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**Ileo-ileal intussusceptions caused by diffuse large B-cell lymphoma of the ileum**

Xu X *et al*. Adult intussusception

Xie-Qun Xu, Tao Hong, Bing-Lu Li, Wei Liu

**Xie-Qun Xu, Tao Hong, Bing-Lu Li, Wei Liu,** Department of General Surgery, Peking Union Medical College Hospital, Chinese Academy of Medical Sciences and Peking Union Medical College, Beijing 100730, China

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**Correspondence to**: **Dr. Xie-Qun Xu**, Department of General Surgery, Peking Union Medical College Hospital, Chinese Academy of Medical Sciences and Peking Union Medical College, Beijing 100730, China. [xiequnxu@gmail.com](mailto:xiequnxu@gmail.com)

**Telephone:** +86-10-69152610 **Fax:** +86-10-6915260

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**Abstract**

The occurrence of adult intussusception from small intestinal lymphoma is quite rare. We present a case of an 82-year-old man with two-month history of intermittent abdominal pain, nausea and fatigue. Clinical symptoms included moderate abdominal tenderness in the right lower abdomen. Computed tomography scan of the abdomen revealed a mass in the terminal ileum with the sign of “bowel within bowel” which was suspected as ileo-ileum intussusception. The patient underwent laparoscopical segmental ileal resection. Pathologic evaluation revealed a diffuse large B cell non-Hodgkin’s lymphoma of the ileum. The postoperative course was uneventful.

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**Key words**: Adult intussusception; Lymphoma; Small bowel; Ileum

**Core tip:** Intussusception is a common cause of bowel obstruction in pediatric patients; primary diffuse large B cell non-Hodgkin’s lymphoma of the small intestine is also rare. Intussusception due to primary diffuse large B cell non-Hodgkin’s lymphoma in the small intestine is even rarer in adult and it is often difficult to diagnose. This article offers the complete diagnosis and management.

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**INTRODUCTION**

Intussusception is most often seen in infants and children and only 5% of all cases occur in adults. It accounts for about 1% of all cases of adult bowel obstruction. Adult intussusception is rare and about 40% were related with malignant lesions[1]. In general, most of the lead points in the small intestine consist of benign lesions. Malignant lesions account for up to 30% of all cases of intussusception in the small intestine. Intussusception occurring in the large bowel is more likely to be malignant lesions in 63%-68% cases[2]. Primary malignant tumors of the small intestine are very rare, accounting for less than 2% of all gastrointestinal malignancies. The malignant lesions resulted in intussusception in the small intestine include primary adenocarcinoma, gastrointestinal stromal tumors (GISTs), lymphoma and carcinoid tumors[3].

Gastrointestinal tract is the most commonly involved site for the primary extranodal non-Hodgkin’s lymphoma (NHL), accounting for 20%-40% of all extranodal disease[4]. The stomach (50%-60%) is the most frequently affected site, followed by the small bowel (20%-30%), whereas 85% of the primary gastrointestinal lymphomas and 60%-80% of the intestinal lymphomas are B-cell type followed by T-cell NHL and Hodgkin’s lymphoma[5]. The ileum is the most common site affected by small intestine lymphoma. Intussusception is very rarely seen in intestinal NHL and the most common type of lymphoma causing intussusception is diffuse B-cell NHL[5].

We herein describe a case of adult ileal intussusception caused by diffuse large B-cell lymphoma of the small bowel in an 82 year old male patient.

### CASE REPORT

An 82-year-old male was admitted with two-month history of intermittent abdominal pain, nausea and fatigue. He also complained of a remarkable weight loss (8 kg) during the last two months. He was diagnosed as incomplete intestinal obstruction and was supported with parenteral nutrition in the community hospital. His past medical history was hypertension for 20 years. Physical examination revealed moderate abdominal tenderness in the right lower abdomen.

The blood test revealed a white blood cell count of 4.1 × 109/L with 63% neutrophils, a hemoglobin level of 96 g/L with a hematocrit of 29%. His liver and kidney function test results, tumor marker (CEA, CA19-9, CA242 and CA724) levels were all normal. The fecal occult blood was positive. The plain abdominal radiograph was unremarkable. The contrast enhanced computed tomography (CT) showed multiple lymphadenoma in the mesentery root of small intestine and posterior peritoneum, a mass in the terminal ileum with the sign of “bowel within bowel” which was suspected as ileo-ileum intussusception (Figure 1). Positron Emission tomography and computed tomography (PET-CT) showed high metabolism in the terminal ileum and multiple lymph nodes with high metabolism in the mesentery root of small intestine, in which malignant lesions in the terminal ileum were considered (Figure 2). Therefore, a ballon assisted enteroscopy was performed. A mass almost filled the ileum cavity about 50 cm away from the ileocecal valve (Figure 3) and did not allow the enteroscopy to pass. The biopsy was got from this lesion. The pathology result of this biopsy showed a diffuse large B cell NHL of the ileum (Figure 4). Laparoscopic exploration was performed because of low hemoglobin, weight loss and mass with suspected intussusception on CT. Laparoscopic exploration revealed a tumor mass of 5.0 cm × 3.0 cm ileo-ileum intussusception which was 40 cm to the ileum distal with multiple lymphadenoma in the mesentery root of small intestine. A segmental ileal resection was performed laparoscopically. After opening the specimen, a mass about 3 cm in the ileum wall almost filling the ileum cavity acted as the leading point. The histopathological findings were suggestive of a diffuse large B cell NHL of the ileum (Figure 4), which was coincident with the preoperative pathological results of endoscopic biopsy. Tumor infiltration was detected in 2 of 16 lymph nodes. The postoperative course was uneventful. Bone marrow biopsies showed a normal marrow without infiltration. Cervical and chest PET-CT scans were unremarkable. Postoperative chemotherapy was performed with clophosphamide, doxorubicin, vincristine, and prednisolone combined with monoclonal antibody rituximab. At the 6-month follow up, there was no evidence of tumor activity.

