

Inflammatory Fibroid Polyp: A Rare Cause of Ileo-ileal Intussusception in Adolescents—Case Report and Review of Literature

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Abstract

Inflammatory fibroid polyp (IFP) of the gastrointestinal tract (GIT) is a rare benign polypoidal mesenchymal tumor. It most often affects middle-aged adults with a variable clinical presentation. The most common site is the stomach, followed by the small intestine. We report a case of a 16-year-old male who presented with the classical features of intussusception. Surgical resection of the ileocecal region revealed a pedunculated polypoidal mass. Histopathological examination showed a submucosal tumor with an onion-skin appearance comprising spindle cells, loose stroma, and mixed inflammatory infiltrate, predominantly eosinophils. IFP should be considered in the differential diagnosis even in pediatric patients with intestinal obstruction because of its bearing on management.

Keywords: Inflammatory fibroid polyp; Vanek's tumor; Gastrointestinal; Intussusception; Polyp

Introduction

Intussusception is the invagination of a proximal portion of the intestine (intussusceptum) into an immediately distal segment (intussusciens) [1]. It can occur at any age but is more common among children. Intussusception in adults differs from that in children in various aspects, including etiology, clinical manifestations, and prognosis [2].

It is usually idiopathic in pediatric patients but almost always pathological, with a lead point, in adults. Pediatric intussusception is often acute with sudden intermittent colicky pain, vomiting, bloody mucoid stools, and the presence of a palpable mass. In contrast, adults may present with acute, subacute, or chronic nonspecific symptoms [3].

Inflammatory fibroid polyp (IFP) is a rare benign tumor-like proliferating lesion that arises from the submucosa of the gastrointestinal tract (GIT). In 1949, Vaněk first described the lesion as a 'submucosal granuloma with eosinophilic infiltration' [4].

IFP can be found throughout the gastrointestinal (GI) tract but is most commonly found in the stomach. Peak incidence occurs in the sixth and seventh decades of life, with a slight male predominance. The majority of cases are asymptomatic

and discovered incidentally. However, clinical manifestations of IFP depend largely on size and location and mostly include intestinal obstruction, abdominal pain, intussusception, and rarely GI bleeding. The small bowel is the second most common site of origin, where IFPs usually present as intussusception or obstruction [5].

Grossly, IFP mimics a pedunculated or sessile lesion, arising from the submucosa and projecting into the lumen, causing ulceration of the overlying mucosa. Characteristic microscopy reveals an onion-skin appearance comprising fibroblast-like spindle cells arranged concentrically around the vessels and mixed inflammatory infiltrate, predominantly eosinophils [6].

We herein present a rare case of ileoileal intussusception due to an inflammatory fibroid polyp in a 16-year-old male patient.

Case Report

A 16-year-old male patient presented to the emergency department with abdominal pain, vomiting, and nausea. Owing to clinical suspicion of intussusception, an exploratory laparotomy was performed, followed by resection of the ileocecal region. The specimen was sent for histopathological examination. Grossly, an intestinal segment measuring 20 cm in length with one cut end (ileal) measuring 4 cm and the other end blind (cecum) was received. The attached appendix measured 8 x 0.5 cm. A pedunculated polyp was identified measuring 7 x 4.5 x 2.5 cm with a stalk measuring 4.5 cm in length (Figure 1). The cut surface of the polyp was gray-white to gray-yellow, homogenous, and firm in consistency (Figure 2). Multiple papillary projections were identified on the ileal mucosa just near the ileal margin. A perforation was identified 8.5 cm away from the ileal margin. Two mesenteric lymph nodes measuring 1.3 cm and 1.5 cm were also identified.

The microsections studied from the polyp show a tumor with ulceration on the surface and the presence of acute inflammatory exudate. The tumor is composed of bland, ovoid, short, spindled, and stellate cells with fine chromatin, inconspicuous nucleoli, and a small amount of eosinophilic cytoplasm. The spindle cells are arranged haphazardly in a loose edematous to myxoid stroma showing inflammatory infiltrate composed predominantly of eosinophils and lymphocytes, plasma cells, capillaries, and small blood vessels (Figures 3 and 4).

Sections studied from the ileal margin show ulceration of the mucosal epithelium with the presence of acute inflammatory exudate. The mucosa and serosa show congested vessels along with infiltration of eosinophils, lymphocytes, and plasma cells.

