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Jejunojejunal Intussusception in a Young Adult Caused by a Polyp Associated with Peutz-Jeghers Syndrome: A Case Report

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Abstract

Introduction: Jejunojejunal intussusception is rare in adults. Peutz-Jeghers syndrome is one of the etiologies in young adults. **Case Report:** This was a 19-year-old patient admitted to the Emergency Department of Saint Louis Hospital in La Rochelle with a clinical presentation of bowel obstruction, characterized by sudden-onset severe abdominal pain and vomiting. On examination, an abdominal mass was palpable in the left periumbilical and epigastric regions, with guarding. Abdominopelvic computed tomography revealed a large jejunojejunal intussusception without signs of ischemia. Surgical exploration by laparotomy showed that the intussusception was located approximately 50 cm from the duodenojejunal flexure. Reduction was performed, followed by surgical resection of 75 cm of jejunum with a mechanical side-to-side anastomosis. The postoperative course was uneventful, and histological examination of the surgical specimen concluded that the lead point was a Peutz-Jeghers hamartomatous polyp. **Rationale:** Limited data are available in France on adult intestinal intussusception. The report highlights an important benign lead point to consider in young adults with small-bowel intussusception.

Keywords

Jejunojejunal Intussusception, Adult, Peutz-Jeghers Syndrome

1. Introduction

Acute intestinal intussusception is the sudden invagination of a proximal intesti-

nal segment into the immediately distal segment, in the isoperistaltic direction. It is a rare entity in adults, accounting for only 1% of all intestinal obstructions and 5% of all cases of intestinal intussusception [1]. In children, it is idiopathic in approximately 75% to 90% of cases [2]. In adults, however, 90% of cases have an identifiable cause, including adenomas, lipomas, neurofibromas, scleroderma, Peutz-Jeghers syndrome, and primary or secondary cancers [3]. The jejunojejunal form is a rare anatomical subtype. We report a case of jejunojejunal intussusception in a 19-year-old male adult secondary to a Peutz-Jeghers syndrome polyp.

2. Observation

This case involves a 19-year-old patient with no significant medical history, including no prior abdominal symptoms, anemia or bleeding history, and any previous episodes of obstruction or intussusception as well as no family history.

He presented to the emergency department of Saint Louis Hospital in La Rochelle with right upper quadrant pain and vomiting of sudden onset occurring approximately 5 hours earlier.

During evaluation in the emergency department, he remained in severe pain despite receiving 10 mg of morphine. Hemodynamic parameters were stable, he was afebrile, and examination revealed a left periumbilical and epigastric mass with guarding. Laboratory tests showed leukocytosis with a white blood cell count of 17,000; C-reactive protein was negative, and the remainder of the workup was unremarkable.

Abdominopelvic CT revealed a large peri-umbilical jejunojejunal intussusception, with the typical "target" appearance on axial images and a doughnut sign, as shown in **Figure 1**. The involved loop was distended, with a maximal measured diameter of 51 mm and a thickened wall.

There was mesenteric fat stranding associated with numerous subcentimetric mesenteric lymph nodes and a thin layer of intraperitoneal fluid in the rectouterine pouch (Douglas pouch). There were no signs of bowel ischemia.

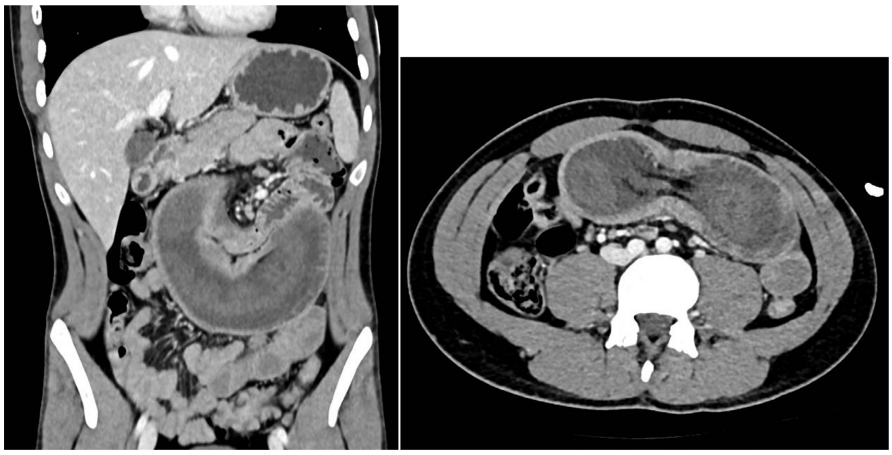

Figure 1. CT images: coronal and frontal sections.

