

## A Case of Spontaneously Reduced Ileoileal Intussusception Caused by a Lipoma

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### ABSTRACT

Intussusception in adulthood is unusual. We describe herein a rare case of adult ileoileal intussusception caused by an ileal lipoma, which was diagnosed preoperatively and was confirmed at the operation to have reduced spontaneously. A 68-year-old woman experienced sudden-onset colicky pain in the upper abdomen accompanied by vomiting and was brought to our hospital by ambulance. Physical examination revealed a distended abdomen and tenderness in the upper abdomen. Laboratory findings showed slightly elevated inflammatory parameters. Abdominal computed tomography (CT) showed a target sign in the ileum, which is a typical sign of intussusception. Additional caudal-side scans showed a homogenous and fatty mass measuring 2.5 cm that was considered to be the leading point for the invagination. These findings led to a preoperative diagnosis of intussusception induced by a lipoma. The patient underwent emergency surgery. Laparotomy revealed a yellowish, soft ileal tumor measuring 2.5 cm in diameter and that the intussusception had already been reduced at laparotomy. Approximately 15 cm of the ileum's length, including the tumor, was resected, and an end-to-end anastomosis was performed. Adult intussusception caused by an ileal lipoma is a rare condition. However, CT is the most useful tool for making a definite preoperative diagnosis based on its typical findings.

**Key words:** *Intussusception, Computed tomography, Lipoma*

Intussusception involves the invagination of one segment of bowel together with its mesenteric fold into the lumen of an adjacent segment of bowel. Intussusception in adulthood is unusual, with an incidence of approximately 2-3 cases per 1,000,000 people per year<sup>7)</sup>. The occurrences of intussusceptions in adults account for less than 5% of all cases of intussusceptions and almost 1% of bowel obstructions<sup>1,2,9)</sup>. There are major differences in the clinical presentation, diagnostic approach, and management of intussusception between the pediatric and adult populations<sup>8)</sup>.

Lipomas of the small intestine are rare benign tumors, and are most commonly encountered incidentally because they are usually asymptomatic. Symptomatic lipomas manifest with hemorrhage or intestinal obstruction<sup>18)</sup>, and intussusception also constitutes a rare clinical entity.

We describe herein a rare case of adult ileoileal intussusception caused by an ileal lipoma, which was diagnosed preoperatively and was confirmed to have reduced spontaneously at operation.

### CASE PRESENTATION

A 68-year-old woman experienced sudden-onset colicky pain in the upper abdomen with vomiting and was brought to our hospital by ambulance. Her medical history included hypertension and arrhythmia. She had undergone appendectomy for appendicitis at the age of 22 years and cholecystectomy for acute cholecystitis at the age of 45 years. She had begun chronic hemodialysis due to end-stage renal disease 8 years previously. Physical examination revealed a distended abdomen and tenderness in the upper abdomen. Laboratory findings showed slightly elevated inflammatory parameters. Her white blood cell count was 9,200/ $\mu$ L, and her C-reactive protein level was 0.3 mg/dL. Abdominal computed tomography (CT) showed a target sign in the ileum, which is a typical sign of an intussusception (Fig. 1a). The intussusception corresponded to the entero-enteric type. Further caudal-side scans showed a fatty (-100 Hounsfield Units) homogenous mass measuring 2.5

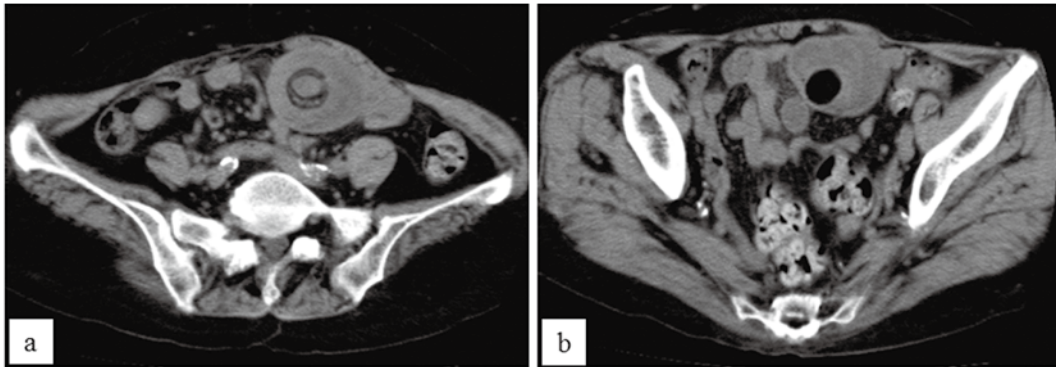
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cm that was considered to be the leading point for the invagination (Fig. 1b). These findings led to a preoperative diagnosis of intussusceptions induced by a lipoma.

