



ISSN: 0975-833X

Available online at <http://www.journalcra.com>

International Journal of Current Research
Vol. 11, Issue, 06, pp.4433-4435, June, 2019

DOI: <https://doi.org/10.24941/ijcr.35350.06.2019>

**INTERNATIONAL JOURNAL
OF CURRENT RESEARCH**

RESEARCH ARTICLE

MULTIPLE JEJUNO-JEJUNAL INTUSSUSCEPTION IN AN ADOLESCENT

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ARTICLE INFO

Article History:

Received 18th March, 2019
Received in revised form
26th April, 2019
Accepted 21st May, 2019
Published online 30th June, 2019

Key Words:

Intussusception: Jejunum:
Multiple: Resection

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Citation: Dr. Cherian Mathew, Dr. Sooraj K, Dr. Binoj Kumar, Dr. Sai Kiran Reddy, Dr. Manohar Babu, Dr. Mary Treasa Pouline and Dr. Sahira 2019. "Multiple jejuno-jejunal intussusception in an adolescent", *International Journal of Current Research*, 11, (06), 4433-4435.

ABSTRACT

Adolescent and adult intussusception (AI) is a rare cause of bowel obstruction and is often associated with a pathologic lead point located in a single region of bowel. The mechanism of intussusception is not fully understood, but it is believed to result from a lesion in the bowel wall that alters the peristaltic pattern of the bowel causing invagination. In cases where no lead point is identified, intussusception may be due to submucosal bowel edema, fibrous adhesions or dysrhythmic contractions. We report a case of multiple simultaneous jejuno-jejunal intussusceptions in a 16-year-old boy who presented with a 1-month history of intermittent, crampy epigastric abdominal pain and nausea. All investigations were normal and Computed tomography imaging was inconclusive. The patient underwent Diagnostic laparoscopy, and five separate regions of intussusception were discovered in the jejunum, which was resected through a mini-laparotomy.

INTRODUCTION

Intussusception refers to the invagination (telescoping) of a part of the intestine into itself. It is the most common abdominal emergency in early childhood, particularly under 2 years of age (<https://www.uptodate.com/contents/intussusception-in-children>). According to current literature, while the incidence in paediatric population in Europe ranges between 0.66 and 2.24 per 1000 children admitted to hospital, the number of new cases in adults is around 2-3 cases per 1,000,000 of the general population annually (Trotta, 2016). Intussusception is considered rare before 3 months and after 6 years of age, resulting, therefore, relatively unusual in adolescents and adults, where the diagnosis is commonly overlooked (Jiang, 2013). Moreover, adult's intussusception cases can be distinguished from paediatric cases in various aspects. First, in children, it is usually primary (idiopathic) and benign. On the contrary, in almost 90% of adulthood intussusception cases an underlying condition (Meckel's diverticulum, polyp, intestinal carcinoma, etc), that serves as a lead point, can be discovered intraoperatively. A second important difference is the clinical scenario. Paediatric intussusception often manifests with sudden onset of intermittent, severe, crampy abdominal pain, inconsolable crying, vomiting, bloody mucoid stools with or without the presence of a palpable mass (Jiang, 2013). Nevertheless, the classic described triad of pain, palpable sausage shaped

abdominal mass, and currant-jelly stool is nowadays seen in less than 15 percent of patients at the time of presentation, especially in older subjects. Adults, on the other hand, may present to emergency department with acute, subacute, or chronic non-specific and long-standing symptoms. Therefore, the initial diagnosis is often missed or delayed and may only be established at the operating theatre. A high index of suspicion is often needed for an early diagnosis, to avoid the developing of serious complications (Lianos, 2013).

Case Report

We report a case of multiple simultaneous jejunal intussusceptions in a 16-year-old boy who presented with a history of intermittent crampy epigastric abdominal pain and nausea for 1 month. He was able to tolerate diet. He had one episode of vomiting. His vital signs, physical examination and laboratory studies were unremarkable. He did not have a history of prior abdominal surgery or malignancy. A CT scan was done which was found to be normal, without evidence of bowel obstruction or mass. The patient underwent diagnostic laparoscopy using a 10 mm 30° camera and two 5 mm working ports. Five separate regions of intussusception within the jejunum were discovered (Fig. 1). All areas of intussusception were long and they recur after reduction, which was resected through a mini-laparotomy. There was no obvious evidence of masses, adhesions or serosal abnormalities along the small bowel.

