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A case report of recurrent intussusception caused by small bowel lymphangioma in an adult

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ABSTRACT

INTRODUCTION: Adult intussusception and lymphangioma in gastrointestinal tract are uncommon entities respectively. Recurrent intussusception due to lymphangioma of the small intestine is extremely rare and mimics adhesive small bowel obstruction (SBO).
PRESENTATION OF CASE: A 37 year old man presented with acute abdominal pain and vomiting. He had been admitted several times for adhesive SBO after laparoscopic cholecystectomy at age 21. He was initially managed with a long tube placement, with which he used to get well. This time, the symptoms once relieved but soon relapsed, so an exploratory laparotomy was performed. Intraabdominal adhesiolysis was performed alongside the excision of a small segment of damaged jejunum. Intussusception of jejunum was noted and its reduction was also performed. Unfortunately, the symptoms continued after the operation, and computed tomography revealed a recurred intussusception of the jejunum. A reoperation with an additional resection of small intestine surrounding intussusception was performed. The symptoms subsided after the second operation and the patient was discharged. Pathological examination revealed lymphangioma within the affected lumen.
DISCUSSION: Intussusception in an adult is often caused by a tumor but can be caused by postoperative adhesion. The reduction is a potential option of treatment if there is no tumor suspected, but sometimes it would be uneasy to affirm the non-existence of tumors.
CONCLUSION: We present this rare case of recurrent jejuno-jejunal intussusception caused by small bowel lymphangioma with review of literature. Taking the possibility of recurrence and malignancy into account, the resection should always be considered in such patients.

1. Introduction

Intussusception occurs when the proximal portion of intestine invaginate into the adjacent distal portion. Adult intussusception is rare, with 90% of the cases caused by a gastrointestinal pathology, including adhesion or tumors [1,2]. Lymphangiomas are benign tumors resulting from congenital lymphatic system malformation [3], generally located in the head and neck regions or the axilla. The gastrointestinal lymphangioma accounts for only 1% of all [4,5]. Small bowel lymphangioma causing intussusception is rare, there have only been a few reports published [1,3,4,6].

We report a young man with intussusception caused by a small bowel lymphangioma, who presented with prolonged symptoms of small bowel obstruction (SBO) apparently associated with postoperative adhesions. This case report has been reported in line with the SCARE criteria [7].

2. Case presentation

A 37 year old male presented to our hospital with acute abdominal pain and vomiting. His abdomen was distended, but no apparent abdominal mass nor rebound tenderness were noted. He had past history of open appendectomy for appendicitis complicated with peritonitis when he was 12 years old, and he underwent laparoscopic cholecystectomy when he was 21. Since then, he had suffered from multiple bouts of SBO and frequently admitted. He also underwent laparoscopic adhesiolysis at the age of 33. On admission, his laboratory data showed hemoconcentration which suggested mild dehydration, and slight signs of inflammation. Contrast-enhanced abdominal computed tomography (CT) scan showed several points of caliber change suggesting the presence of adhesions, with no signs of ischemic bowel nor structural abnormality.

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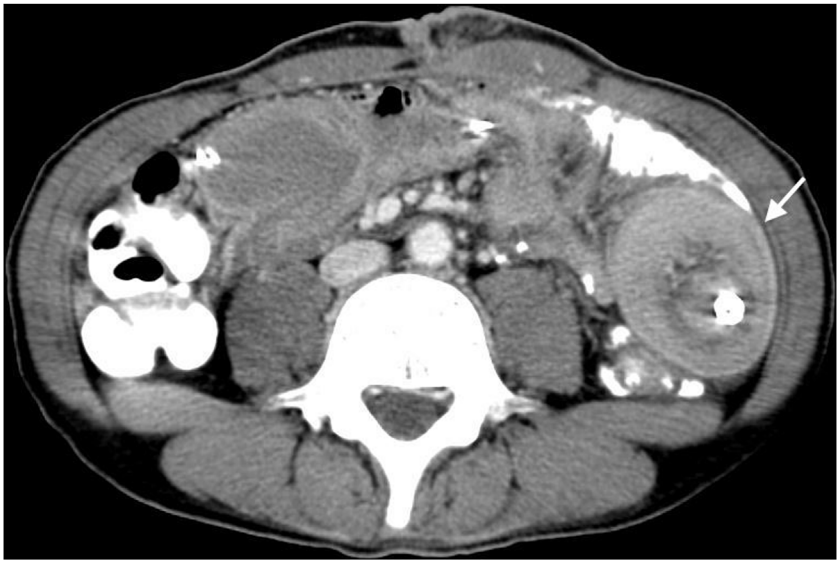

Fig. 1. Computed tomography: The presence of mesenteric fat within a bowel suggests intussusception (arrow). The long tube is also inside the small bowel.

