was diagnosed with intussusception of the ileum due to multiple lipomatosis and underwent partial ileal resection. Three years after the first surgery, he again experienced intermittent abdominal pain and nausea, and was referred to our department. Contrast-enhanced computed tomography at this time also showed intussusception near the ileocecal valve, with several fat-density tumors. He underwent partial ileal resection as an emergency surgery, with the histologic diagnosis confirming ileal multiple lipomatosis. Repeated **surgical resections are sometimes required for patients with intestinal intussusception due to lipomatosis, since lipomas not causing symptoms are often left untouched after minimal resection. Close follow-up such as contrast-enhanced CT and small-bowel endoscopy after surgery should be performed regularly so that the patients can avoid emergency surgeries because of the insufficient preoperative examinations.**

Key words: Multiple lipomatosis – Recurrent intussusception – Resection

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Adult intestinal intussusception is a rare disease known to be associated with intestinal tumors.[1–3] Lipoma, one of the most common causes of intestinal intussusception, usually occurs singly and few reports have described multiple lipomatosis.[4] We describe here a case requiring partial ileal resection on 2 occasions due to intussusception from multiple lipomatosis.

Case Presentation

A 45-year-old Japanese man was referred to our hospital for detailed examination after presenting with intermittent abdominal pain and showing positive results from a fecal blood test. Endoscopic examinations revealed multiple soft, yellowish masses in the ileum and contrast-enhanced computed tomography (CT) also showed multiple fat-density nodules inside the ileum with intussusception and pancreatic fat infiltration, suggesting a diagnosis of multiple lipomatosis (Fig. 1a–c). The patient underwent segmental ileal resection for histologic diagnosis and release of the intussusception. Tumor masses were distributed from 150 cm on the oral side of the ileocecal valve, and the intussusception was located 20 cm on the oral side of the ileocecal valve (Fig. 2a and 2b). Since the main purpose of surgery was to confirm the diagnosis of lipomatosis and release the intussusception, and also to avoid short-gut syndrome due to massive enterectomy, we decided to resect only the site of the intussusception as a 20-cm segment of ileum (Fig. 2c). The pathologic diagnosis was multiple lipomatosis of the ileum with no sign of malignancy. Postoperative course was uneventful. The patient was lost to regular follow-up without notice after discharge, although we were planning to perform contrast-enhanced CT regularly to examine the changes in size and numbers of the lipomas, because additional resection might be necessary to avoid possible emergencies. Three years postoperatively, the patient again experienced intermittent abdominal pain and nausea and was referred to our department. Physical examination revealed a movable mass in the right lower abdomen and contrast-enhanced CT revealed ileocecal intussusception with a leading fat-density tumor and several masses of the same density also evident inside the intestine (Fig. 3a). White blood cell count was also increased to 11,700/μL on blood testing. We therefore suspected that the intussusception was due to residual lipomas from the first surgery and decided to perform emergency surgery. Although the location of the leading lipoma was detected with the CT scan, we could not perform small-bowel endoscopy preoperatively. Since it seemed difficult to determine the extent of resection only with laparoscopic inspection without palpation, we decided to perform open surgery rather than laparoscopic surgery this time. An intussusception 3 cm in length was found 10 cm on the oral side of the previous anastomosis (Fig. 3b). Most of the lipomas were located on the oral side of the previous anastomosis and a 30-mm leading tumor for the intussusception was found 6 cm on the oral side of the ileocecal valve (Fig. 3c). The site of the previous anastomosis was slightly congested due to the intussusception and was resected with a 30-cm length of ileum, from 10 cm to the anal side to 20 cm to the oral side from the previous anastomosis, where the surgically palpable tumors were distributed (Fig. 4a). The histologic diagnosis was again submucosal lipomatosis without malignancy (Fig. 4b). The patient was discharged without any postoperative morbidity and has remained asymptomatic on regular postoperative follow-up for 6 months.

