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Intestinal Invagination in an Adult with Vanek's Tumor: A Case Report

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Abstract | **Case Report**

Introduction and Significance: Inflammatory fibroid tumors (IFT) or Vanek's tumors are rare benign neoplasms. They can affect any part of the digestive tract. They are under-diagnosed conditions, usually revealed by a life-threatening complication such as intussusception. The final diagnosis is made on the resection specimen after curative surgery. ***Case Presentation:*** A 25-year-old female patient presented with ileo-ileal intussusception revealed by an emergency CT scan. The etiology was unclear, but we suspected small bowel tumor etiology. Emergency surgery was therefore performed and the tumour was resected with margins. The diagnosis of Vanek's tumour was established on pathological examination. ***Discussion:*** Inflammatory fibroids are mesenchymal tumours without malignant potential. However, they may be revealed by a dangerous complication requiring emergency surgery. Complete resection is necessary, and anatomopathological examination enables the diagnosis to be made. ***Conclusion:*** Surgeons should include IFT among the differential diagnoses of adult ileal intussusception, as it mimics other small bowel tumours. The diagnosis can only be made on pathological examination.

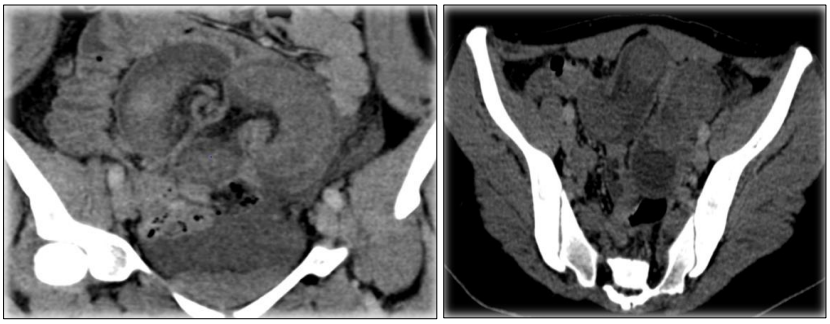
Keywords: Inflammatory Fibroid Tumor Vanek's Tumor, Small Intestine Intussusception.

1. INTRODUCTION

Inflammatory fibroid tumors (IFTs) are rare conditions, usually single, benign, polyp-like neoplasms that can affect any part of the gastrointestinal tract, most in the gastric antrum and ileum. The underlying cause is still controversial [1]. Preoperative diagnosis is difficult. In complicated cases such occlusion or intussusception, an inflammatory fibroid tumour may be discovered incidentally during postoperative pathological examination [2]. We present here the case of a young woman presented with an acute abdomen related to intussusception caused by an inflammatory fibroid tumor. Complete resection by emergency laparotomy was performed, and the diagnosis was established on pathological examination.

2. CASE PRESENTATION

A 25-year-old woman with no previous surgical history had been complaining of abdominal pain for a week. The patient was admitted to the emergency department for diffuse abdominal pain, with no defensiveness or palpable mass, evolving for 2 days. On physical examination, the patient was apyretic and hemodynamically and respiratorily stable. Biological tests revealed no remarkable abnormalities. An emergency abdominal CT scan showed intussusception of the small intestine (Fig. 1).



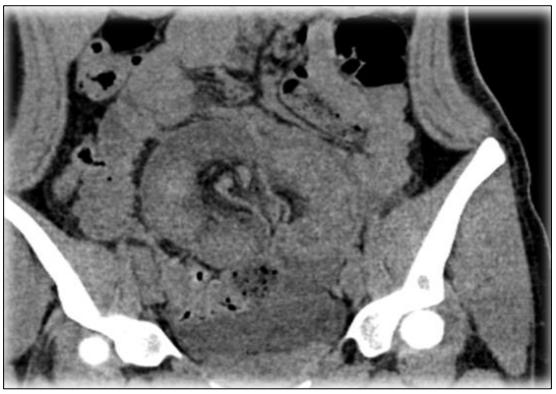


Figure 1: Abdominal CT scan in axial and coronal sections without and with gadolinium injection

Image of ileo-ileal invagination of the hypogastric and peri-umbilical region, without upstream digestive distension. Doubtful parietal pseudo-nodular contrast-enhancing formation.

Signs of digestive distress revealed by the spontaneously hyperdense ileal parietal appearance and peritoneal effusion ()