A diagnosis of adhesive SBO was made and he was initially managed with a long tube placement. After several days, the symptoms were relieved and the tube was removed, but abdominal distension and pain soon relapsed in three days. After re-insertion of a long tube, there was no clinical improvement this time. We decided to perform an exploratory laparotomy, upon which we recognized the adhesion throughout the intestinal tract. We dissected the adhesion, and some area of the small intestine was found damaged during the procedure, and a short portion of the jejunum was resected. During the procedure, we found an unexpected jejunojejunal intussusception. The intussuscepted portion did not appear to be diseased, therefore we performed a manual reduction of the intussusception. The long tube was left inside the small intestine for the decompression of the enlarged small intestine.

Unfortunately, the symptoms continued for days after surgery, and repeated CT scan on postoperative day 7 revealed a recurred intussusception of the small intestine (Fig. 1). The contrast fluoroscopy performed while moving the long tube confirmed the position of the intussusception at the same site where we performed the primary reduction (Fig. 2). Non-operative management was not successful, so a reoperation was performed and the segment of the small intestine involved in the intussusception was resected (Figs. 3 and 4). The symptoms resolved after the reoperation and subsequently the patient was discharged without major complications. The gross pathological examination of the opened specimen of the resected bowel showed several cystic lesions inside the small intestine, that were judged to be the lead point of the intussusception (Fig. 5). The histological examination confirmed the diagnosis of lymphangioma, with the positive immunohistochemical staining of D2-40. The patient has not been admitted for the recurrence of the symptoms for several years thereafter.

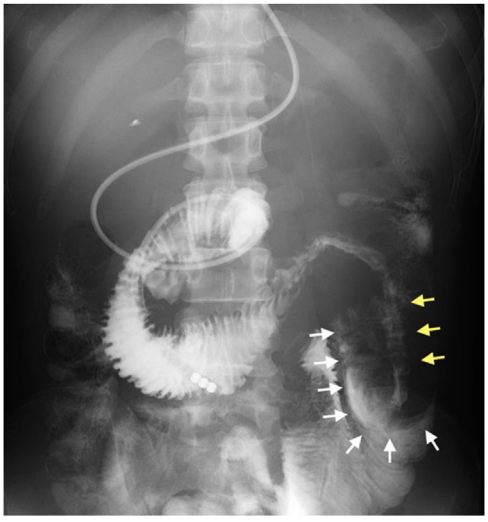

Fig. 2. Contrast fluoroscopy of the jejunum: Note the proximal lumen (yellow arrows) and the contour of the distal lumen (white arrows) of the intussuscepted jejunum.

3. Discussion

Intussusception in an adult is rare, and mostly caused by a structural abnormality such as tumors [1,8,9], which provide a lead point lesion for the invagination to occur. It can be classified by its location: enteric (confined to the small bowel), ileocecal (ileocecal valve leads the intussusception), ileocolic (terminal ileum prolapses into the ascending colon), and colocolic (confined to the large bowel) [8].

In previous reports, 38%–66% of intussusception occurs in small bowel (enteric intussusception). In enteric intussusception, tumors were found in up to 75% of the patients, and incidence of malignancy is as high as 29%–38% [8,9]. Other etiology of enteric intussusception includes Meckel's diverticulum, inflammatory lesions and postoperative adhesions [2,8,9]. In addition, an enteric tube placement can cause iatrogenic intussusception [10].

The classical triad of intussusception (currant-jelly stools, abdominal pain and palpable mass) is rarely seen in adults. Instead, they present with nonspecific abdominal pain, nausea and vomiting [2,8], which resemble the symptoms of adhesive SBO. In our case, the symptoms on the admission were those caused by adhesive SBO, but retrospectively, the similar symptoms after the first surgery are supposed to have been those of intussusception, judging from the resolved symptoms after the reoperation. CT findings are pathognomonic. The mesenteric fat within the the mass of intussusception makes it look like a "target" when the image is perpendicular to its longitudinal axis as was in our patient [11].