Discussion

Adult intussusception is rare, accounting for only 5% of all cases.[1,2] More than 80% of cases are caused by organic disorders, with intestinal tumors accounting for 59%.[3] About 60% to 70% of the intestinal tumors involved are benign, and lipoma ranks as the second most common cause of intestinal intussusception, accounting for 17%.[5] Around 70% to 80% of lipomas are reportedly found in the ileum, especially within 60 cm of the terminal ileum,[1,5,6] and the mean size of the lipoma causing intussusception is reportedly around 35 mm.[2] Secondary symptoms such as abdominal pain from diverticula and bleeding from ulceration of the overlying mucosa at the top of the tumor have been reported, usually in association with lipoma over 20 mm in diameter.[4,7] Only 5% of reported cases involve multiple intestinal lipomas, and these are sometimes accompanied by malnutrition due to the overgrowth of bacteria in diverticula or exocrine pancreatic insufficiency from fat infiltration of the pancreas.[4]

Few reports have described intussusceptions due to multiple lipomatosis as in this case, because of the infrequency of the disease. In our case, the leading lipoma of the intussusception was 30 mm in diameter, and hemorrhage from the lipoma due to

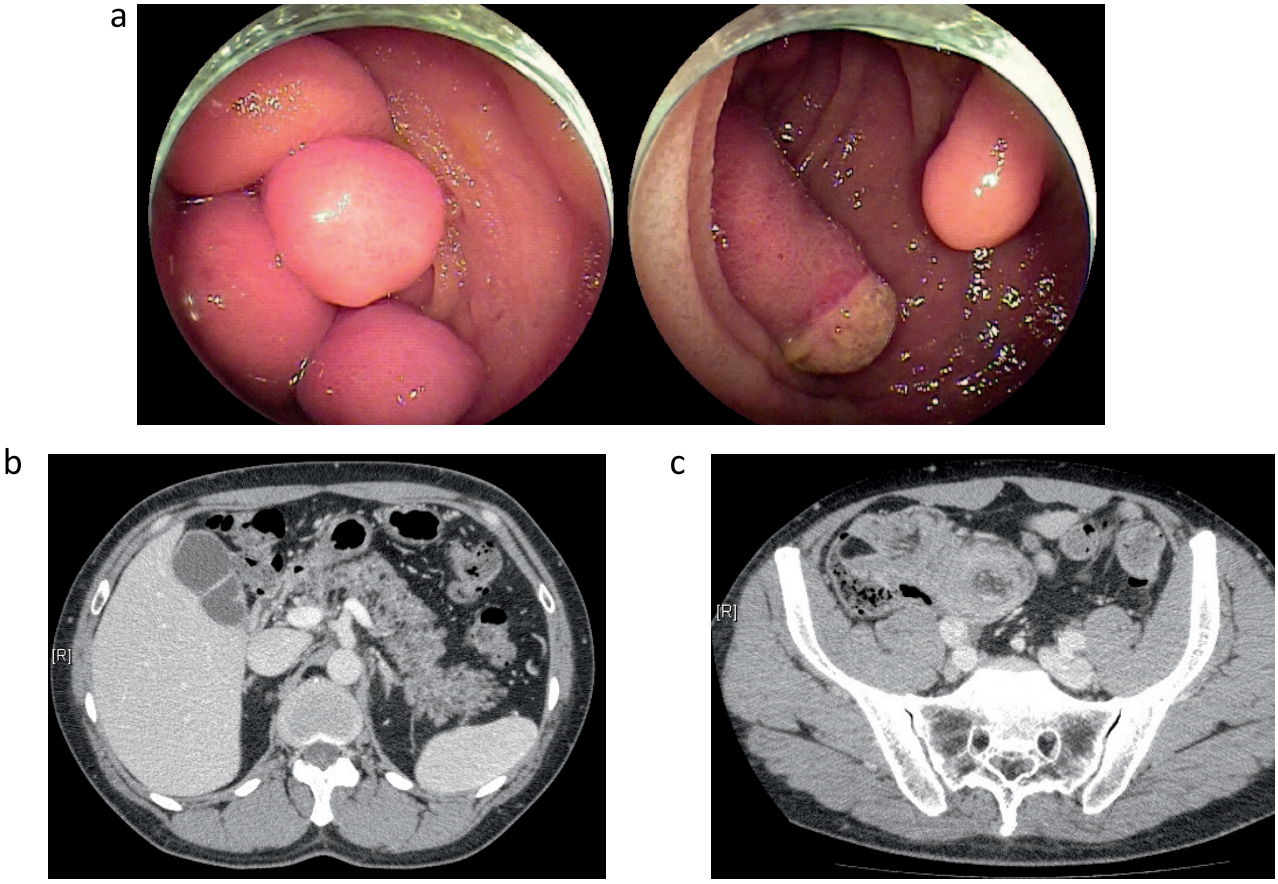

Fig. 1 Clinical findings from endoscopy and CT before the first surgery. (a) Enteroscopy before the first surgery shows multiple soft, yellowish masses with mucosal erosion. (b) Pancreatic fat infiltration is evident. (c) Concentric sign suggesting ileocecal intussusception due to leading lipoma.

mucosal ulceration was first detected as a positive result on a fecal occult blood test. The patient also suffered from repeated abdominal pain and constipation, but had not experienced weight loss or malnutrition, and was discharged without any surgical complications after each of the surgeries.