The patient underwent emergency surgery. Laparotomy revealed a yellowish, soft ileal tumor measuring 2.5 cm in diameter and that the intussusception had already been reduced (Fig. 2). Approximately 15 cm of ileum, including the tumor, was resected, and

an end-to-end anastomosis was performed. The post-operative course was uneventful, and the patient was discharged on the thirteenth postoperative day.

Macroscopic assessment of the resected specimen showed the presence of a submucosal tumor that was  $2.5 \times 2.0 \times 1.5$  cm in size with the features of lipoma (Fig. 3a, b). Histological examination revealed mature fat cells, connective tissue, and scattered blood vessels within the removed submucosal tumor.



**Fig. 1.** Abdominal computed tomography: An axial image showing a typical target sign (a) and a fatty mass as the leading point of the intussusception (b).

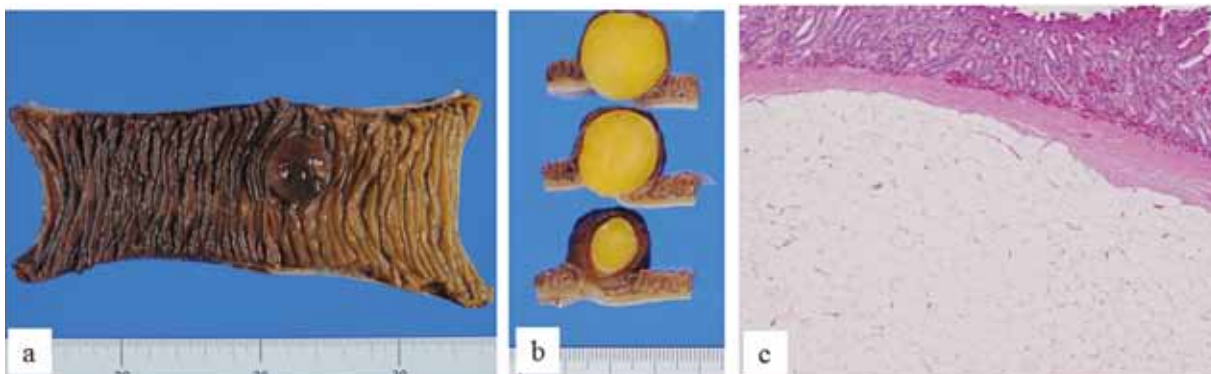


**Fig. 2.** Operative findings: Laparotomy revealed a yellowish, soft ileal tumor measuring 2.5 cm in diameter and that the intussusception had already been reduced.

There was no evidence of dysplasia or malignancy (Fig. 3c). The patient is alive without symptoms 5 years postoperatively.

## CONCLUSIONS

The exact mechanism that precipitates intussusception in adults is still unknown. However, it is generally suggested that any lesion in the bowel wall or irritant within the bowel lumen could alter the normal peristaltic pattern and thereby initiate an invagination leading to intussusception<sup>16</sup>. This pathologic process often results in obstruction. Intussusceptions have been classified according to the site of the lesion. The most common classification system divides intussusception into four categories: enteric, ileocolic, ileocecal, and colonic<sup>5,16</sup>. Two-thirds of intussusceptions in adults are of the en-



**Fig. 3.** Macroscopic and histologic findings of the excised specimen.

a,b: A submucosal tumor, measuring  $2.5 \times 2.0 \times 1.5$  cm.

