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Small Bowel Intussusception Secondary to a Peutz-Jeghers Hamartomatous Polyp: A Rare Cause of Acute Abdominal Pain

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ABSTRACT

Background:
Intussusception in adults is an uncommon cause of intestinal obstruction, differing from pediatrics cases where idiopathic aetiologies predominate. In adults, it is usually associated with an underlying pathological lead point, frequently neoplastic. Peutz-Jeghers syndrome (PJS), an autosomal dominant disorder characterized by mucocutaneous pigmentation and hamartomatous gastrointestinal polyps, is a rare but important cause. Although PJS predisposes to both benign and malignant neoplasms, small bowel intussusception secondary to hamartomatous polyps is infrequently reported.

Case Report:
We present the case of a 27-year-old male with no significant past medical history who presented with progressive severe abdominal pain, nausea, and a single episode of vomiting. Physical examination revealed a vague abdominal mass, and laboratory investigations demonstrated leucocytosis and mild lactic acidosis. Contrast-enhanced abdominal CT showed a whirlpool sign and mural hematoma suggestive of volvulus or intussusception. Emergency laparoscopy converted to mini-laparotomy revealed a 20–30 cm ischemic small bowel segment with intussusception approximately 140 cm proximal to the ileocecal junction. A firm intraluminal polypoid lesion served as the lead point. Segmental small bowel resection with primary anastomosis was performed. Histopathological analysis confirmed a Peutz-Jeghers-type hamartomatous polyp without dysplasia or malignancy. The patient had an uneventful postoperative recovery and was discharged in stable condition.

Conclusion:
This case highlights the diagnostic and therapeutic challenges of small bowel intussusception in adults. Although rare, PJS-related intussusception should be considered in young adults presenting with nonspecific abdominal pain and bowel obstruction. Early recognition with cross-sectional imaging and prompt surgical intervention are essential to prevent ischemia and improve outcomes. Surveillance strategies remain critical in patients with PJS to prevent recurrence and other complications.

Keywords: Abdominal Pain, Hamartoma, Intussusception, Peutz-Jeghers Syndrome, Small Bowel Obstruction

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INTRODUCTION

Intussusception, a rare cause of bowel obstruction in adults, is even less common in the elderly population. Unlike pediatric cases, adult intussusception is primarily associated with pathologic diseases acting as lead points, often requiring surgical intervention.[1] One such cause is Peutz-Jeghers syndrome (PJS), a hereditary condition marked by the presence of hamartomatous polyps, which may result in intestinal obstruction.

CASE REPORT

A 27-year-old male with no significant past medical history presented to the emergency department with a few days of progressive, severe abdominal pain. The pain was diffuse but most pronounced in the epigastric region. He also reported nausea and a single episode of vomiting. He denied any history of trauma, fever, diarrhea, or urinary symptoms. Notably, the patient had no other clinical features suggestive of Peutz-Jeghers syndrome, such as melanocytic macules or a family history of gastrointestinal neoplasms.

On examination, the patient had stable vital signs, and a vague, palpable mass was noted in the left abdomen. Laboratory results showed leukocytosis (WBC 16 x 10^9/L) and mild lactic acidosis (lactate 3 mmol/L). A contrast-enhanced abdominal CT scan revealed a small bowel mural hematoma and a whirlpool sign suggesting the possibility of volvulus (Figure 1).

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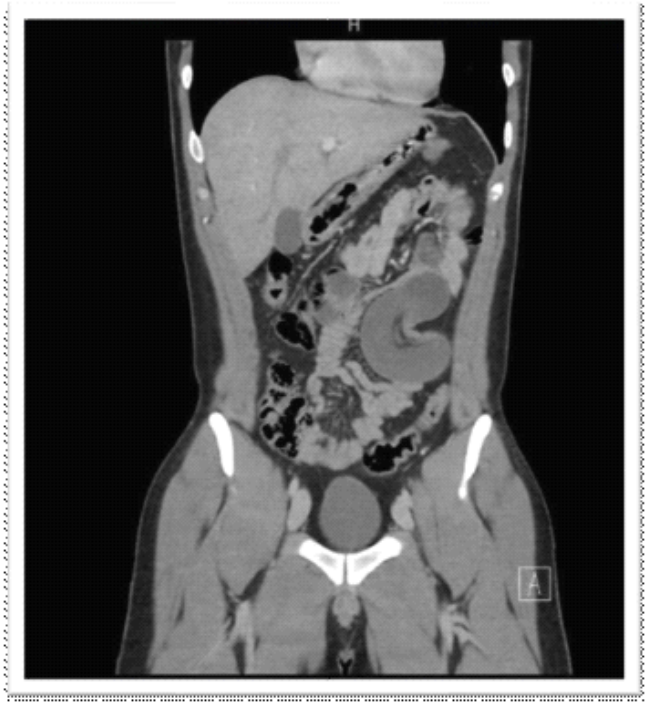

Figure 1: Whirlpool Sign on Contrast-Enhanced CT Abdomen.

