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Intestinal Invagination Secondary to Vanek Tumor, A Rare Cause of Intestinal Obstruction in Adults: Case Report

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ABSTRACT

Introduction: Mechanical intestinal obstruction is a common surgical condition with a wide range of causes, including intrinsic intestinal problems such as intestinal invagination. This condition is extremely rare in the adult population and is mostly caused by neoplasms. The clinical presentation, although varied, is typically gradual and nonspecific. Among intestinal neoplasms, inflammatory fibroid polyps (IFPs) are very rare, constituting less than 0.1% of gastrointestinal tract polyps.

Case Presentation: We present a 48-year-old female patient who presented with abdominal pain lasting for 1 month, with a 4-month history of constipation. A CT scan showed findings suggestive of pseudo-obstruction secondary to intestinal invagination. After an emergency laparotomy, histopathological analysis reported a Vanek tumor in the terminal ileum.

Discussion: Intestinal intussusception in adults is a rare condition, often caused by benign tumors that act as lead points. Inflammatory fibroid polyps (IFP), commonly located in the ileum, are such tumors. The symptoms are typically nonspecific, making early diagnosis challenging. In the presented case, a laparotomy was performed to address the obstruction and resect the tumor, which was histologically confirmed as an IFP. This case highlights the importance of considering these tumors in the differential diagnosis of adult intussusception and the need for timely surgical intervention.

Conclusion: IFPs should be considered in the differential diagnosis of adult intussusception, especially in cases of chronic, non-specific symptoms and intestinal obstruction.

KEYWORDS: Intestinal Obstruction, Intussusception, Inflammatory Fibroid Polyp, Vanek tumor, Laparotomy.

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INTRODUCTION

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Intestinal obstruction is the most common surgical disorder of the small intestine (1). It primarily arises from two main causes: first, and more commonly, a mechanical issue, and second, a motility abnormality of the intestine, known as functional obstruction (2).

For the purposes of this article, we will focus on mechanical obstruction, which is pathophysiologically based on the obstruction of the passage of food bolus, chyme, or fecal matter along the digestive tract due to a physical barrier. Causes include extrinsic factors (adhesions, hernias, extraintestinal neoplasms – including carcinomatosis –

endometriosis, intra-abdominal abscesses, and idiopathic sclerosis), intrinsic (intramural) diseases [congenital (malrotation, atresia, stenosis, congenital cysts, and bands), inflammatory (inflammatory bowel disease, diverticulitis, radiotherapy, tuberculosis, lymphogranuloma venereum, and schistosomiasis), neoplastic (primary small bowel cancer, obstructive colon cancer), traumatic (hematomas, stenosis from anastomoses), and other causes (intestinal invagination, volvulus, aganglionosis, ischemic injury, radiotherapy, or duodenal obstruction due to superior mesenteric artery)], and finally, causes due to intraluminal anomalies (bezoars, feces,

foreign bodies, gallstones in cholecystoenteric fistulas, and enteroliths) (1, 2).

Globally, from 1990 to 2019, the prevalence of intestinal obstruction (IO) increased by 56.91%, and the incidence by 86.67%. A higher prevalence has been reported in men compared to women, with a significantly greater incidence in economically developed countries. Age plays an important role in the incidence and mortality of IO, as older patients have a higher mortality rate, while middle-aged and younger individuals have a lower mortality (3).

As previously mentioned, one cause of mechanical obstruction is intestinal invagination, also known as

intussusception, which was first described in 1674 by Paul Barbette in Australia (4), and the term was coined by John Hunter in 1789. It primarily involves the telescoping of one segment of the intestine into an immediately adjacent segment. The intussusceptum is the proximal segment that invaginates or is introduced into the distal segment, referred to as the intussuscipiens (the receiving segment). Pathophysiologically, this begins with a lead point (anchor) that acts as a focal point of traction, drawing the proximal intestine into the lumen of the distal intestinal segment, driven by the peristalsis of that segment (5) (Fig. 1).

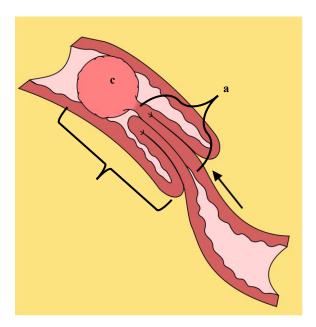


Figure 1: Representation of intestinal invagination. (a) Invaginated intestinal segment (*intussusceptum*). (b) Segment of intestine receiving the previous one (*intussuscipiens*). (c) Traction point.

