Surgical and Experimental Pathology

CASE REPORT

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Inflammatory fibroid polyp– a rare aetiology for adult ileocolic intussusception

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Abstract

We report a case of an adult ileocolic intussusception secondary to an inflammatory fibroid polyp (IFP), a rare and unexpected association between a surgical condition, and an underlying histopathological process. Intussusception is an uncommon surgical diagnosis in adults, and an IFP is rare mesenchymal tumour. Surgical intervention of the intussusception in an adult patient, given the uncertain aetiology of the lead point, is recommended as gold standard.

Keywords Intussusception, Surgical pathology, Inflammatory fibroid polyp

Background

This case reports a rare case of adult ileocolic intussusception secondary to an inflammatory fibroid polyp (IFP). Intussusception is an uncommon adult surgical condition, representing 5–16% of all cases of intussusception, and only 1–5% of all causes of adult intestinal obstruction (Azar and Berger 1997). Up to 90% of cases are secondary to an underlying pathology, with approximately 65% being secondary to benign of malignant neoplasms (Poudel et al. 2022). The diagnostic uncertainty of the underlying lead point highlights the need to surgically intervene on these lesions promptly.

The inflammatory fibroid polyp, which is also known as the Vanek tumour, was first described by Vanek in 1949, and then proposed by Helwig and Ranier in 1953 (Poudel et al. 2022). It is now defined as a benign tumour which arises from the gastrointestinal tract and derives from the submucosa. The most common site from which IFPs arise is the gastric antrum is 66–75% of cases, followed by the small bowel in 18–20% of cases (Poudel et al. 2022). The ileal segment, however, is the most common site where they occur. Historically, aetiology was thought to be an inflammatory response to a submucosal granuloma usually associated with an irritating stimulus such as helicobacter pylori, trauma, tuberculosis, etc. However, activating mutations of the proto-oncogene platelet derived growth factor alpha (PDGFRA) gene have been recently as a potential underlying true neoplastic origin (El-Sergany et al. 2015).

The appropriate surgical management of adult intussusception remains debatable when deciding on a primary en bloc resection versus an initial reduction followed by a limited resection. Here we report a case of a laparoscopic ileocolic resection.

Case presentation

A 54-year-old female presented to the emergency department with abdominal pain, distention, vomiting, having had an outpatient colonoscopy 5 days prior identifying a medium-sized caecal mass. This was on a background of worsening generalised abdominal pain, 8 kg unintentional weight loss and night sweats over several months. There was no surgical history. The abdomen was soft and mildly distended, with a palpable right lower quadrant

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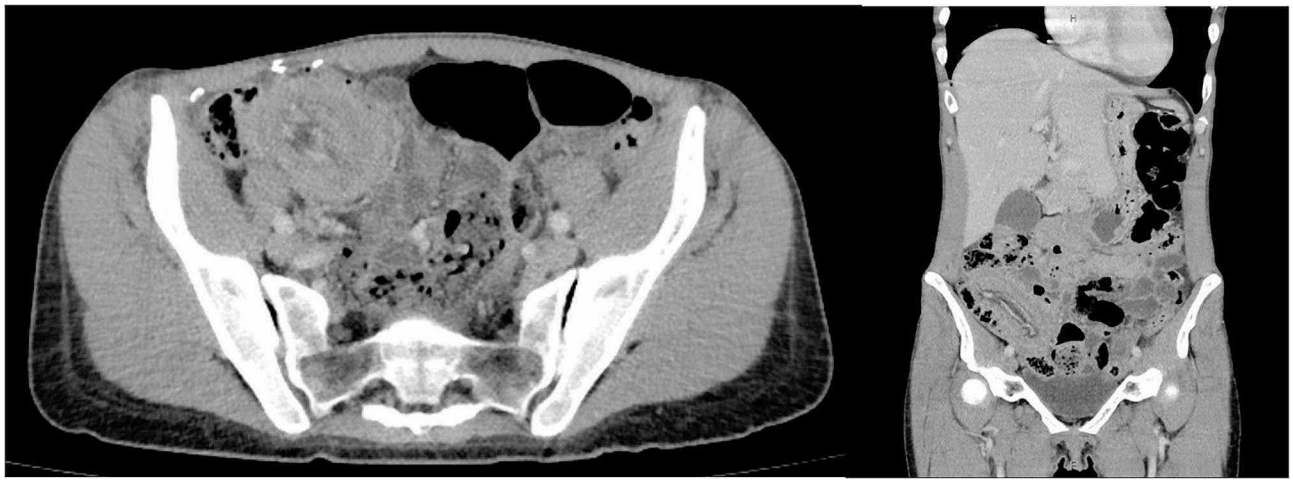