The patient was kept NPO and scheduled for emergency diagnostic laparoscopy with possible bowel resection. Intraoperatively, the small bowel was examined from the ligament of Treitz to approximately 230 cm distally revealing a dilated segment leading to an area of small bowel intussusception at approximately 140 cm from the ileocecal junction. Attempts at laparoscopic reduction were unsuccessful, prompting conversion to a mini-laparotomy for better control. A 20-30 cm segment of the intussuscepted bowel was identified, appearing severely congested with ischemic changes. A firm, intraluminal polypoid mass with a stalk, measuring approximately 3 x 4 cm was palpated 200 cm distal to the ligament of Treitz, serving as the lead point (Figure 2,3).

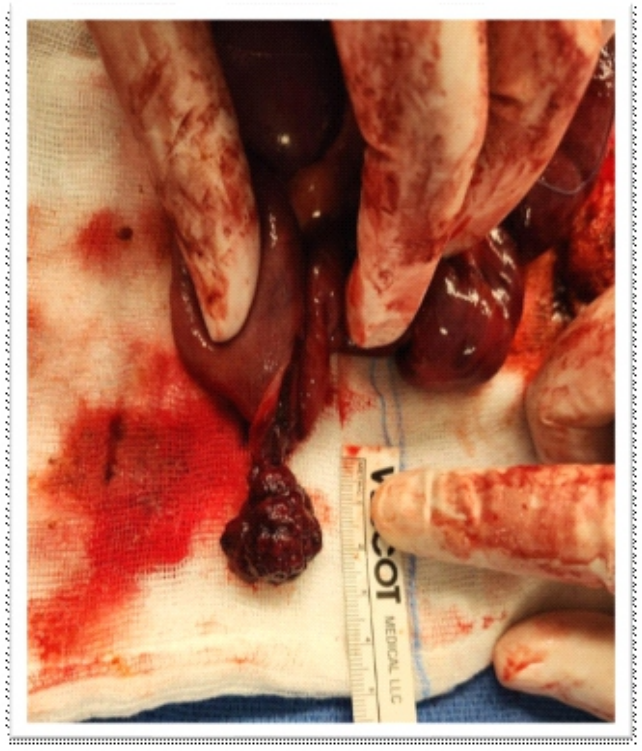

Figure 2: Intramural Polypoidal mass with a stalk.

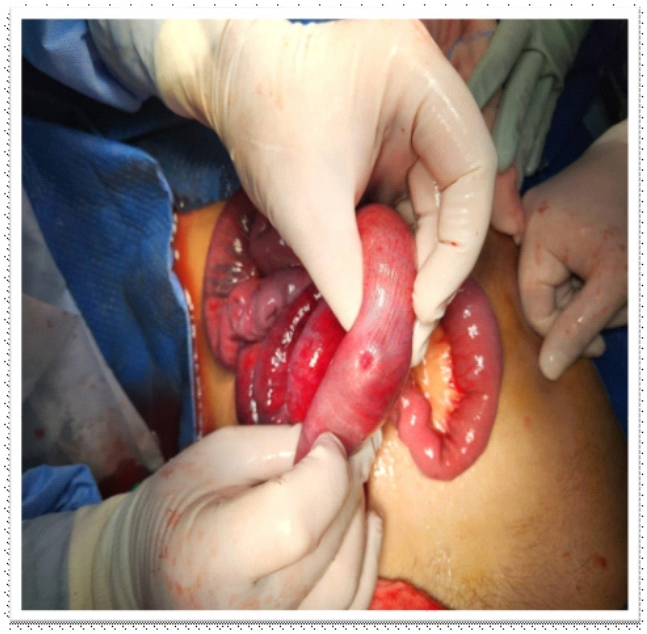

Figure.3: lead point seen.

A 50 cm segmental small bowel resection with primary anastomosis was performed. The small bowel was carefully examined during the procedure, and no other areas of pathology or associated lesions were identified. The patient had an uneventful postoperative recovery and was discharged on analgesia and a PPI after stabilization. Histopathological examination of the resected specimen confirmed a Peutz-Jeghers-type hamartomatous polyp (2.5 x 2 x 2 cm) with no dysplasia or malignancy. The resection margins were viable, and two reactive mesenteric lymph nodes were identified (Figure 4).

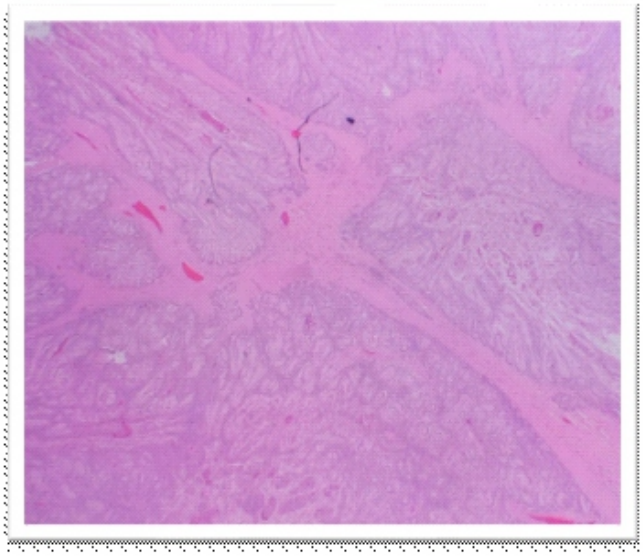

Figure .4: - Histopathological appearance showing characteristic arborizing tree-like pattern.