Symptoms arise from edema and reduced vascularization of the affected segment, leading to subsequent obstruction, which, if untreated, progresses to necrosis and ultimately intestinal perforation (5). While more common in children under 5 years, where it is the most frequent cause of intestinal obstruction (IO), it can also occur in adults, typically being idiopathic and mediated by multiple factors (5, 6). Its incidence in this age group has been reported as 2 to 3 cases per million inhabitants (7), with benign tumors (37.4%), malignant tumors (32.9%), and idiopathic causes (15.1%) as the most common etiologies. The most frequent locations are enteric (49.5%), ileocolic (29.1%), and colonic (19.9%) (8). Clinically, adult patients with this pathology present with nonspecific symptoms, including a history of poorly localized, insidious, long-standing colicky abdominal pain, often accompanied by symptoms consistent with partial obstructions, such as nausea, vomiting, constipation, gastrointestinal bleeding, changes in bowel habits, or abdominal distension. It is important to note that the presentation of classic acute intestinal obstruction symptoms

in these patients is variable (7). On physical examination, patients may show marked abdominal distension, reduced or absent peristalsis, tympany on percussion in the mesogastrium, and tenderness to palpation in the right flank and iliac fossa. No hepatomegaly or splenomegaly is usually found. If the condition has progressed, signs of peritonitis or ischemic bowel may be present. Additionally, tachycardia or hypotension may be observed. Given the broad spectrum of clinical presentation, a wide range of differential diagnoses must be considered, and intussusception may not always be included (7).

Laboratory tests typically reveal leukocytosis and elevated acute-phase reactants such as C-reactive protein (8, 9). On plain abdominal radiographs, intestinal loop dilation and absence of gas in the colon may be observed, along with the "target sign" – although radiography lacks diagnostic value for this pathology. Ultrasound plays an important role in the differential diagnosis, particularly in ruling out an acute appendicitis process (10). Additional imaging studies, such as

CT scans, barium contrast studies, and colonoscopy, may also be useful (8).

IFPs were first described in 1949 by J. Vanek in Prague as a lesion composed of: (I) connective tissue with mesenchymal elements, (II) an inflammatory infiltrate of eosinophils and lymphocytes, and (III) vascularization from arterioles, blood vessels, and lymphatic capillaries (11). The term "fibroid polyp" was later coined by Helwig and Rainer in 1953. While this lesion can occur anywhere in the gastrointestinal tract, the most common locations are the gastric antrum and ileum (12). They account for 0.1% of all gastric polyps, and the incidence outside of the stomach is much lower. The average age of presentation is 60 years (ranging from 26 to 87 years), and the etiology of these lesions has been debated in recent years. Several hypotheses suggest that they may be part of granulomatous lesions, mesenchymal tumors (close to gastrointestinal stromal tumors), characterized by a CD34.9 cluster of differentiation, and dendritic cell origin. Genetic associations have been identified with mutations in the platelet-derived growth factor receptor alpha (PDGFR-A), and some cases have been linked to Helicobacter pylori infection (12, 13).

Given the infrequent presentation of intussusception as a nonspecific intestinal obstruction in adults, large multicenter studies are lacking, thus requiring considerable preoperative diagnostic skill.

We present the case of a 48-year-old female patient with the following relevant history: no chronic degenerative diseases, no allergies, no smoking or alcoholism, a recent diagnosis of diverticular disease without current treatment, and surgical history including a hysterectomy 4 years ago for uterine myomatosis, and laparoscopic appendectomy 6 weeks prior to admission.

Her current symptoms began 4 months ago with intermittent, colicky abdominal pain localized in the epigastrium, right

flank, and right iliac fossa, rated 8/10 on the numerical pain scale (10 being the worst), accompanied by changes in bowel movements, including alternating constipation and decreased stool consistency, without mucus or blood. Based on these symptoms, a barium contrast intestinal transit study was performed externally, showing diverticular disease of the descending colon and sigmoid and a filling defect in the ileum (32 mm), altering the typical mucosal pattern. The patient chose not to pursue further diagnostic workup for these findings.

She presented to the emergency room with exacerbation of abdominal pain, described as 10/10, initially localized in the epigastrium and later becoming generalized. Associated symptoms included nausea and vomiting (3 episodes of gastric contents), and absence of bowel movements for 48 hours. No other significant symptoms were reported. Upon admission, vital signs were within normal limits (BP: 122/80 mmHg, HR: 70 bpm, RR: 20 bpm, Temp: 36.5°C, SpO2: 95%).

Physical examination revealed a globally distended abdomen with decreased peristalsis, tympany over the mesogastrium, and tenderness to palpation in the right flank and iliac fossa, with no hepatomegaly or splenomegaly. There were no signs of peritoneal irritation.

Laboratory tests showed: Hemoglobin 14.4 g/dL, Platelets 364,000/mm³, Leukocytes 8.08/mm³, Neutrophils 65.3%, Lymphocytes 25.9%; Prothrombin time 12.2 sec, APTT 26.8 sec; Blood chemistry (Glucose 93.3 mg/dL, Urea 15.5 mg/dL, Creatinine 0.7 mg/dL), and Electrolytes (Na: 139.9 mEq/L, K: 4.2 mEq/L, Cl: 109.3 mEq/L).