Given this acute intestinal intussusception, an indication for emergency surgery was established, consisting of a segmental small-bowel resection via laparotomy. The patient was placed in the supine position under general anesthesia. A midline supraumbilical incision was performed, extending slightly below the umbilicus.

Upon entering the peritoneal cavity, a very small amount of clear, straw-colored fluid was observed. A markedly dilated loop was present in the mid-abdomen (**Figure 2**), located approximately 50 cm from the duodenojejunal flexure. The dilated segment measured more than 20 cm. Distal to this, the small bowel was completely collapsed. A large intussusception was located within the dilated segment. An attempt at reduction, given the degree of dilation, immediately caused a serosal tear. It was therefore necessary to resect the entire intussuscepted small-bowel segment, which showed signs of ischemic compromise with vascular congestion.

Enlarged mesenteric lymph nodes exceeding 1 cm were observed, and after successful reduction of the intussusception, the mesenteric resection was completed, removing as many lymph nodes as possible.

The specimen (**Figure 3**) was submitted for histopathological examination.

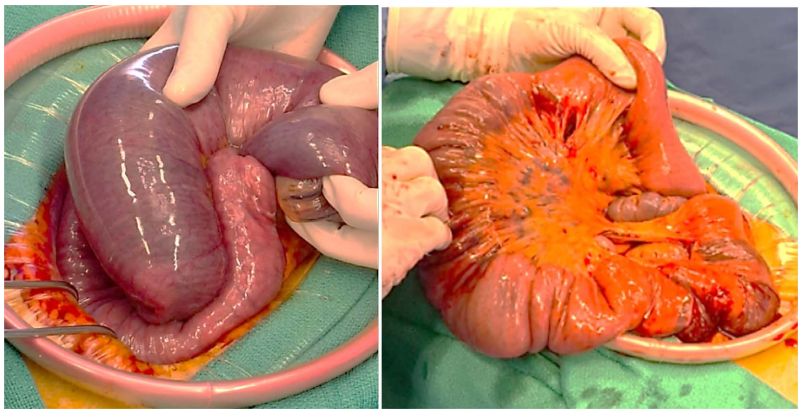

Figure 2. Intraoperative images before and after reduction of the intussusception.

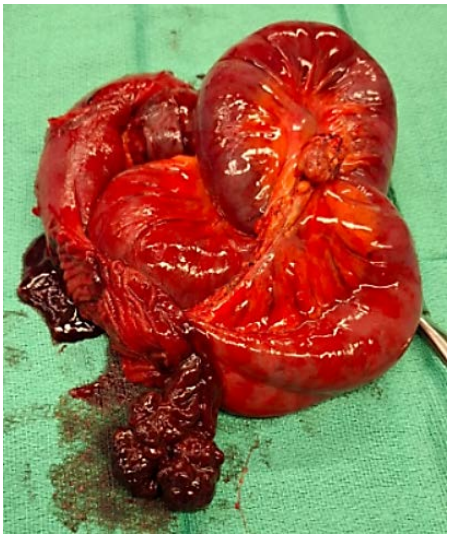

Figure 3. Image of the surgical specimen and the polyp.

Near the initial site of intussusception, it contained a polypoid lesion measuring more than 3 cm, which was responsible for the intussusception. A mechanical side-to-side jejunojejunal anastomosis was performed using an Echelon 80 stapler, followed by peritoneal lavage. Palpation of the duodenum and the remaining small bowel revealed no other polypoid lesions.

The postoperative course was uncomplicated, and the patient was discharged on postoperative day 7.

Concerning the postoperative follow-up, MR enterography and colonoscopy were performed 2 months after surgery.