Fig. 1 CT abdomen pelvis (axial and coronal views) demonstrating Ileocolic intussusception with mild dilatation of small bowel loops

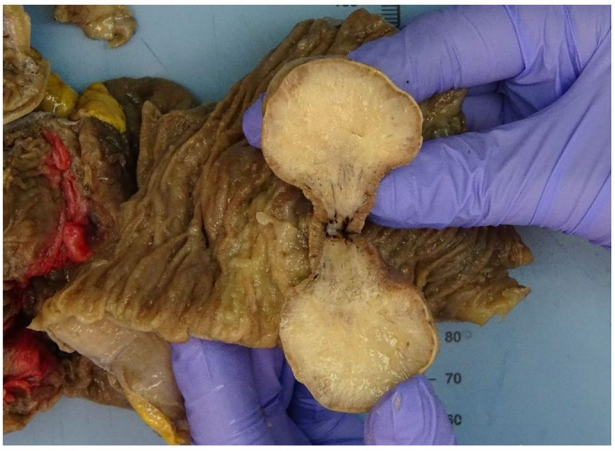

Fig. 2 Polypoidal mass in the ileum measuring 43 × 25 × 30 mm

mass. A computed tomography revealed an ileocolic intussusception (Fig. 1). The patient underwent a laparoscopic ileocolic resection, finding an ileocolic intussusception secondary to a polypoid mass in the terminal ileum 20 cm from the ileocecal valve. The patient recovered well without complications.

The outpatient colonoscopy biopsies were inconclusive. The histopathology of the resected mass revealed an inflammatory fibroid polyp (IFP), limited to the mucosa and submucosa (Fig. 2). It demonstrated a typical proliferation of spindle shaped cells, with concentric peri-vascular arrangement ("onion skinning") dispersed amongst a mixed inflammatory cell infiltrate rich in eosinophils (Fig. 3A-C). The spindle cells demonstrated positive staining for vimentin and negative for CD34, Ckit, beta-catenin, Bcl-2, ALK1, SMA and desmin (Fig. 3D). The IFP was diagnosed as the lead point for the ileocolic intussusception.

Discussion and conclusions

Intussusception, while common in children, is a rare surgical condition in adults accounting for 1–5% of intestinal obstructions(Azar and Berger 1997). The aetiology in adults is an identifiable pathology in 90% of cases, whereas it is mostly idiopathic in children (Poudel et al. 2022). Given the risk of underlying neoplasia, resection with oncological surgical principles is advised. However, there is controversy regarding the need for intra-operative reduction versus en-bloc resection without reduction. The former can potentially limit the extent of resection, but other studies concur reduction can promote peritoneal or venous dissemination of tumour cells and bowel perforation (El-Sergany et al. 2015). In this case, intra-operative reduction proved challenging, and given the known presence of an ileocecal mass, an en-bloc resection was preferred. Laparoscopic management, as performed in this case, has been only occasionally reported in the literature.

IFP's are rare and benign mesenchymal tumours arising from the submucosa of the gastrointestinal tract. A total of 119 published cases have been published in the literature (Kao and Chen 2020; Akbulut 2012).. Activating mutations of proto-oncogene platelet derived growth factor alpha (PDGFRA) gene has been confirmed as the underlying neoplastic origin (Schildhaus et al. 2008). Invasive growth or metastatic disease has not been reported.