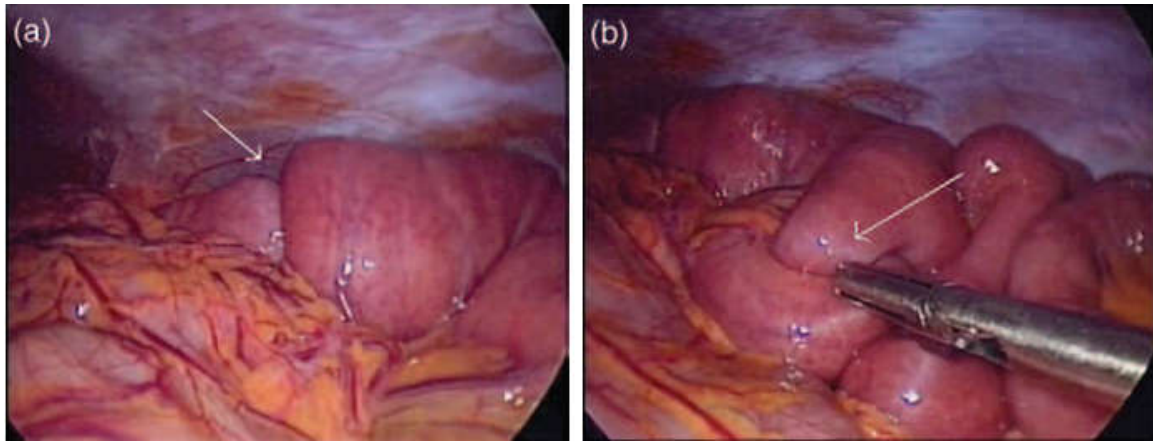


Figure 1. Small bowel intussusception (arrow) that did not reduce spontaneously

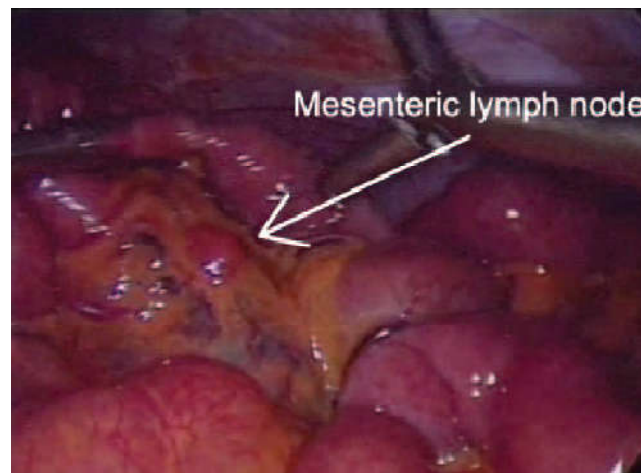


Figure 2. Prominent mesenteric lymph nodes (arrow) discovered during laparoscopy

There were prominent lymph nodes noted in the small bowel mesentery (Fig. 2). The postoperative course was uneventful and he was discharged 5 days after his operation. He had resolution of symptoms and had no recurrence after 1 years of follow-up. Pathologic examination of the small bowel demonstrated no evidence of malignancy, lead point or pathologic changes. Likewise, there was no evidence of acid-fast organisms, fungal or helminth parts identified. The enlarged mesenteric lymph nodes demonstrated reactive follicular hyperplasia.

DISCUSSION

Intussusception is the most common abdominal emergency in early childhood (<http://emedicine.medscape.com/article/937730-treatment#d11>) whereas it represents a relatively rare clinical scenario in adolescents (like our patient) and young adults, frequently implicating a significant delay or even an erroneous conclusion in the diagnostic process. Clinic is usually nonspecific and possibly misleading, especially during the seasonal epidemic peak of gastrointestinal infection. Our patient, for example, presented with recurrent abdominal pain and no palpable mass or other characteristic physical signs and symptoms, thus hardly discernible from other more common causes. A literature review of 1214 patients with intussusception revealed that 63 % of adult intussusception was tumor-related, 50 % of which were malignant. A malignant tumor was the etiology in 48 % of patients with colocolonic intussusception and in 17 % of those with enteric intussusceptions (Felix, 1976).