Intestinal intussusception due to lipoma is often subacute or chronic, and therefore is difficult to diagnose based on the clinical presentation alone. Small-bowel endoscopy is useful for detecting small intestinal tumors, and lipoma is detected as a soft, yellowish mass arising from the submucosa and not invading into the muscle layer. Lipomas look like a cushion when pressed with endoscopic forceps (cushion sign).[8] The sites and distributions of lipomas can also be diagnosed precisely. CT is another useful tool for detecting lipomas and consequent intussusception before intervention, because lipomas appear as fat-density masses (–40 to –120 Hounsfield units) inside the intestine and the site of intussusception is seen as a multiple concentric ring.[9] This finding was seen in our case. Ultrasonography and X-ray fluoroscopic examination are sometimes helpful in detecting tumors.

Surgical resection of the involved segment is commonly performed for intussusception due to lipoma, but no guidelines have been set to determine the resection range for multiple lipomatosis. Although the usefulness of laparoscopic surgery has recently been reported for intussusception,[10] detailed preoperative examination such as small bowel endoscopy may be necessary to determine the extent of bowel resection for patients with multiple lipomatosis. Since resection of the entire involved intestine may lead to short-gut syndrome, and lipomas themselves are benign, minimum resection around the intussusception site seems reasonable. However, since lipomas not causing symptoms are left untouched after minimum resection, close postoperative