Sections studied from papillary projections in other areas of the mucosa also show ulceration of the mucosal epithelium and the presence of acute inflammatory exudate. Papillary projections seen grossly turned out to be an associated inflammatory change on histology. The mucosa shows congestion. Mucosa and serosa show infiltration of polymorphs, lymphocytes, plasma cells, and eosinophils. Sections studied from the appendix show histological features of acute appendicitis. Sections studied from the two lymph nodes isolated show features of reactive lymphadenitis. Sections studied from the margins of perforation show ulceration and necrosis of the mucosa, submucosa, and muscularis propria. The serosa shows congested blood vessels and infiltration of polymorphs and lymphocytes. On the basis of this histomorphology, a final diagnosis of inflammatory fibroid polyp was given.



Figure 1: Gross specimen of ileocecal gut segment showing a polyp measuring 7x4.5x2.5 cm.

Discussion

Inflammatory fibroid polyp (IFP) represents a rare cause of a polypoid condition of the gastrointestinal (GI) tract in childhood. They tend to affect patients in the 5th and 6th decades of life. IFP has been described in the literature by a variety of names, such as “Vanek’s tumor,” eosinophilic or submucosal granuloma, granuloblastoma or gastric fibroma with eosinophilic infiltration, inflammatory pseudotumor, inflammatory fibroid tumor, and hemangiopericytoma [7].

IFP is usually a solitary polypoid benign tumor that can affect any part of the gastrointestinal (GI) tract, with the stomach being the most common site. It is characterized by submucosal fibrous connective tissue growth with spindle-shaped cells in a hypervascular stroma infiltrated by nonspecific inflammatory cells, especially eosinophils [8].



Figure 2: Cut surface of polyp reveals a homogenous gray-white to gray-yellow appearance.

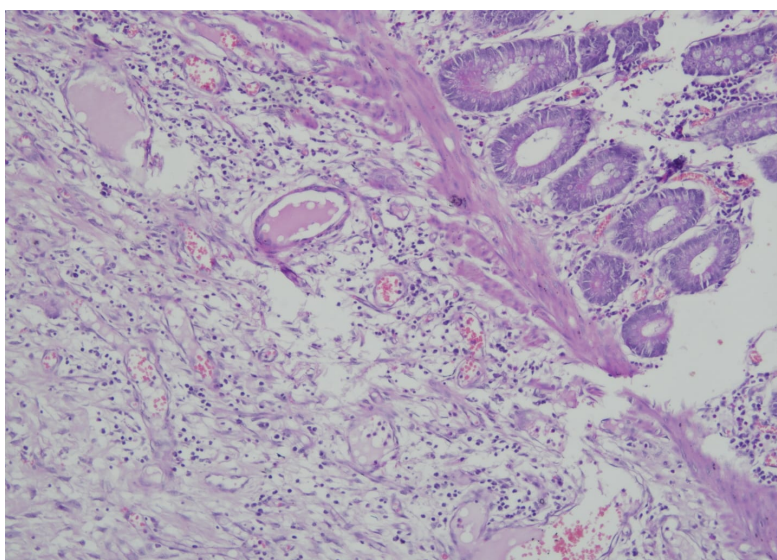


Figure 3: Photomicrograph showing a submucosal tumor consisting of spindle cells, inflammatory cells, and small vessels (H and E stain; 100x).

The origin of the mesenchymal spindle-shaped cells that comprise the polyp remains enigmatic. Recent studies have shown familial predisposition and oncogenic PDGFRA (platelet-derived growth factor receptor) mutations in many cases, suggesting their neoplastic nature. Similar mutations in the PDGFRA gene have been found in gastrointestinal stromal tumors (GISTs), which imply a common oncogenetic pathway [9].

The most common site of IFP in the GIT in adults is the gastric antrum (66%-75%), followed by the small bowel, colorectal region, gallbladder, esophagus, duodenum, and appendix. However, the ileum is the most common site responsible for intussusception, especially in children, similar to our case [10].

A myriad of clinical features may be associated with IFP depending on their size and location. The most common symptoms of IFP include colicky abdominal pain, nausea, vomiting, constipation, and distension, as seen in the present case. IFP can be asymptomatic and detected incidentally on endoscopy or abdominal imaging [11].

