Repeated Intussusception Induced by Intestinal Lipomatosis: Report of a Case

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We describe a man with intestinal lipomatosis in association with repeated episodes of intussusception. A 50-year-old man came to our emergency department for intermittent epigastric cramping pain. Abdominal computed tomography scan revealed a doughnut sign and suspected ileo-colic intussusception. Emergent laparotomy showed ileo-colic intussusception involving the ileocecal valve, cecum and approximately 30 cm of the ascending colon. Multiple submucosal tumors were noted to involve the whole small intestine and one tumor of the ileum formed the leading point of the intussusception. Right hemicolectomy with primary anastomosis was performed. Pathologic examination confirmed that the multiple lesions were benign submucosal lipomas. One month later, another episode of intussusception was noted. The affected intestinal segment was resected with ileo-ileostomy. In patients with intussusception caused by submucosal lipomatosis, surgery might be curative. If a large segment of bowel was affected by submucosal lipomatosis, resection of all lipomas might not be feasible. Smaller lipomas may be left in place because these tend to be asymptomatic. But they may become leading points of recurrent intussusception.

Key words: submucosal lipomatosis, intussusception

Small intestinal neoplasms are uncommon in clinical practice. Benign small intestinal tumors (e.g., leiomyoma, lipoma, hamartoma, or desmoid tumor) are usually asymptomatic but may present with intussusception.[1] Because the small intestine is relatively inaccessible to routine endoscopy, diagnosis of small intestinal neoplasms is often delayed after onset of symptoms.[2] Intussusception is common in the pediatric population but quite uncommon in adults.[3] The treatment of the adult intussusception is always operative. We report here by a case of intestinal lipomatosis in association with repeated episodes of intussusception, and discuss the clinical course, image findings, and treatment.

Case Report

A 50-year-old man without a past history of surgery had episodic abdominal discomfort for more than 5 years. He came to the emergency department several times for help but no specific lesion was identified. The pain became more and more frequent, and was unresponsive to antacids in the previous 2~3 months. KUB revealed distended bowel loops, and hyperactive bowel sounds were noted. Then he was admitted to the gastrointestinal department under the impression of intestinal obstruction. During the admission, the abdominal distension became more severe, and the intermittent epigastric cramping pain still worse. On physical examination, he was found to have rebound tenderness, and a palpable ill-defined, sausage-like mass in the right upper abdomen. Computed tomography (CT) scan revealed a target/doughnut sign and suspected ileo-cecal intussusception (Fig 1).

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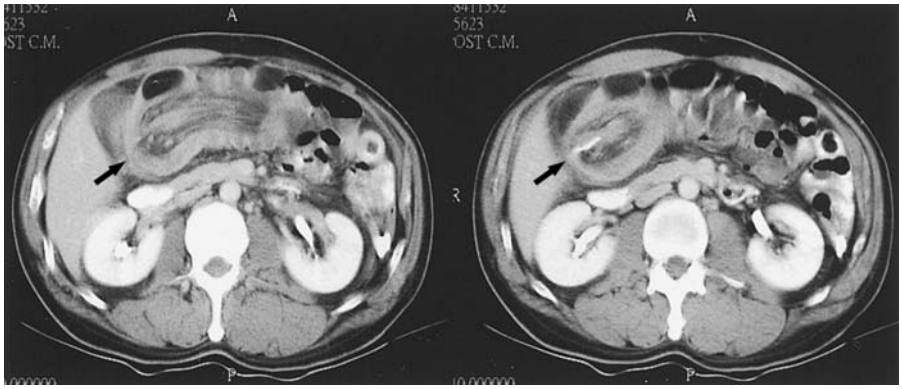

Fig 1. Abdominal CT revealing target/doughnut sign involved cecum and ascending colon (arrow).

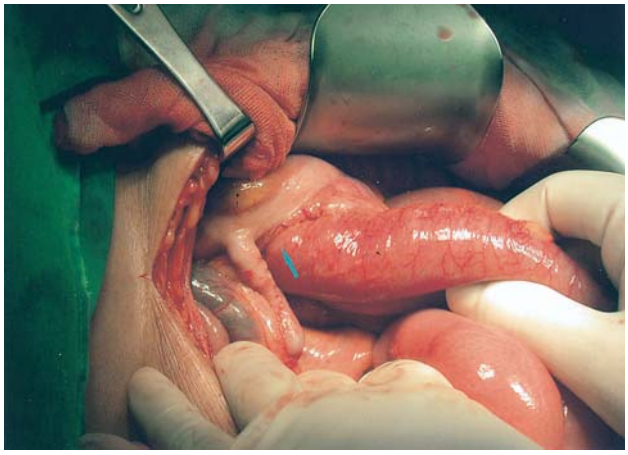

Fig 2. Laparotomy showing ileo-colic intussusception involving approximately 30 cm of ascending colon (arrow).