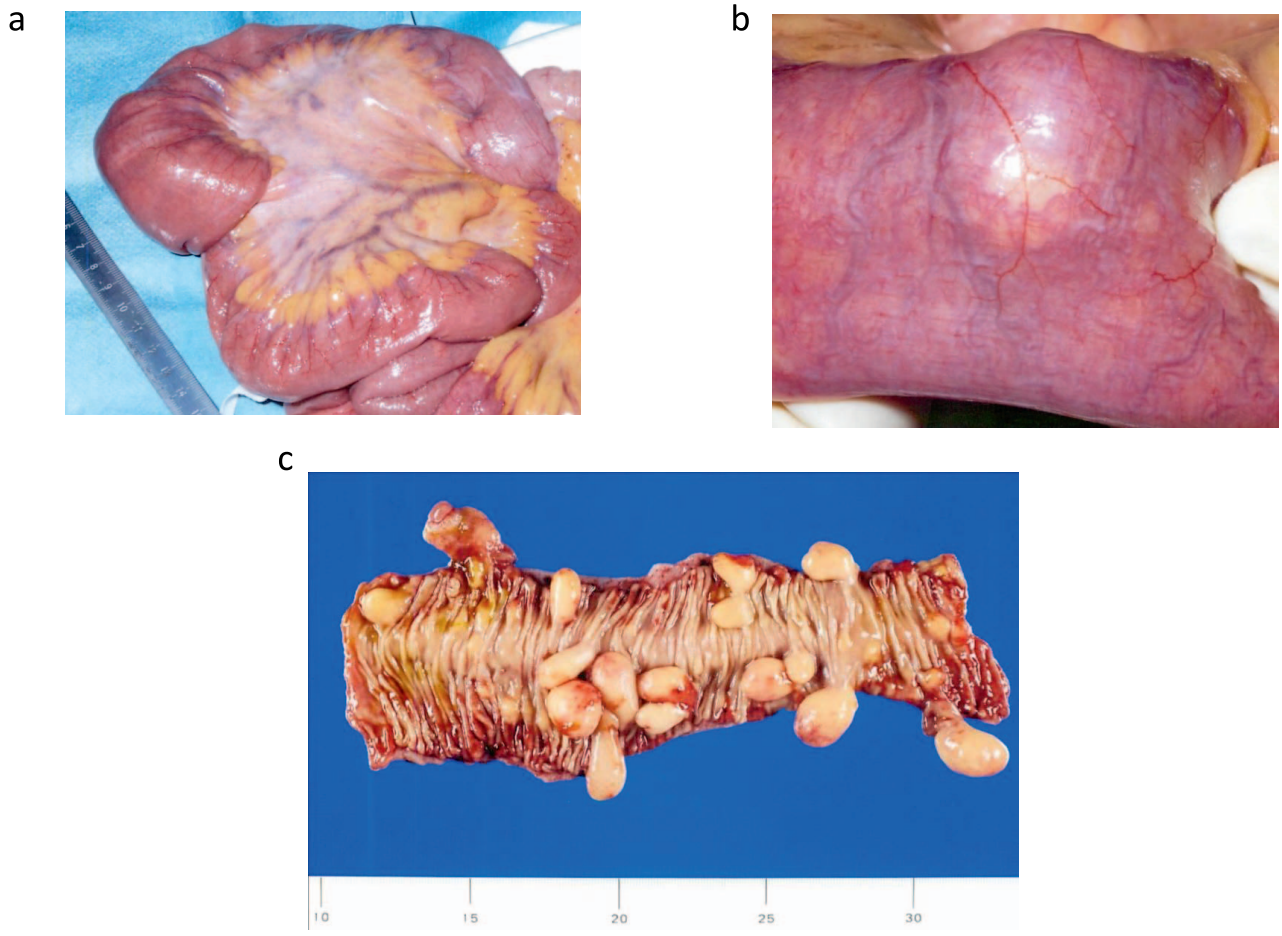

Fig. 2 Surgical findings from the first surgery. (a) Intussusception site 20 cm on the oral side from the ileocecal valve in the first surgery. (b) Soft, yellowish masses can be seen through the intestinal wall. (c) Resected ileum (20 cm in length) including the intussusception site. Pathologic diagnosis was multiple lipomatosis.

follow-up should be performed regularly. A case of additional endoscopic resection for lipomas using double-balloon endoscopy has also been reported,[11] and this kind of intervention represents another option when the lipomas are small enough to safely resect. To date, few reports have described cases requiring repeated surgical resections for intussusception due to lipomatosis. However, since any follow-up examinations were not done after the first surgery due to the patient's inconvenience, we had to perform a second emergency surgery in this case. However, this may have been avoidable if regular image examinations such as small-bowel endoscopy and contrast-enhanced CT had been performed after the first surgery. Detailed imaging examinations are strongly recommended after minimal surgical resection for multiple lipomatosis.

Conclusion

We have reported a rare case requiring repeated surgical resection for intestinal intussusception due to lipomatosis. Patients who undergo minimal bowel resection due to multiple tumors such as lipomatosis should receive regular follow-up after surgery using imaging to detect new lesions as early as possible and allow interventions such as endoscopic resection before resorting to second surgery.