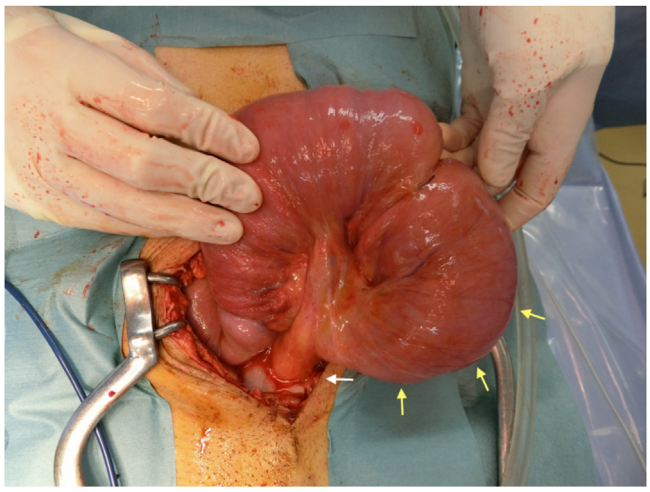

Fig. 3. Intraoperative findings: Intussusception (white arrow) and the dilation of the proximal jejunum (yellow arrows).

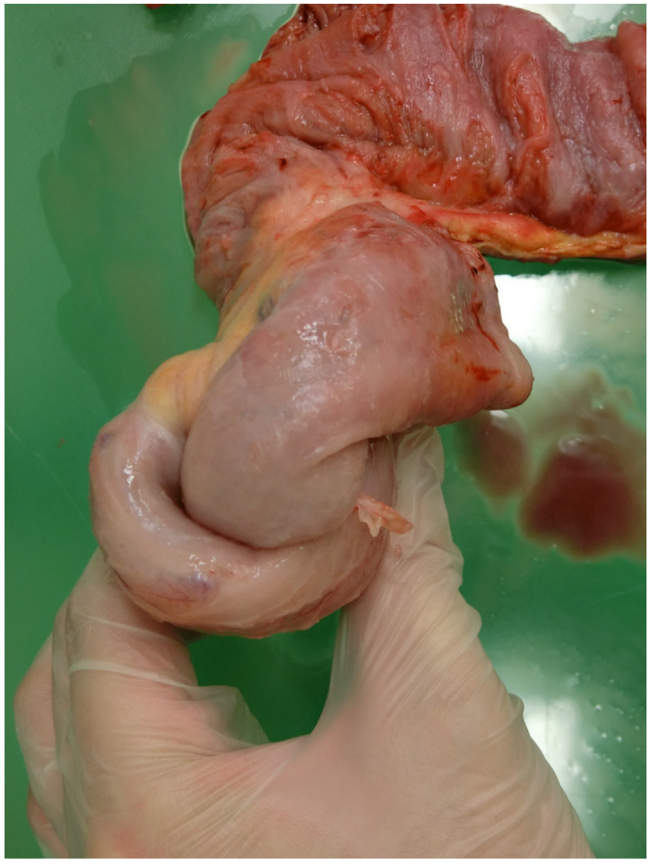

Fig. 4. Excised specimen: Intussuscepted portion of the small intestine.

Surgical resection is usually recommended in adult intussusception [2,9,12]. However, there are several reviews suggesting that in cases of small bowel intussusception, reduction can be attempted when the segment involved is viable and a malignancy is not suspected [2], or when it is associated with serosal adhesions without demonstrable intraluminal disease [8].

There have been several case reports on the patients with small intestinal lymphangioma causing intussusception in adults [1,3,4,6]. In all these reports, the patients presented with abdominal pain with or without nausea. Those case reports represent patients without past surgery on their abdomen, so the symptoms were not suspected to be those caused by adhesive SBO.

In our case, however, the intussusception occurred in a patient with history of several episodes of postoperative adhesive SBO. There was no sign of tumor causing intussusception recognizable preoperatively or intraoperatively. During the first operation, we considered that the symptoms should have been due to adhesions, and that the intussusception itself was the result of adhesions, so that the adhesiolysis would be sufficient in treating the etiology. That being said, considering the substantial incidence of tumors in adult patients with intussusception, whether it is benign or malig-