c: Mature fat cells in the submucosa to the muscularis propria of the ileum (hematoxylin and eosin staining,  $\times 100$ ).

teric type<sup>15</sup>) that was seen in this case. The clinical presentation is complicated and non-specific in adults. Although abdominal pain is the most common presentation, it is accompanied by varying symptoms and usually has a subacute or chronic onset. Fewer than 20% of cases present acutely with complete bowel obstruction. The diagnostic radiologic method that is most useful in making the diagnosis of adult intussusception is CT<sup>11</sup>. It helps in both making an accurate diagnosis and also for identifying any associated underlying causes. Previous reports regarding adult intussusception showed that most patients had organic lesions as the leading point, and the majority of the lesions were neoplasms<sup>13,16</sup>. A recent report showed that benign (24%) and malignant (21%) neoplasms were the most common etiology for adult intussusception; 30% of cases with any organic leading point were idiopathic<sup>15</sup>. The sensitivity of CT to diagnose intussusceptions correctly has been reported to range from 58% to 90%, as verified by subsequent surgery<sup>3,4,11</sup>.

Gastrointestinal lipomas are most commonly located in the colon (65% to 75%), small bowel (20% to 25%), and occasionally in the foregut (< 5%)<sup>6</sup>. Common symptoms of intestinal lipoma include abdominal pain (68%), intussusception (44%), hemorrhage (29%), vomiting (24%), and obstructive symptoms (18%)<sup>17</sup>. Thus, intussusception is a common complication of gastrointestinal lipoma, although the disease is classified as rare. Lipomas account for 5.1% of the causes of adult intussusception<sup>15</sup>. Namikawa et al<sup>14</sup> reviewed 50 cases of adult intussusception caused by a lipoma and reported in the English literature during the past decade. The cohort comprised 28 men and 22 women with a mean age of 52.1 years. The lipoma size ranged from 1.2 to 12.0 cm, with a mean size of 4.9 cm. Among them, 50% of lipomas were located in the small intestine, and the most frequent location was the ileum (21 of 25 cases). A preoperative diagnosis of intussusception was made in 21 (42%) of 50 patients. Their case was reported as the sixth case of adult ileoileal intussusception caused by a lipoma diagnosed preoperatively<sup>14</sup>.

Surgical resection remains the recommended treatment for adult intussusception due to the large proportion of organic causes and the relatively high incidence of malignancy<sup>1,9,13</sup>. The advantages of intraoperative reduction of the intussusceptions prior to resection, especially when the small bowel is affected, are that it may preserve a considerable length of bowel and thereby prevent development of short-bowel syndrome, but most surgeons agree that reduction should not be attempted if there are signs of irreversible bowel ischemia, inflammation, or when malignancy is suspected. However, the resection of a long segment of intussusception often necessitates the excision of more of the intestine

than necessary to remove the tumor without reduction. Therefore, reduction should be attempted if the involved bowel segment is viable but only in patients for whom a definitive benign diagnosis has been made preoperatively<sup>9,13,14</sup>.

The rare case presented here could have a preoperative definitive diagnosis of ileoileal intussusception caused by an intestinal lipoma. Typical CT findings of intussusception and the existence of a lipoma as the leading point contributed to a definitive preoperative diagnosis. In addition to the rarity of the disease, the present case developed a spontaneous reduction that was confirmed at operation. There have been few reports of spontaneous reduction of adult intussusception of the appendix<sup>12</sup>. Another report involved a case of intussusception caused by blunt trauma to the abdomen<sup>10</sup>. These cases were confirmed to be spontaneously reduced on follow-up CT. Spontaneous reduction in adult intussusception can be a phenomenon occurring in the setting of idiopathic or incidental intussusception. However, to the best of our knowledge, there have been no reports of spontaneous reduction of adult intussusception caused by neoplasms.

In conclusion, adult intussusception caused by an intestinal lipoma is a rare condition. CT is the most useful tool for making a definite preoperative diagnosis based on its typical findings. Furthermore, the present case highlights the possibility of spontaneous reduction of intussusception caused by neoplasms.

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