Abdominal ultrasound did not show intestinal alterations, and a CT scan (simple and contrast-enhanced) revealed slight gastric distention, dilation of small bowel loops, and changes suggestive of intestinal invagination, resulting in a pseudoobstruction (Image 1).



Image 1: Computed tomography, axial cut. (a) "Intestine within intestine."

The patient underwent exploratory laparotomy, with the following findings: a 20 cm ileo-ileal intussusception located 270 cm from the Treitz angle (Figure 2). Enteropexy was performed on the invaginated segment, with no vascular

compromise; however, a palpable mass was identified (Figure 3). An enterotomy was then performed, revealing a 3 cm polypoid tumor with a 0.5 cm pedicle, which was identified as the lead point for the intussusception (Figure 4).



Image 2: Intestinal invagination; (a) Intussusceptum. (b) Intussuscipiens.



Image 3: Post-enterolysis intestinal segment; (a) Mass compatible with the etiology of the condition.

Primary closure was achieved using Connel-Mayo and Lembert stitches on the serosa. Intestinal permeability was confirmed with appropriate coloration and peristalsis.

The patient remained under observation in the general surgery ward, with stable vital signs, tolerating ambulation, and showing adequate peristalsis. Oral intake was initiated with a liquid diet and later progressed to a soft diet, without complications. The patient was discharged home on postoperative day 5 with clinical improvement, with follow-up scheduled at the general surgery outpatient clinic.



Image 4: Intestinal polyp.

The surgical specimen was sent for histopathological analysis, which reported a macroscopic appearance of a 3.1 x 3 x 2.6 cm ovoid, smooth, gray, mucoid, and soft tissue nodule. Microscopically, the sections revealed an intestinal epithelial lining with regular villi, covered by cylindrical epithelium and mature goblet cells, with no atypia and preserved polarity. A non-encapsulated lesion with regular, non-infiltrating borders was observed, composed of mature spindle cells, capillaries, abundant eosinophilic and mast cell infiltration, and defined lymphoid follicles. histopathological diagnosis was inflammatory fibroid polyp (Vanek's tumor) of the terminal ileum (Figure 5).

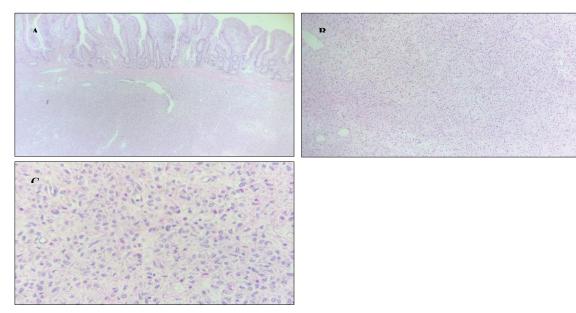


Image 5: Histological sections of the resected surgical specimen. (A) Intestinal epithelial villi. (B). (C).

DISCUSSION

Intestinal intussusception is a rare but significant cause of bowel obstruction in adults, with reported rates of 2-3 cases per million people. The most common etiology in this age group is benign tumors, which typically create a focal lesion acting as a lead point, causing one segment of the intestine to pull another segment into itself. This leads to ischemia, edema, and potential necrosis of the affected segment, which can quickly progress to perforation and peritonitis if not treated promptly.

As described in the literature, adults typically present with chronic, nonspecific symptoms such as episodes of abdominal pain, intermittent bowel obstruction, and vague gastrointestinal symptoms, which align with the presentation in our patient. This often leads to a delayed diagnosis, so it is crucial for surgeons to keep this possible etiology in mind to avoid treatment delays.

Among the various benign tumors that can act as lead points in adult intussusception is the inflammatory fibroid polyp (IFP), also known as Vanek's tumor, which is a rare gastrointestinal lesion. It can develop in different locations, with the ileum being a common site, as in the case presented. Histopathologically, IFPs are characterized by a fibromuscular stroma rich in eosinophilic infiltrates and mast cells, with capillaries and lymphoid follicles.

This case underscores the importance of considering IFPs in the differential diagnosis of adult intussusception, particularly when typical causes have been excluded. Management of intussusception can be complex; in this case, laparotomy was required to address the obstruction, identify the underlying lesion, and prevent further complications. Successful surgical resection and primary closure of the intestine, followed by histopathological confirmation of Vanek's tumor, highlights the crucial role of timely diagnosis and surgical intervention.

CONCLUSION

While IFPs are rare, they should be considered as a possible cause of intussusception in adults, particularly in patients presenting with chronic, vague symptoms and evidence of intestinal obstruction. Early recognition through imaging and histopathological analysis, followed by appropriate surgical intervention, is key to managing these cases effectively.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

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