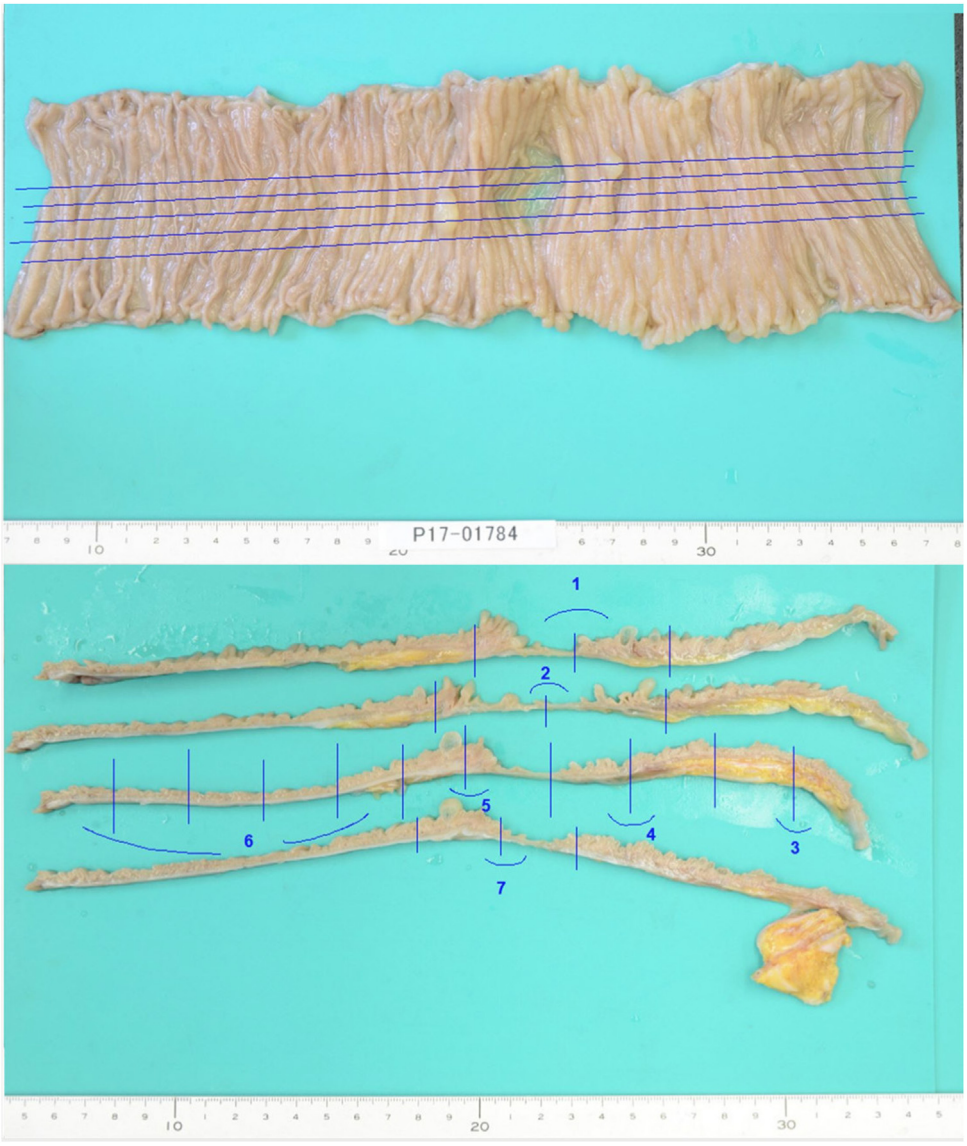

Fig. 5. Gross pathological findings: Multiple cystic lesions are noted inside the small bowel. Lymphangioma was noted in the areas 1, 2, 5 and 7.

nant, we now believe that it would have been a better option to perform resection at the first surgery.

4. Conclusion

We report a rare adult case with jejuno-jejunal intussusception resulting from a small bowel lymphangioma, that recurred soon after primary reduction. Surgical resection is warranted even if there is no clinical sign of structural abnormality, because the causative tumors are not easily detectable before pathological examination.

Conflicts of interest

No conflicts of interest.

Sources of funding

No source of funding.

Ethical approval

Ethical approval is not required by our institution. Permission and consent has been taken from the patient.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Author contribution

Toru Akashige – Abstract, Figure collections, writing.
Kota Sato – writing, editing.
Hajime Odajima – review, others.
Shigeru Yamazaki – review, editing.

Registration of research studies

This case report is not “first-in-man”, so registration to www.reserchregistry.com cannot be done.

Guarantor

Toru Akashige.

Provenance and peer review

Not commissioned, externally peer-reviewed.

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