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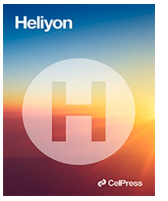

# Ileum intussusception secondary to submucosal liposarcoma in adult : A case report

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ABSTRACT

Intussusception in adults is a rare surgical emergency. Unlike in children, most adult intussusceptions arise from a pathological lead point. Ileal intussusception caused by a submucosal liposarcoma is a particularly rare phenomenon. This report describes the diagnosis and management of adult ileal intussusception secondary to submucosal liposarcoma in adult to provide a reference for future clinical work. A 64-year-old female presented to the emergency department with worsening abdominal pain associated with an 8 h history of intermittent vomiting. Based on physical examination, laboratory investigations, and computed tomography, the most likely diagnosis was ileal intussusception secondary to liposarcoma. Thus, emergency laparotomy was performed. During exploration, an ileal invagination was visualised approximately 30 cm from the ileocecal valve, and a flexible polypoid mass was palpable at the lead point of the intussusception. Subsequently, the patient underwent radical resection of pathological tissues with a primary end-to-end ileal anastomosis. Histopathological examination revealed a well-differentiated submucosal liposarcoma. Postoperatively, the patient recovered uneventfully and was doing well at the 6-month follow-up in the outpatient clinic. Thus, clinicians should consider the origin of submucosal liposarcomas in adult with intussusception. Once ileal intussusception secondary to submucosal liposarcoma is diagnosed, timely radical resection is recommended.

## 1. Introduction

Intussusception is the prolapse of a segment of the intestine and its mesentery into the lumen of the adjacent intestinal segment. It was first described by Paul Barbette in 1674. Intussusception is relatively common in children, although its presence in adults is rare, accounting for 5 % of all cases of intussusception. Unlike in children, 90 % of adult cases of intussusception are caused by a definitive underlying disorder, with only 10 % being idiopathic [1]. Neoplasms are the most common causes of intussusception and are found in approximately two-thirds of adult cases. Liposarcoma is a malignant tumour that originates from mesenchymal cells and tends to differentiate into adipose tissue, accounting for approximately 15–20 % of all soft tissue sarcomas [2]. Liposarcoma often occurs in the deep soft tissues of the lower limbs, mesenteric area and retroperitoneum with high fat content. However, liposarcoma originating in the ileum is extraordinarily rare [3]. To date, intussusception due to ileal liposarcoma has not been reported. This report describes the diagnosis and management of an extraordinarily rare case of adult ileal intussusception caused by submucosal liposarcoma to add to experience of clinicians.

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## 2. Case presentation

A 64-year-old female presented to the emergency department with worsening abdominal pain associated with an 8 h history of intermittent vomiting for 8-h. She had experienced similar episodes of pain for the past 3 months, which was relieved without treatment. The patient had no history of abdominal surgery or systemic disease. Physical examination revealed abdominal distention and mild tenderness around the right iliac fossa, with no muscle guarding or rebound tenderness.

Laboratory investigations revealed an elevated white blood cell count ($10.2 \times 10^9$/L), and serum C-reactive protein and D-dimer levels of 25.1 mg/L and 2.20 mg/L, respectively. Other laboratory results were within normal limits. Abdominal contrast-enhanced computed tomography (CT) revealed a classic bowel-in-bowel appearance of intussusception, which was characterised by the "target sign" (Fig. 1a). Furthermore, a fat attenuation mass (−90 Hounsfield units) accompanied by a strip with a slightly high-density shadow was identified as the lead point within the lumen of the intussusception (Fig. 1b).

Based on these findings, the most likely diagnosis was ileal intussusception secondary to intestinal liposarcoma. Thus, emergency laparotomy was performed. During exploration, an ileum invagination, measuring 8 cm in length, was visualised approximately 30 cm from the ileocecal valve, and a flexible polypoid mass was palpable at the lead point of the intussusception. Intraoperative exploration confirmed the diagnosis of ileal intussusception secondary to intestinal liposarcoma. Subsequently, the patient underwent radical resection of the pathological tissue with a primary end-to-end ileal anastomosis. During the laparotomy, no attempt was made to reset the intussusception, and the pathological tissue was completely removed. The surgical process was uneventful, with a surgical duration of 83 min and an intraoperative bleeding volume of 20 ml. Macroscopic examination of the specimen revealed a polypoid pedunculated lesion, measuring $58 \times 35 \times 32$ mm, arising from the ileum (Fig. 2). Histopathological examination confirmed a well-differentiated submucosal liposarcoma with negative margins (Fig. 3). Postoperatively, the patient recovered uneventfully and was discharged from the hospital six days after surgical resection. The patient was doing well at the 6-month follow-up in the outpatient clinic.