As these lesions are intramural, mucosal biopsies during endoscopic are often non-diagnostic, as occurred in the current case. Histochemical analysis allowed differentiation from other spindle cell lesions namely GIST, leiomyomas and inflammatory myofibroblastic tumour. GISTs are positive for CD117 and CKIT, leiomyomas are positive for SMA and Desmin, and inflammatory fibroblastic tumour show positive expression for ALK (Liu et al. 2013). IFPs are typically positive for vimentin and do

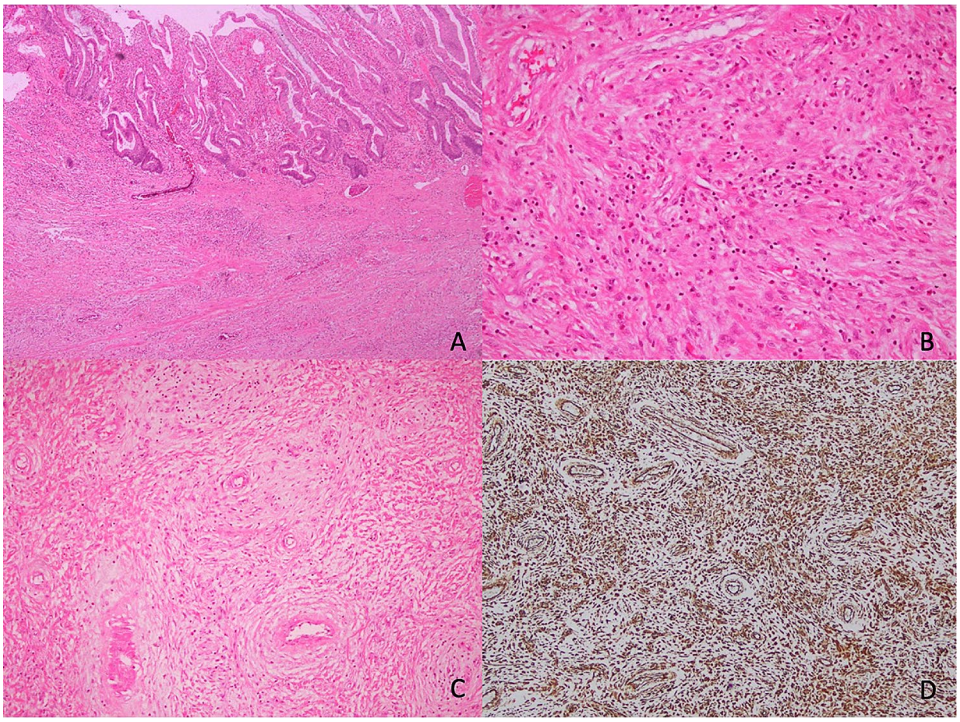

Fig. 3 (A) Haematoxylin and Eosin stain, 100x magnification. The lesion involves the submucosa and lamina propria of the bowel wall. (B) Haematoxylin and Eosin stain, 200x magnification. The spindle shaped lesional cells with inflammatory cell infiltrate rich in eosinophils. (C) The spindle shaped lesional cells concentrically arranged around the blood vessels imparting an onion skin appearance. (D) Vimentin, 200x magnification. The lesional cells exhibit positive staining for vimentin

not demonstrate positivity for ALK. Thus, the features and immunoprofile is consistent with an IFP.

We report a rare case of an adult ileocolic intussusception secondary to an IFP. Given the uncertain aetiology of the lead point, and to avoid complications, surgical intervention of adult intussusception is recommended. Future research should focus on elucidating the optimal surgical approach.

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Author contributions
DTT– writing– original draft; investigation. WS– writing– review and editing; investigation. KB– writing– review and editing; supervision.

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Data availability
Data sharing is not applicable to this article as no datasets were generated or analysed during the current study.

Declarations

Ethics approval and consent to participate
This is a case report that does not require a formal ethical committee approval in the Northern Territory of Australia.

Consent for publication
Formal consent was obtained from the patient prior to submission. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Competing interests
The authors declare that they have no competing interests.

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