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CASE REPORT

Rare Ileal Inflammatory Fibroid Polyp Causing Intussusception: A Case Report

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ABSTRACT

An inflammatory fibroid polyp (IFP) is a rare, benign tumor most commonly presenting in the gastric antrum (60–70%), followed by the intestines (18–20%). We present a case of intussusception due to an IFP. A middle-aged female was admitted with right iliac fossa pain and abdominal distention. A CT of the abdomen showed a 5 cm smooth, oval, fluid-like (24 HU), simple hypoattenuating lesion at the terminal ileum complicated with intussusception. An open laparotomy and end-to-end anastomosis were performed. The tumor was resected. Histopathological results showed an IFP. IFP can be identified on CT abdomen as oval, hypoattenuating, fluid like lesion, However, it is not specific and other differential diagnoses of bowel lesions should be considered. Histopathologically, it is rare that this lesion extends into the muscularis propria and subserosa.

Keywords: *Inflammatory fibroid polyp, Intussusception, CT abdomen*

INTRODUCTION

Intussusception is a condition whereby a proximal bowel slides into the distal portion of the bowel, thus blocking food particles or fluid from passing through the bowel. Intussusception can also cuts off blood supply to the affected bowel loop. Intussusception is rare in adults, affecting 1 in 1,000,000 patients per year worldwide. About 90% of those who had intussusception have an underlying lesion that caused it, where half of the lesions were malignant (Guerci et al., 2022). Benign causes of intussusception are gastrointestinal stromal

tumours (GISTs), polyps, Meckel's diverticulum, inflammatory lesions and trauma (Choi et al., 2004).

Inflammatory fibroid polyp (IFP) is a rare, benign tumor that most commonly presents in the gastric antrum (66 to 75%), followed by the small intestine (18 to 20%) (Akbulut, 2012) and the large intestine and rectum (4 to 7%) (Sugawara et al., 2018). The tumor is present in those aged 4 to 80 years old (Akbulut, 2012; Garmpis et al., 2021). Rarely it happens in the gall bladder, esophagus, duodenum, or appendix (Akbulut, 2012). Females have a slightly higher chance of getting an IFP with a female-to-male ratio of 1.3:1 (Garmpis et al., 2021).

IFPs usually do not cause symptoms and are found incidentally during endoscopic procedures or laparotomy. In the small bowel, it can cause intussusception or intestinal obstruction (Akbulut, 2012). The size of an IFP is usually 2 to 5 cm, but some may reach up to 20 cm. They originate from the submucosa and grow into the intestinal lumen. IFPs originate from the mesenchymal cells of the intestinal tract (Akbulut, 2012). Initially, the presence of eosinophils within the lesion suggests it is of inflammatory origin, thus, the name "inflammatory fibroid polyp" is conceptualized. However, later studies showed mutations in platelet-derived growth factor receptor alpha (PDGFRA), causing its development to be in favor of a neoplastic origin (Rais et al., 2017). In terms of immunohistochemistry, an IFP stains positive for CD 34 and vimentin and stains negative for CD 117, S100, and DOG-1 (Akbulut, 2012).

Due to the rarity of this tumor, epidemiological data has been sparse. Since the attenuation of IFP is similar to surrounding intestinal mucosa, it is often missed during CT scans as a potential lead point for intussusception. Besides, small size of the polyp (< 1cm) and collapsed bowel also makes the detection of the polyp difficult on CT. We present a case report of a 51-year-old female who presented with IFP terminal ileum intussusception. We also performed a literature review on its imaging features.

CASE PRESENTATION

A 51-year-old female presented with right iliac fossa pain for one week, which was colicky and non-radiating in nature, with a pain score of 6. She had abdominal distention for 2 days, vomiting for one day, and loose stools for one week. She did not have a fever, and her vital signs were stable. Blood pressure was recorded to be 130/83 mmHg with a pulse rate of 93/min and normal oxygen saturation on room air.

On admission, full blood count showed a hemoglobin level of 10.9 g/dL, a white cell count of 10.1x 103/uL, and a platelet count of 609 x 103/uL. -rayA peripheral blood film (PBF) was performed because the patient was mildly anemic. The PBF showed mild normocytic normochromic anemia with thrombocytosis.

An abdominal radiograph (Figure 1) showed small bowel dilatation. Next, a contrast-

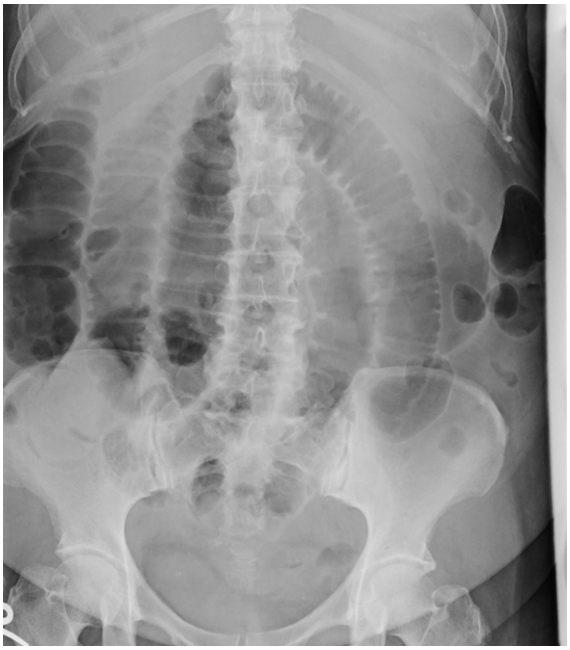

Figure 1: Abdominal radiograph showed small bowel dilatation.