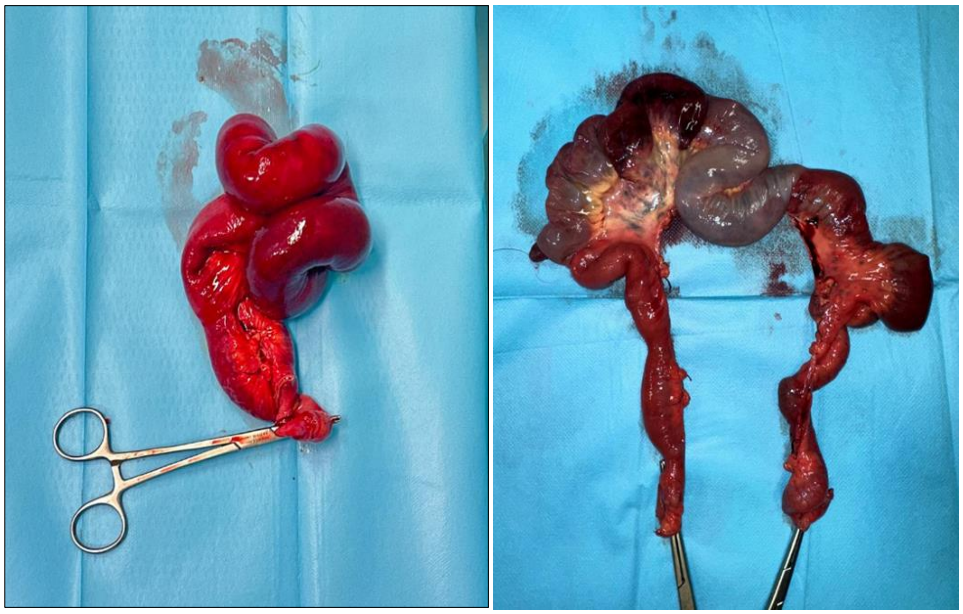


Figure 2

The invaginated part of the small bowel was resected with a safety margin on either side of the lesion, with mechanical lateral anastomosis (Fig. 3). The postoperative course uneventful.

Pathological examination revealed a submucosal nodule in the bowel wall, measuring 2.5x1.5 cm, corresponding to a dense, pauci-cellular fibrosis of thick collagen clusters. This is consistent with a reactive nodular fibrous pseudotumor. The proximal and distal resection margins pass into viable care tissue. No histological evidence of malignancy.

3. DISCUSSION

Inflammatory fibroid tumors are rare benign tumors described by Vanek in 1949 [4], as a "submucosal granuloma with eosinophilic infiltration". They can affect any part of the gastrointestinal tract. The most common site is the gastric antrum (66%-75%), followed by small intestine (18%-20%), colorectal region (4%-7%), gallbladder (1%), oesophagus (1%), duodenum (1%) and appendix (< 1%) [5, 6]. The etiology of TFI is unknown, and many theories have been proposed [1-7]. TFI remains asymptomatic for a long time. Symptoms are non-specific and depend on size, location and complications [6]. More than half of patients with TFI of the small intestine had intussusception [6]. Intussusception is a complication seen mainly in children, but is rare in adults. The etiology and clinical presentation differ between children and adults. Diagnosis is generally difficult. Biological workup and PSA are not helpful in suspecting the diagnosis, as lesions are only found in cases of intestinal obstruction with non-specific features [8, 9]. CT is considered the most sensitive imaging modality for intussusception [10]. Consequently, CT is mandatory in cases of emergency presentation such as intussusception. It

provides sufficient preoperative assessment of the obstructive lesion and may suggest the presence of vascular involvement and signs of severity [11], as in our case. However, preoperative diagnosis of small bowel TFI is difficult, particularly when the size of the lesion is less than three centimetres [1]. Abdominal ultrasound is known to represent an accurate tool diagnosing intussusception. It shows a "target sign" on the axial view and a "trident" image on the longitudinal sections [12]. In addition, a polypoid lesion may also be observed [10]. In cases of chronic colonic pain, investigations may include endoscopy or colonoscopy for gastric and colonic lesions. For small bowel lesions, double balloon endoscopy is useful for assessing the polyp [1-6]. However, endoscopic resection may miss a malignant tumour [2]. MRI can contribute to the radiological diagnosis of intussusception, but cannot clearly determine the etiology in cases of TFI [1, 2]. Emergency surgery is usually required in the event of a complication, as in our case. Laparoscopy is not always possible in cases of small bowel distension Small bowel loops are not wide enough, which does not allow sufficient exploration of the abdominal cavity. However, Saeed *et al*., [9], and Guerci *et al*., [13], described two cases where laparoscopic-assisted small bowel resection was successfully performed for intussusception caused by inflammatory fibroid tumours. In some cases, laparoscopy combined with enteroscopy can be a useful tool, particularly when the tumour is small [14]. Complete resection is the gold standard and the only possible route to a final diagnosis. On pathological examination, IFTs are usually single, macroscopically polypoid or sessile, measuring< 5 cm [15]. They stain positively for CD34 and vimentin, and occasionally for smooth muscle actin, calponin, CD35 and cyclin D1. Unlike mesenchymal tumors, TFIs do not stain for CD 117 and S100 [16]. After complete resection, TFIs generally do not recur, and the lesion appears to be devoid of malignant potential [1].

4. CONCLUSION

Inflammatory fibroid tumours (IFT) or Vanek's tumours of the small intestine are an under-diagnosed condition. The difficulty lies in the existence of several differential diagnoses. It requires pathological examination of the resected specimen, the only means of definitively establishing the diagnosis. In our case, a life-threatening complication led to emergency surgery, fortuitously revealing an uncommon benign tumour of the digestive tract.

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