Total colonoscopy identified a large pedunculated polyp in the sigmoid colon, approximately 3 cm in maximal diameter, which was resected using a diathermic snare, as well as a 1-cm polyp in the rectum and another in the right colon.

Postoperative MR enterography, as well as esophagogastroduodenoscopy revealed a large polyp, apparently pedunculated between the superior genu and the second portion of the duodenum, away from the papilla, measuring 53 × 30 × 50 mm, with no upstream obstructive syndrome. The patient was referred to Bordeaux University Hospital for resection of his duodenal polyp.

Examination of the surgical specimen, approximately 75 cm in length, revealed a well-demarcated hemorrhagic infarction extending over about 30 cm surrounding a single 36-mm pedunculated polyp that was also markedly infarcted, ulcerated, and hemorrhagic, with 90% necrosis. Its arborizing architecture was suggestive of a Peutz-Jeghers hamartomatous polyp, with a few glandular atypia (at most low-grade dysplasia). The polyp was located 4 cm from a surgical margin that was also partially infarcted; the other margin was essentially unremarkable, aside from mild intraepithelial lymphocytosis. Similarly, polyp biopsies from the duodenum, right colon, and sigmoid colon supported the diagnosis of a Peutz-Jeghers hamartomatous polyp. STK11 testing and oncogenetic assessment was planned.

The seven excised lymph nodes were variably enlarged, with hemorrhagic infarction and/or sinus distension. Moreover, there was no histological evidence of malignancy.

3. Discussion

Intussusception is an unusual cause of intestinal obstruction in adults.

Our case involves a 19-year-old young adult. A study of a series of 13 cases reported ages ranging from 24 to 61 years [4]. Another larger series involving 170 adults found a mean age of 41 ± 17 years (range, 15 - 95 years) [5].

The clinical presentation of adult intussusception is variable. The most common initial symptom is abdominal pain, reported in almost all patients, followed by cessation of stool passage (constipation) and/or vomiting (approximately two-thirds of cases) [4]. The classic triads described in the pediatric population—the Ombredanne triad (abdominal pain, vomiting, rectal bleeding) and the Fèvre triad (pain, rectal bleeding, palpable intussusception mass)—are rare in adults [6]. In our case, the patient presented with severe abdominal pain that persisted despite morphine ad-

ministration, vomiting, and a palpable intussusception mass.

The consultation delay was 6 hours. As in our case, approximately 60% of patients present with acute symptoms lasting from 6 hours to a few days and are admitted to the emergency department [4].

Abdominal computed tomography is the reference examination for confirming the diagnosis, specifying the location, and, when present, identifying signs of bowel compromise and complications (perforation with free intraperitoneal air). The typical CT finding is a heterogeneous soft-tissue mass with a target- or sausage-like appearance, showing a layering effect [6]. In our case, there was little discrepancy between the preoperative CT and the operative findings, it described as showing no ischemia, whereas surgery later found ischemic compromise and vascular congestion. In fact, the diagnosis is not always established preoperatively. CT accuracy is approximately 75% [7].

Abdominal ultrasonography is an important diagnostic tool, with lower radiation exposure and lower cost. Classic imaging signs include the "target" or "ring" appearance on transverse sections and the "sandwich" appearance on longitudinal sections. However, it is an operator-dependent examination, as it requires performance by an experienced operator. Similarly, obesity and the presence of a large amount of air in distended bowel loops limit image quality and, consequently, diagnostic accuracy [6].

Regarding the location of intussusception, an analysis of 170 computed tomography scans in adult patients found that intussusception was predominantly entero-enteric (87.6%), whereas ileocecal (4.7%) and colocolic (5.9%) forms were less common [5].

Adult intussusception has an identifiable cause in 70% to 90% of cases [8]. When the lead point is in the small intestine, the cause tends to be benign (adenoma, lipoma, polyp, Meckel's diverticulum). In contrast, it is often malignant when the lead point is in the colon (carcinoma, lymphoma, gastrointestinal stromal tumors, metastases from melanoma, or intestinal sarcoma) [8]-[10].

Most polyps measure between 2 and 5 cm in diameter [11]. In our case, it measured 3 cm.