### DISCUSSION

Intussusception is rare in adults (< 5%), but it is the most common cause of intestinal obstruction in infants aged 6-18 mo[1]. The characteristic pediatric presentation triad of abdominal pain, palpable abdominal mass and bloody stool is rarely seen in adult cases. An abdominal mass could be palpated in 12%-30% cases, whereas 6%-25% cases would present with a complications of the disease such as obstruction, hemorrhage, perforation, or intussusception[5]. Most patients present with subacute (24.4%) or chronic (51.2%) symptoms of abdominal pain, nausea, vomiting and constipation. The non-specific clinical presentation makes the preoperative diagnosis difficult[6]. CT scan is one of the most useful preoperative diagnosis modalities for intussusception for it could show a thickened segment of bowel with an eccentrically placed crescent-like fatty area (bowel within bowel )[5]. If the patient doesn’t have a complete intestinal obstruction, colonoscopy would be helpful for a clinical and pathological diagnosis of the intussusception. PET-CT is useful for detecting the primary benign or malignant lesion sites and the distal regions which may be involved by lymphoma although it is not often routinely used[7].

About 52%-55% adult intussusceptions occur in the small intestine[8]. There were 63% adult small intestinal intussusception cases which were usually associated with benign lesions, 23% cases were idiopathic, and 14% cases were associated with malignant lesions[9] . The incidence of primary lymphomas in the small intestine accounts for less than 2% of all gastrointestinal malignancies and 10%-20% of small intestine malignancy[1].

For the management of small intestine intussusception caused by NHL, most of the authors think that the surgical intervention is mandatory for adult intussusception due to the high incidence of underlying malignancy in intussusceptions and the inability to differentiate non-operatively benign from malignant causes in enteric intussusceptions[5]. For adult small intestinal intussusception caused by NHL, primary surgical resection of the localized intestinal lesions with NHL is the major treatment modality, especially for those who have complications of intussusception.

In conclusions, we offered the relatively complete process for the diagnosis of adult ileal intussusceptions with detailed medical pictures in this specific case. Adult intussusception must be considered in the differential diagnosis in patients with abdominal pain and vomiting. The work-up must include X-Ray, ultrasound and CT scan of the abdomen; even the PET-CT in special cases. Surgical interventions are required and warranted once the diagnosis of intussusception is made, due to the high risk of malignancy and bowel obstruction.

**COMMENTS**

***Clinical diagnosis***

Ileo-ileal intussusceptions caused by diffuse large B-cell lymphoma of the ileum.

***Differential diagnosis***

Adult intussusception must be considered in the differential diagnosis in patients with abdominal pain and vomiting.

***Imaging diagnosis***

The imaging diagnosis must include X-Ray, ultrasound and CT scan of the abdomen which would show the sign of “bowel within bowel”.

***Treatment***

Surgical interventions should be required and warranted once the diagnosis of intussusception is made, due to the high risk of malignancy and bowel obstruction.

***Peer review***

This is an interesting case report, which shows a rare case of ileo-ileal intussusception caused by diffuse large B-cell lymphoma of the ileum.

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**Figure 1 Contrast-enhanced computed tomography showing suspected right ileo-ileum intussusception with a sign of “bowel within bowel” in the ileum (A, arrow, axial view), (B, arrow, coronal view).**

**Figure 2 Positron Emission tomography and computed tomography showing high metabolism in the right ileum (arrow) and multiple lymph nodes with high metabolism in the mesentery root of small intestine, in which malignant lesions in the terminal ileum was suspected.**

**Figure 3 Endoscopic findings showing a mass almost filling the ileum cavity about 50 cm far away from the ileocecal valve.**

**Figure 4 Histological and immunohistological examination of the endoscopic and surgical specimens showing diffuse large B-cell non-Hodgkin’s lymphoma**. A: × 400, HE staining; B: ×400, CD5 (-); C: × 400, CD10(-); D: × 400, CD20(+); E: × 400, CD23(-); F: × 400, MUM-1(+); G: × 400, Bcl-6(-); H: ×400, Cyclin D1(-).