These polyps are extremely difficult to diagnose preoperatively due to nonspecific imaging features. Even through endoscopy, biopsy may be difficult as these lesions may arise from the submucosa with surface erosion and granulation. CT is considered an invaluable imaging modality for the evaluation of the site, size, and complications of IFP, such as intussusception. However, an accurate diagnosis of IFP is only possible with histopathological examination [7, 11].

Macroscopically, these tumors may be pedunculated or sessile, measure 0.2-20 cm in diameter, arise from the submucosa, and project into the bowel lumen. The mucosal surface is usually ulcerated and pale [3].

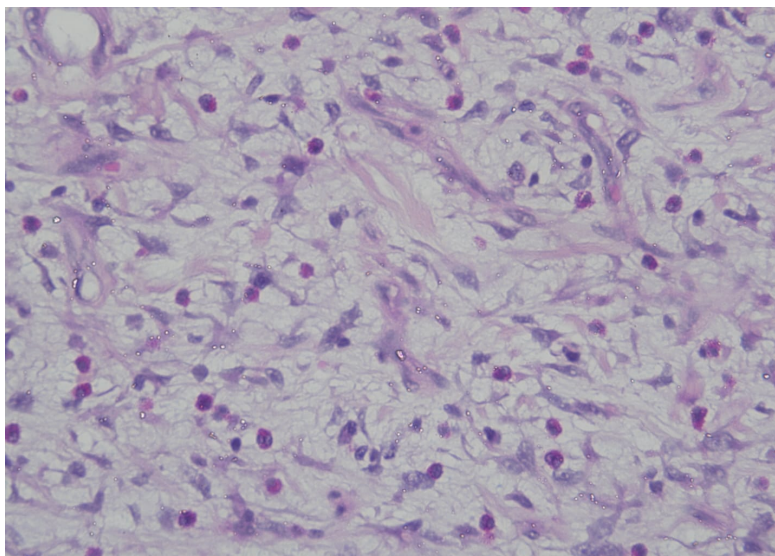


Figure 4: Photomicrograph showing bland spindle cells, small vessels, and prominent eosinophils (H and E stain; 400x).

Histologically, IFP is composed of mononuclear spindle-shaped cells arranged in fascicles, giving an onion-skin appearance with vascular proliferation and mixed inflammatory infiltrate predominantly comprising eosinophils [12]. IFP has been categorized into four histopathological groups: classical fibrovascular, nodular, sclerotic, and edematous. A fifth category of IFP with nuclear pleomorphism has been described but is rather atypical [13].

The histopathological differential diagnoses of IFP include gastrointestinal stromal tumor (GIST), inflammatory myofibroblastic tumor, leiomyoma, and neurofibroma. A comprehensive immunohistochemistry (IHC) panel is used to assist in diagnosis, wherein IFP are generally reactive for CD34, for vimentin, and occasionally for SMA and fascin. The cells of IFP are nonreactive for CD117, S100, desmin, and DOG1. In contrast, GISTs show characteristically positive CD117 and CD34 immunostaining. However, this distinction is not definitive. The identification of similar PDGFRA mutation implies a common oncogenetic pathway with GISTs [11, 14].

Although IFPs are biologically regarded as benign and noninvasive lesions, a few cases have been reported to invade the muscularis propria layer. This implies that IFPs may behave as locally aggressive neoplasms with infiltrative growth patterns and exhibit local recurrence after inadequate resection [15].

Surgical or endoscopic excision is the treatment of choice for symptomatic cases. In the majority of reported cases, surgical excision alone was curative, and no reports exist of a malignant transformation. Because of the paucity of data in the literature, a cautious approach with periodic clinical surveillance of the affected is advised, with follow-up endoscopy reserved only for a few cases [7, 11]. Limitations: IHC was not performed due to resource constraints, and thus, mimics cannot be definitively excluded.

Conclusion

Inflammatory fibroid polyp is a rare benign submucosal tumor of idiopathic origin. IFPs should be considered in the differentials of intestinal obstruction due to intussusception, even in children. An immunohistochemical staining panel comprising C-KIT, CD34, SMA, and S-100 must be applied for differentiating IFPs from other mesenchymal tumors, particularly GISTs. However, IHC was not applied in our case. A CT scan should also be considered in the assessment of such patients.

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Competing Interests None declared.

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