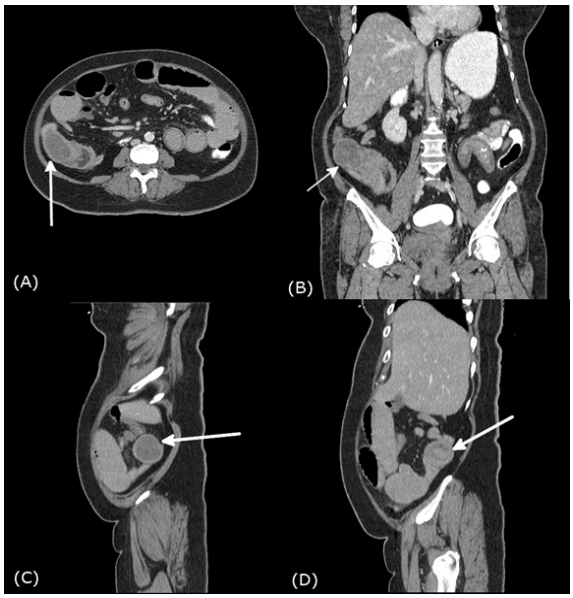

Figure 2: CECT abdomen pelvis in (A) axial, (B) coronal, (C and D) sagittal views. Figures 2A, 2B, and 2C showed the position of the cross-sections of the inflammatory fibroid polyp at the terminal ileum. Figure 2D showed the target appearance of the bowel, in keeping with intussusception.

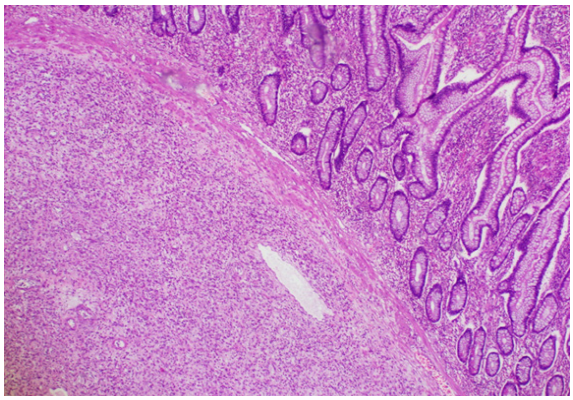

Figure 3: Haematoxylin & Eosin stain, 100x magnification. Submucosal lesion composed of a proliferation of haphazardly arranged short spindled and stellate cells. Small and intermediate-size blood vessels are seen within the lesion.

enhanced CT of the abdomen was performed to find the cause of the bowel obstruction. Both oral and rectal positive contrasts were administered. The abdominal CT (Figure 2) showed a dilated small bowel from the distal jejunum to the distal ileum. The terminal

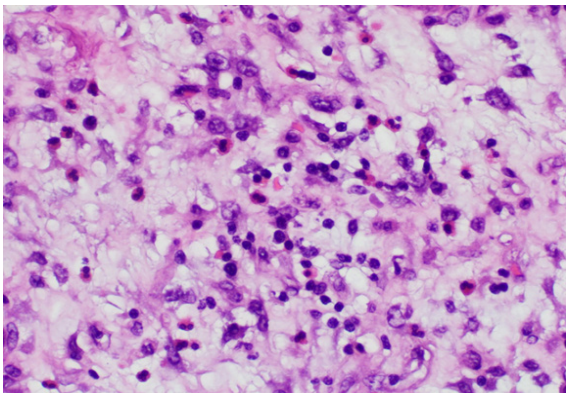

Figure 4: Haematoxylin &Eosin stain, 400x magnification. IFP cells have fine chromatin, indistinct nucleoli, and scant eosinophilic cytoplasm. The stroma is edematous to myxoid with prominent eosinophils, lymphocytes, and plasma cells.

ileum was also distended. The ascending and transverse colon colon werecollapsed while the descending colon was partially distended with contrast. A well-defined, oval, hypodense lesion measuring 4.7 x 3.0 x 4.1 cm (AP x W x CC) with mucosal hyperenhancement was seen at the terminal ileum. The lesion showed hypoattenuation, measuring +24 Hounsefield Units (HUs).