Fig 3. Multiple submucosal tumors on surface of small and large intestine.

Laparotomy showed ileo-cecal intussusception involving 30 cm of the ascending colon (Fig 2). Multiple submucosal tumors were noted over the whole small intestine (Fig 3); one tumor of the ileum formed the leading point of intussusception. Reduction was not performed due to tight telescopical impaction of the intussusceptive segment. Right hemicolectomy with primary anastomosis was performed. Pathologic examination confirmed that the multiple lesions were benign submucosal lipomas (Fig 4). Some larger lipomas were resected. The other lipomas were left in place because they were small, numerous and total resection was deemed impracticable. One month later, another episode of bowel obstruction was noted. After conservative treatment with total parenteral nutrition (TPN) for one week, distension and peritoneal signs progressed. Abdominal CT revealed ileo-ileal intussusception (Fig 5). The patient received a second laparotomy, and the affected intestine (Fig 6) was resected with ileo-ileostomy. Severe adhesion and fixed small and large intestine were noted during the second laparotomy, adhesionlysis was not performed in consideration of fixed bowel loops which may decrease risk of further intussusception. After the second operation, the patient has been free of symptoms and signs of bowel obstruction for the past two years.

Discussion

Most common benign tumors of the small intestine are leiomyoma, adenoma and lipoma. Lipomas can be found anywhere but are more commonly located at the distal intestine.[1] More than two thirds of the lipomas

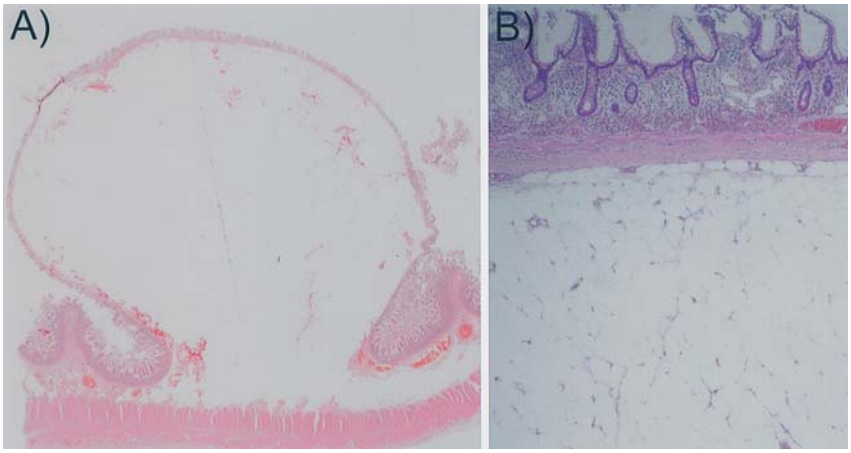

Fig 4. Microscopic examination showing multiple benign submucosal lipomas. (Hematoxylin and eosin stain; A) ×40, B) ×100).

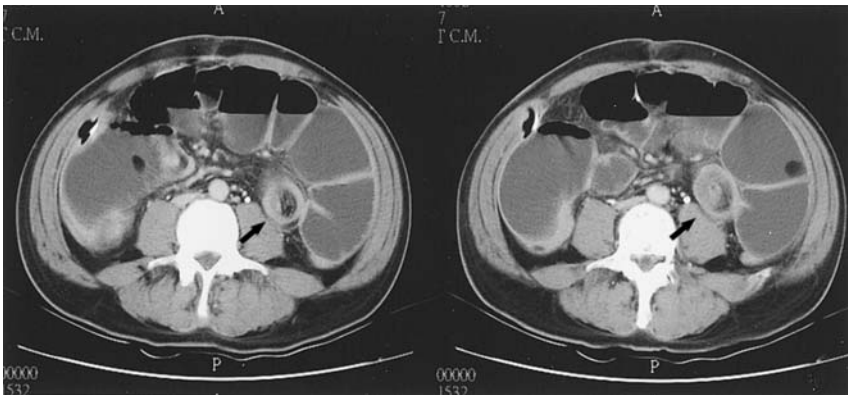

Fig 5. Abdominal CT revealing severe ileus and second episode of intussusception (arrow).