## 3. Discussion

Intussusception occurs when a portion of the proximal bowel is distally driven into the bowel via peristalsis. Most intussusceptions in adults are caused by a definitive underlying disorder. Adult intussusception is often secondary to pathological factors, such as tumours, polyps, inflammation, and/or diverticula, the most common being intestinal tumours. Liposarcoma is a malignant tumour that originates from mesenchymal cells and tends to differentiate into adipose tissue. Liposarcoma is a common soft tissue malignancy that accounts for approximately 20 % of all soft tissue malignancies. The World Health Organization classifies these tumours into four categories: well-differentiated, myxoid/round cell, pleomorphic, and dedifferentiated liposarcomas. Different liposarcoma subtypes exhibit varying oncological and histological characteristics. Well-differentiated liposarcomas are the most common subtype, and have a lower grade and metastatic potential [2]. According to the positional relationship, liposarcomas can be divided into three types: submucosal type, intramuscular type, and subserous type. Although the small intestine accounts for 75 % of the length and 90 % of the surface area of the gastrointestinal tract, primary submucosal liposarcomas are rare. Submucosal liposarcoma in adults is usually asymptomatic, although continuous growth of the tumour may cause intussusception. The main mechanism underlying intestinal

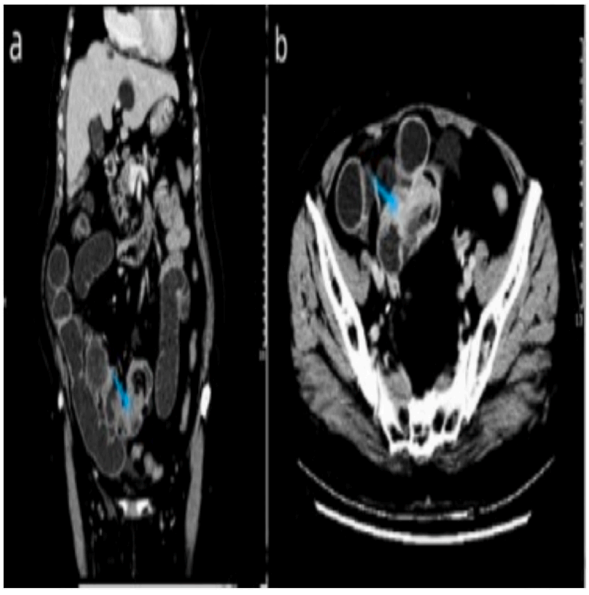


**Fig. 1. a**. Coronal CT scan showing invaginated mesenteric vessels and mesenteric fat (arrow). Fig. 1b. Axial CT scan showing an intraluminal enhancing lesion at the lead point of the intussusception (arrow).

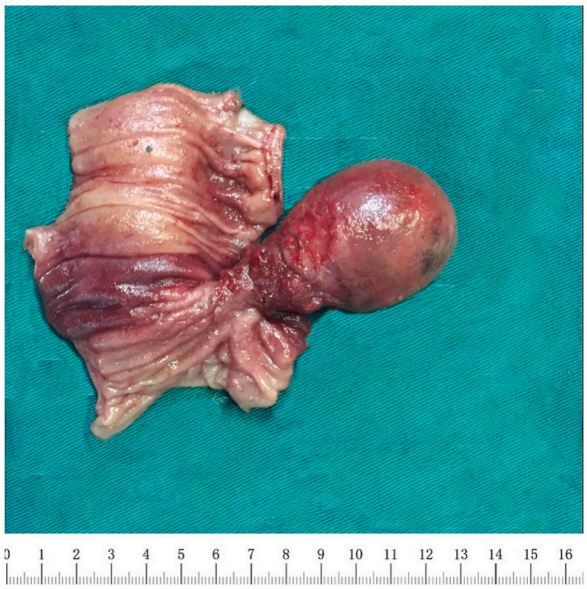


**Fig. 2.** The specimen revealed a polypoid pedunculated lesion (58 × 35 × 32 mm) in the ileum.

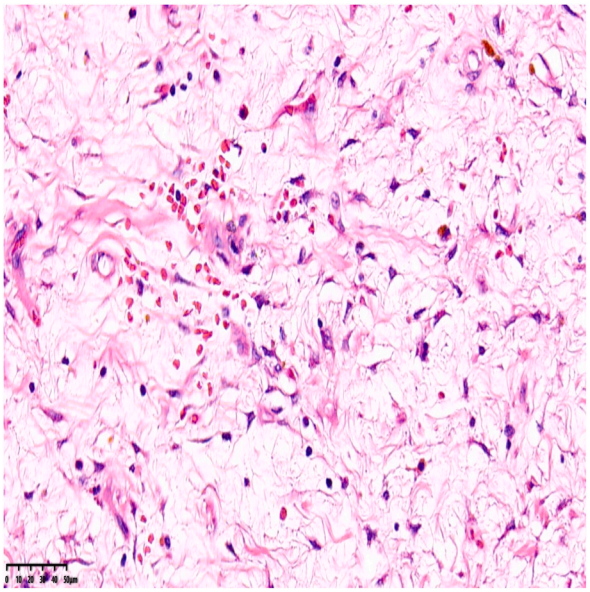

**Fig. 3.** Histopathology shows well-differentiated liposarcoma. (Original magnification × 200).

tumour-induced intussusception is that the tumour may be grasped and pulled forward through traction, causing the surrounding intestinal portion of the tumour to move easily to the distal intestine.