An exploratory laparotomy was performed. Intra-operatively, the proximal small bowel was dilated. An ileal intussusception was found 50 cm from the ileocecal valve. The small bowel collapsed distal to the intussusception. The intussusception was reduced, and a polypoidal tumor, sized at 5 x 3 x 3 cm (AP x W x CC), was found. The tumor was resected, and a primary end-to-end anastomosis was performed. The postoperative recovery was uneventful. The patient was discharged on day 5 post-operation. The resected tumor was sent for histopathological examination.

Gross pathology showed a hemorrhagic and tan-cut surface. Microscopy examination shows a poorly marginated, hypocellular lesion in the submucosa, extending into the muscularis propria and subserosa. It contained spindle-shaped cells and stellate cells with fine chromatin, (Figure 3) indistinct nucleoli, and

scant eosinophilic cytoplasm. Additionally, the stroma of the tumor was myxoid to edematous in appearance, containing eosinophils, lymphocytes, and plasma cells (Figure 4). There are also some small to intermediate-sized blood vessels with concentric fibrosis. Immunohistochemistry stains were positive for CD 34 and had multifocal positivity for SMA. It tested negative for CKAE1/AE3, CD 117, DOG-1, S100, ALK 1, and Desmin.

DISCUSSION

CT scan has a 58 to 100% accuracy in diagnosing intussusception (Akbulut, 2012). Small bowel tumors, whether benign or malignant, account for no more than 2% of all gastrointestinal tumors (Gill et al., 2001). Ivanis N et al. reported 77 cases of IFP in the English literature from 1976 to 2019 (Ivanis et al., 2020). In all the IFPs that were imaged by CT, only 46–50% of the cases could a lead point be found (Han et al., 2015; Lee & Yuen, 2014). However, CT scan is the standard diagnostic tool not only for polyp, but also for any lead point in the abdomen, whether intussusception is present or not because of its wide availability and fast diagnosis

Two research articles systematically explored the imaging features of IFP on CT. Han G et al. characterized the imaging features of IFP based on size, shape, margins, contour, growth patterns, mucosal hyperenhancement, homogeneity, and degree of enhancement in a case series of 27 patients from a single institution. The majority of IFP lesions appear as round or oval, hypoattenuating, smooth masses (Han et al., 2015). Mucosal ulceration is associated with hyperenhancement of the mucosa on a CT scan (Han et al., 2015). Meanwhile, Lee CY et al. performed a literature search from 1990 to 2013, focusing on the size, shape, margin, and attenuation of the lesion with HU values, if available. They showed similar results as Han G et al. Meanwhile, the HUs for IFP for these cases range from 16 to 17 (Lee & Yuen, 2014). Our case agreed closely with these two articles on the general imaging features of IFP. The density IFP in our case was fluid-like with an attenuation of 24 HUs. However, other gastrointestinal tumours may show similar features on CT scans such as gastrointestinal stromal tumour (GIST), plexiform myxofibroma, and schwannoma, thus should be included as differential diagnoses for IFP (Mocellin, 2021). In addition, our sample showed a hemorrhagic cut surface, which may be associated with the small and intermediate sized blood vessels found in histopathological study as shown in Figure 3. Proliferation of blood vessels in tumours are commonly due to neovascularization. When brittle blood vessels ruptured, they may contribute to chronic blood loss in the patient, causingmild anemia.

Other authors came across the imaging features of IFP on endoscopic ultrasound, abdominal ultrasound and MRI, On endoscopic ultrasound, IFP is presented as a homogenous, hypoechoic lesion (Inayat et al., 2018)..On transabdominal ultrasound, IFP is described as round, hypopechoic soft tissue mass (Akbulut, 2012; Bhutia et al., 2016). In MRI abdomen, IFP is described as homogenously T1-weighted isointense soft tissue mass when compared to surrounding muscles; central hypointensity with peripheral ring on T2-weighted sequence. While on DWI sequence, IFP resembles a “target” sign (Feldis et al., 2015). However, endoscopic ultrasound can only reach until duodenum; Bowel lesion is not normally seen on transabdominal ultrasound unless the bowels are distended and fluid filled. Meanwhile, MRI has limited availability and time consuming. Therefore, CT scan is still the best modality in finding lead point or small bowel lesion in emergency setting.

In this case, the IFP stroma is oedematous to myxoid based on histopatholoical study. The infiltration of the tumor into the muscularis propria and subserosa, which is found in our case, is a rare feature of IFP. However, it is impossible to differentiate between the IFP

subtype and the degree of mucosal invasion on CT scan.

CONCLUSION

CT scan is the most practical modality for the diagnosis of IFP in case of intussusception. The CT of the abdomen shows a smooth, oval, fluid-like, hypoattenuating lesion. However, such features are not specific and other types of bowel lesions should also be considered. It is also rare that such lesions can extend into the muscularis propria and subserosa on histopathological study

CONFLICT INTEREST

No funding was received for the preparation of this article. No financial competing interests declared.

CONSENTS

Written consent was obtained from the patient to publish this case report. A copy of the written consent is available for review by the Chief Editor.

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