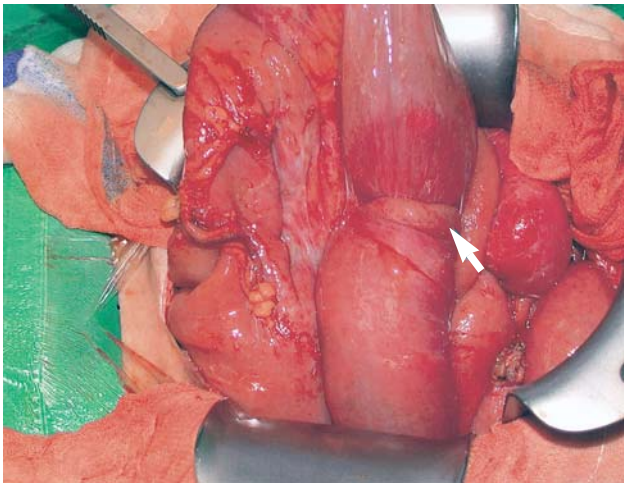

Fig 6. Laparotomy showing ileo-ileal intussusception (arrow).

remain asymptomatic and are found incidentally either at surgery or at autopsy. For those tumors that become symptomatic, intermittent intestinal obstruction and gastrointestinal (GI) bleeding, often attributable to intussusception, are the cardinal manifestations.[2] One of the lipomas was a leading point of ileo-cecal intussusception in our patient and another lipoma induced a second episode of ileo-ileal intussusception. Intussusception is common in the pediatric population but quite uncommon in adults.[3] The clinical findings of adult intussusception are variable. Acute intestinal obstruction is not common. Most of the adult patients presented with subacute, chronic, or intermittent intestinal obstruction. Intussusception can occur anywhere in the gastrointestinal tract. Ultrasonography in the emergency setting has a role in the evaluation of nonspecific abdominal pain. Emergent ultrasonography can be useful to prevent a misdiagnosis

in an atypical clinical course of intussusception, acute cholecystitis, or aortic aneurysm.[3] The most useful diagnostic radiologic method of adult intussusception is the CT scan.[4] A CT scan demonstrates the collapsed intussuscepted proximal intestine (intussusceptum) as well as the mesenteric fat and vessels lying within the wall of the distal intestine (intussuscipiens). On cross-sectional images, the intussusception demonstrates a target appearance.[5] It may appear as a sausage-shaped or reniform mass as the disease progresses. The treatment of adult intussusception is always operative. As almost 50% of adult intussusceptions harbor malignant lesions,[6] resection without reduction is advocated as the best treatment of adult intussusception. In adult intussusception, the leading point of any diagnostic possibility should be always kept in mind. Solitary gastrointestinal lipomas are infrequently encountered in clinical practice. Small bowel lipomatosis, or multiple lipomas limited to the small bowel, is exceedingly rare. Fewer than 25 cases have been reported.[7] No pattern of Mendelian inheritance has been described for the disorder.[7] Ileo-ileal intussusception is rare. In patients with repeated intussusception caused by intestinal lipomatosis, surgery is curative for each respective episode. Cases of small bowel intussusception can be reduced in patients unless strangulation of the bowel is present. Therefore, unnecessary resection that may result in short bowel syndrome can be avoided.[8] In our case, as a large segment of bowel was affected by lipomatosis, resection of all lipomas was considered impossible. Large lipomas may be safely removed through an enterotomy. Smaller lipomas may be left in place because these tend to be asymptomatic,[9] but recurrent episode of intussusception may be encountered.

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多發性小腸脂肪瘤導致反覆性腸套疊：病例報告

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在我們的病例報告中，患者為50歲男性，腹部斷層掃瞄顯示疑為腸套疊。剖腹探查確立為多發性小腸脂肪瘤併發迴腸—結腸套疊。患者在接受右半側大腸切除術後，恢復良好。但術後一個月，再度出現腸阻塞症狀，腹部斷層掃瞄顯示為迴腸—迴腸套疊，剖腹探查確立為多發性小腸脂肪瘤導致之反覆性腸套疊，在切除套疊部位及迴腸—迴腸吻合術後，患者之腸阻塞解除，並已兩年無症狀。

多發性小腸脂肪瘤，為良性腫瘤，一般多無症狀，當有明顯的阻塞性症狀或造成腸套疊時，則需手術治療。在腸套疊的診斷上，腹部斷層掃瞄最具診斷價值。多發性小腸脂肪瘤導致之反覆性腸套疊，由於無法將多發性小腸脂肪瘤完全根除，仍有再發腸套疊之風險。

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