We retrospectively reviewed 16 years of data from patients with intussusception and found that 77.3 % were related to a tumor, 73.5 % of which were malignant. Among them, 11.3 % occurred in a postoperative setting, and 11.3 % were idiopathic. Malignant tumor was the etiology in 90.0 % of patients with colocolonic intussusception and in 25.0 % of those with enteric intussusception. More than 90 % of adult intussusception patients have distinct causes that are related to the small or large intestine. This is in contrast to the 90 % of childhood intussusception cases that are idiopathic (Felix, 1976). Etiologies of adult intussusception include tumor- or surgery-related, idiopathic, and "other." Benign or malignant tumors are the most frequent cause of intussusception in adults. Many colocolonic intussusceptions were related to primary adenocarcinoma of the colon. Generally, many malignant tumors of the small bowel are metastatic tumors. Postoperative factors are the second most common etiology of intussusception in adults (Azar, 1997 and Felix, 1976). Most affected patients have pre-diagnosis episodes of intermittent abdominal pain and vomiting (Azar, 1997). The most common symptoms are due to bowel obstruction and include crampy abdominal pain (71 %), nausea and vomiting (68 %), and sensations of abdominal fullness (45 %) and tenderness (60 %) (Brayton, 1954). The emergence of acute symptoms due to complete intestinal obstruction occurs in fewer than 20 % of patients. Similar results were seen in our study, and the incidence of long-term symptoms was 27.3 %. It is often difficult to diagnose adolescent intussusception because the clinical findings are not clear. In the past, colon intussusception was diagnosed with a contrast enema showing

a crab claw-like shadow, but the accuracy of preoperative diagnosis was only 20–25 %. Intussusception in adults and adolescent is often discovered only during exploratory surgery (Gordon, 1991). Abdominal US and CT diagnose intussusception with high sensitivity. CT examination typically reveals a three-layer structure that includes the intestinal wall, its mesentery, and wrapping intestine. It sometimes reveals the tumor as a lead point (Iko, 1984). In a report by Azar and Berger, abdominal CT accurately diagnosed intussusception in 78 % of patients (Iko, 1984), CT is the most useful examination for diagnosing intussusception. In our study, US and CT assessments and Plain abdominal radiography was of no value.

In adults, it is important to diagnose the organic intussusception lesion to help guide treatment decisions. Enema or colonoscopy examinations can reveal and reduce the intussusception as well as facilitate qualitative diagnosis of the organic lesion (Omori, 2003). Before the mid-1950s, intraoperative reduction of intussusception followed by removal of the organic lesion was the treatment of choice. Subsequently, it was recommended that colocolonic-type surgical resection be performed without reduction because of the high incidence of malignant disease associated with intussusception (Brayton, 1954). Other investigators then suggested that intussusception in adults should be resected without reduction regardless of the site because malignant disease is highly associated with enteric intussusception (Weilbaecher, 1971). More recently, surgical treatment has been determined according to the length of the affected small intestine in patients with enteric intussusception because of the relatively low incidence of primary malignant disease. In other words, surgery is performed without reducing the intussusception if the affected portion of the small intestine is not extensive. If resection of a long segment of bowel is required, intraoperative reduction is attempted to reduce the length of the resection. Surgical resection without reduction should be limited to primary malignant disease (Felix, 1976). Sarr et al (1981) at the Mayo Clinic recently questioned the accepted notion that preoperative reduction of intussusception was not recommended because it was associated with malignant disease.

Tumor cells are always flowing out of primary lesions, and the reduction of intussusception causes little damage to the intestinal mucosa. Patients with enteric and ileocolic intussusceptions do not have leading points, so there is no need for removal. Preoperative reduction serves several functions, including avoidance of emergency surgery, allowing radical surgery for cancer, and reducing the extent of the intestinal resection. It also allows time for preoperative preparation of the bowel. Careful radiologic or endoscopic evaluation can detect strangulated intussusception that is impossible to reduce preoperatively. These observations have resulted in a shift in the clinical paradigm for reducing the intussusception. In our case as the patient had recurrent symptoms and intraoperatively found to have long segments of jejunum-jejunal intussusceptions, we decided to go for a limited resection of jejunum.

Conclusion

Adolescent and adult intussusception (AI) is a rare cause of bowel obstruction and is often associated with a pathologic lead point located in a single region of bowel. Adolescent jejunum-jejunal intussusception is a rare cause of abdominal pain and clinicians must be vigilant in considering intussusception as a potential cause for intestinal obstruction in children of all ages. The diagnosis of intussusception is often a challenge to emergency physicians because most patients present with nonspecific signs and symptoms. The index of suspicion should be particularly high in older children who often have pathologic lead points and atypical presentations: delayed diagnosis can lead to ischemic complications.

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