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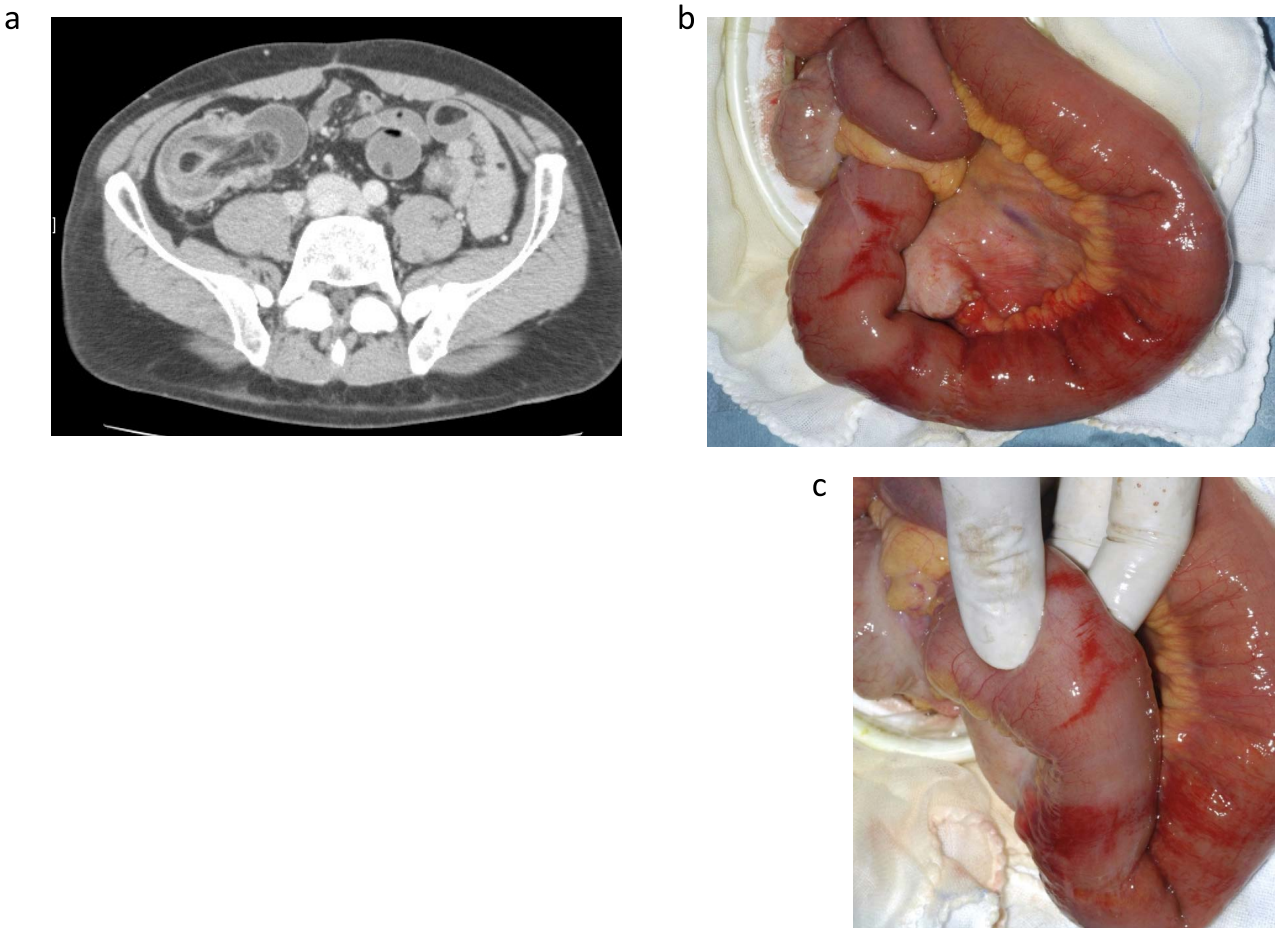

Fig. 3 Preoperative CT and surgical findings from the second surgery. (a) Multiple fat-density masses and ileocecal intussusception with leading lipoma are seen on preoperative CT. (b) Previous anastomotic site appears congested and indicates that intussusception occurred near this area. (c) Leading lipoma palpable 6 cm from the ileocecal valve.

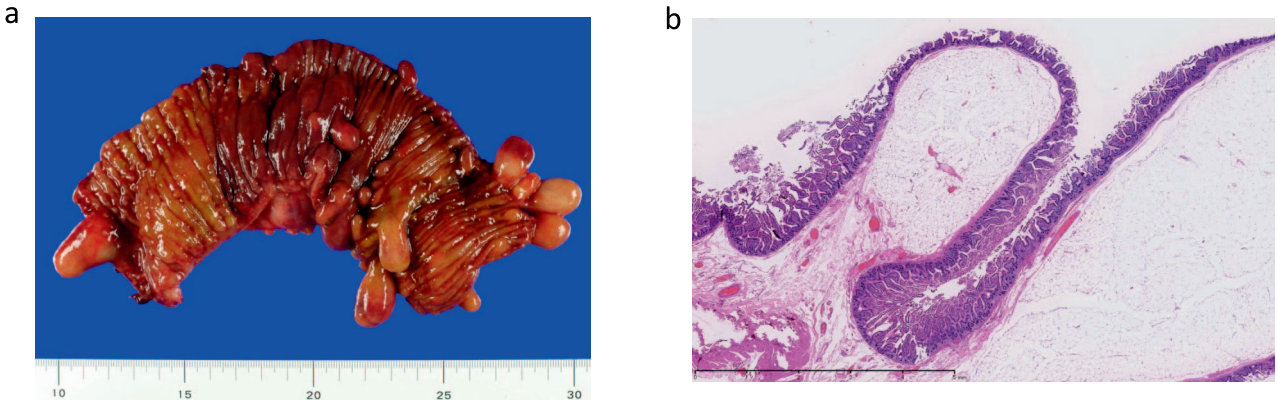

Fig. 4 Specimen taken from the second surgery and histologic findings. Surgical and pathologic findings. (a) Resected ileum (30 cm in length) with the congested mucosa at the previous anastomotic site. (b) Mature fat cells are seen in both specimens beneath the muscularis mucosae, and no malignant findings are evident. There is also no sign of diverticula.

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