From a surgical standpoint, management is immediately operative given the risk of necrosis and perforation of the involved bowel loops. Other major considerations also support surgical treatment, including the frequent presence of an underlying etiology that itself requires operative management and the prevalence of malignant disease as the initiating cause [12].

However, some authors have reported cases of nonoperative treatment in adults, resulting in improvement in a small minority, but with recurrence or symptom chronicity in most cases [5] [9].

Once the decision to proceed with surgical management has been made, the question arises as to whether to use a laparoscopic or open (laparotomy) approach. The choice of surgical procedure is determined by the location, size, etiology, and viability of the intestine [13].

From a diagnostic standpoint, the laparoscopic approach offers the advantage of confirming the diagnosis and identifying the anatomical nature of intestinal intussusception, which may be misinterpreted on imaging. From a therapeutic standpoint, however, the challenge lies in attempting reduction and resection laparoscopically, as technical feasibility depends closely on the experience of the team managing the patient. Overall, laparoscopic management of adult intestinal intussusception is comparable to open surgery. However, the use of laparoscopy in emergency settings and its acceptance as a valid approach for adult intestinal intussusception remain controversial [10]. In our case, laparotomy was chosen over laparoscopy because of the marked distension of the intussuscepted bowel loop, with a thickened wall and a maximum diameter measuring 51 mm. The risk of perforation being significant.

With regard to the intraoperative procedure, considerable debate surrounds two approaches: reduction followed by resection versus routine primary resection without reduction.

The disadvantages of reduction include the theoretical risk of intraluminal seeding or venous embolization at sites of ulcerated mucosa. Other concerns related to reduction include the possibility of perforation during manipulative maneuvers and the risk of dissemination in cases of primary tumors or isolated metastases [9]. The advantage of reduction is that it allows assessment of bowel viability and thereby reduces the number of unnecessary resections.

Therapeutic approaches therefore appear to converge toward primary reduction in cases of benign lesions and systematic resection in cases of malignant lesions. However, the main challenge remains distinguishing malignant from benign lesions before reduction. [7]. A definitive diagnosis is provided by histopathological examination, which in most cases is performed on the surgical specimen.

Predictive factors for malignant etiology include, among others, advanced age, colonic location, and chronic symptoms. In our case, the patient was young, with acute pain and a jejunal location. The overall presentation suggested a benign etiology; therefore, we initially performed reduction before completing the resection.

Histological examination initially suggested a Peutz-Jeghers hamartomatous polyp with some glandular atypia. Slightly more than half of Peutz-Jeghers syndromes are familial, with the remainder corresponding to sporadic de novo mutations (of the STK11 gene). It is a rare syndrome, with an incidence ranging from 1 per 8300 to 1 per 120,000 births depending on the series [14].

Polyps in Peutz-Jeghers syndrome (PJS) are generally not true neoplasms; however, their sometimes substantial size can cause small-bowel obstruction. Jejunal polyps may number as many as 100 and require multiple surgical interventions, potentially leading to short bowel syndrome. The mean age at surgery is typically approximately 21 years [15].

An oncogenetics consultation was requested, given the multiple polyps detected throughout the gastrointestinal tract. Regarding to surveillance, colonoscopy, upper gastrointestinal endoscopy, and small bowel assessment by magnetic reso-

nance enterography or video capsule endoscopy are recommended every two to three years from the age of 18 years onward. Annual pancreatic surveillance by endoscopic ultrasonography or magnetic resonance imaging/magnetic resonance cholangiopancreatography (MRI/MRCP) is recommended between 30 and 35 years of age [16].

4. Conclusion

Jejunojejunal intussusception is a rare cause of intestinal obstruction in adults. The main clinical sign is sudden-onset acute abdominal pain. Computed tomography provides high diagnostic accuracy. In adults, an underlying lesion serving as a lead point is identified in most cases. This etiology is generally benign in young adults, and Peutz-Jeghers syndrome is one such cause. Surgical resection is the recommended treatment, with low postoperative morbidity and mortality.

Authors' Contributions

All authors contributed to the patient's management and to the writing of the article.

Ethical Considerations

The patient gave consent for the anonymous publication of data from his medical record.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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