DISCUSSION

Small bowel intussusception in adults is a rare condition, often secondary to an underlying pathological lead point. Hamartomatous polyposis is a rare cause of intussusception in adults. But this complication is the most frequent for patients with Peutz-Jeghers syndrome.[2] In older adults, these lead points are commonly malignant tumors, while in younger individuals, benign causes such as polyps or benign tumors, including hamartomas, are more frequently identified. This case report discusses a 27-year-old male who presented with small bowel intussusception caused by a Peutz-Jeghers-type hamartomatous polyp, a rare but noteworthy etiology for adult intussusception.

Peutz-Jeghers syndrome is an autosomal dominant disorder characterized by melanocytic macules of the lips, buccal mucosa, and digits, multiple gastrointestinal hamartomatous polyps and an increased risk of various neoplasms.[3] PJS also predisposes individuals to various malignancies, including those of the gastrointestinal tract, pancreas and breast. While PJS is a recognized cause of gastrointestinal polyps, small bowel intussusception due to these hamartomatous polyps is uncommon. Intussusception can involve any portion along the gastrointestinal tract but is most commonly reported in the small or large intestine with only a few, scattered case reports of gastro-gastric intussusception.[4] In addition to the more commonly reported small and large bowel involvement, cases of jejunoduodenal and ileojejunal intussusception have also been described in patients with PJS. Aydin et al reported a rare case of jejunoduodenal intussusception caused by a solitary duodenal polyp, underscoring the anatomic variability and clinical heterogeneity of this syndrome.[5] In most adult cases of intussusception, malignancy is the predominant cause, as it forms a more substantial lead point. However, in PJS, these benign hamartomatous polyps can serve as a lead point for intussusception, as demonstrated in this patient.

The clinical presentation of adult intussusception can often be nonspecific, with symptoms such as abdominal pain, nausea, vomiting and bloating. These symptoms are frequently mistaken for other gastrointestinal disorders, which can lead to delayed diagnosis.[6] In this case, the patient's abdominal pain and nausea, along with a palpable abdominal mass, initially raised concerns for bowel obstruction or volvulus. However, imaging studies, particularly a contrast-enhanced abdominal CT, provided more definitive findings, including the characteristic whirlpool sign and bowel mural hematoma which suggested intussusception rather than other causes of obstruction. These imaging findings guided the decision for surgical intervention.

Surgical management of adult intussusception typically involves either reduction or resection, depending on the severity of bowel compromise and the underlying etiology.[7] In this case, the intussuscepted bowel segment was severely ischemic, requiring bowel resection. The discovery of a polypoid mass with a stalk as the lead point confirmed the diagnosis and necessitated resection of the affected segment. The patient's uneventful recovery post-surgery highlights the importance of early and appropriate surgical intervention in such cases.

Histopathological examination of the resected specimen confirmed the presence of a Peutz-Jeghers-type hamartomatous polyp measuring 2.5 x 2 x 2 cm, with no evidence of dysplasia or malignancy. This finding aligns with previous reports indicating the benign nature of these polyps, despite their potential to cause significant complications such as intussusception or bowel obstruction. The resection margins were viable and two reactive mesenteric lymph nodes were identified indicating a localized inflammatory response rather than metastatic disease.

Given the potential for these polyps to cause acute complications, regular surveillance in patients with PJS is crucial. Gene Reviews recommends periodic small-bowel imaging via magnetic resonance enterography (MRE) or video capsule endoscopy (VCE) starting in adolescence, ideally every 2–3 years. Such proactive screening can aid in the early detection and endoscopic removal of polyps before they reach sizes likely to cause obstruction or intussusception.[8]

This case underscores several important points in the management of small bowel intussusception in adults, particularly in the context of underlying genetic disorders like Peutz-Jeghers syndrome. The rarity of intussusception due to hamartomatous polyps calls for a broad differential diagnosis, especially when encountering young adults with nonspecific abdominal symptoms. While imaging techniques such as CT can assist in diagnosing intussusception, surgical intervention remains the cornerstone of treatment, particularly when bowel ischemia is present.

CONCLUSION

Hamartomatous polyposis is a rare cause of intussusception in adults, but this complication is most frequent in patient with Peutz Jeghers syndrome. Early recognition, appropriate imaging and timely surgical intervention are essential to prevent complications such as bowel ischemia and ensure favorable outcomes. This case emphasizes the importance of comprehensive evaluation and underscores the need for surgical intervention in managing adult intussusception.

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