Ileal intussusception secondary to liposarcoma is an emergent surgical situation. Early diagnosis and appropriate treatment are essential because the mesentery of the involved segment is trapped between the overlapping layers of the bowel and its vascularity may be compromised. If intussusception is not treated in a timely manner, it can lead to serious complications, such as intestinal perforations [4]. Due to the lack of specific clinical symptoms of ileal intussusception secondary to liposarcoma, its clinical diagnosis relies more on imaging. Currently, CT is considered to be an ideal imaging technique for diagnosing intussusception secondary to liposarcoma because it reveals a pathognomonic bowel-within-bowel configuration while defining location and nature [5]. CT has a high sensitivity and specificity for the diagnosis of intussusception. CT can reveal the characteristic "target sign" on sagittal view and the "double tube sign" on axial view. In fact, a target-like or double-tube-like image depends on the angle of the beam relative to the intussusception. In addition, CT can accurately identify the lead point of intussusception. Different pathological types of liposarcomas exhibit characteristic CT features that can provide supportive evidence for their diagnosis. For well-differentiated liposarcomas, the

most common CT feature is characterised mainly by fat density with strip-like septa and/or small nodular soft tissue density shadows visible within it. On enhanced scanning, the lesion capsule and septa exhibit mild-to-moderate enhancement. However, identifying the lead points for liposarcomas can be challenging because they require differentiation from tumours rich in adipose tissue [6]. Lipoma is a tumour originating from the adipose tissue. The incidence of lipoma in adult intussusception has been reported to be 11.25 %. Currently, the clinical diagnosis of intussusception secondary to lipomas relies primarily on CT [7]. The CT image depicts the same image as fat tissue, with a CT value of approximately −80 to −120, and its density or signal is uniform, the boundary is clear and the morphology is regular. On enhanced scanning, the capsule and septa of the lesion do not exhibit any enhancement. However, the CT feature may be atypia when fat occurs ulcers and necrosis.

Currently, the prognosis for non-surgical treatment for submucosal liposarcoma is not favourable. Liposarcomas are malignant tumours composed of adipocytes with varying degrees of differentiation and atypia. Due to the presence of a pseudocapsule composed of cell fragments, inflammatory cells, and potential tumour tissue around liposarcomas, most submucosal liposarcomas exhibit invasive biological behaviours. Radical resection is the preferred treatment for submucosal liposarcomas. During the surgical procedure in this case, the tumour was completely removed with healthy edges. If a negative margin cannot be achieved with liposarcoma, the local recurrence rate increases after surgical resection. When submucosal liposarcoma induces intussusception, the surgical procedure is more challenging because rupture of the liposarcoma can cause tumour cells to spread, and injury of the intestinal tract can increase the risk for postoperative infection and death.

With the development of laparoscopic technologies, the advantages of laparoscopy have been increasingly accepted by both surgeons and patients. In recent years, surgeons have continuously performed laparoscopy for intussusception [8]. However, the use of laparoscopy or laparotomy for neoplasm-related intussusception remains controversial [9]. Currently, it is believed that laparoscopy can be attempted for intussusception associated with benign lesions. If malignant neoplasm-related intussusception is suspected before surgery, laparotomy is recommended. Laparotomy can flexibly use hand techniques, which is beneficial for reducing the risk for malignant tumour cell implantation in the abdominal cavity and avoiding the possibility of abdominal contamination caused by tearing or cutting of the intestinal canal. The patient in this case underwent radical resection with primary end-to-end ileal anastomosis via laparotomy. The patient recovered uneventfully and was discharged from hospital six days after surgical resection. The patient was doing well at the 6-month follow-up in the outpatient clinic.

## 4. Conclusion

Ileal intussusception in adults is a rare phenomenon caused by submucosal liposarcoma. This article describes a rare case of intussusception secondary to submucosal liposarcoma, which was successfully treated. Early diagnosis and appropriate treatment of neoplasm-related intussusception in adults are essential. Clinicians should consider the origin of submucosal liposarcomas in adult patients with neoplasm-related intussusception. Abdominal CT is regarded to be the ideal imaging technique for the diagnosis of intussusception secondary to liposarcoma. Once ileal intussusception secondary to submucosal liposarcoma is diagnosed, timely radical resection via laparotomy is recommended.

## Ethics declarations

The authors confirm that written consent for the submission and publication of this case report, including images and associated text, was obtained from the patient in accordance with COPE guidelines. And informed consent was received from participants. Review and/or approval by an ethics committee was not needed for this study because not applicable.

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## Data availability statement

Data associated with this study has been deposited at Wenzhou Medical University affiliated Zhoushan Hospital Data Set. http://www.zs-hospital.com/

## CRediT authorship contribution statement

**Hong-wei Yu:** Writing – review & editing, Writing – original draft, Validation, Investigation, Funding acquisition, Formal analysis, Conceptualization. **Jin-gang Yan:** Validation, Resources, Methodology, Formal analysis. **Lei Zheng:** Writing – original draft, Visualization, Resources. **Jun-hua Huang:** Writing – original draft, Project administration, Methodology, Investigation, Formal analysis, Data curation.

## Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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