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CASE REPORT

Multiple Inflammatory Fibroid Polyps in a Female; A case Report

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ABSTRACT

The inflammatory fibroid polyp (IFP) usually presents as a solitary benign polyp that arises from the submucosa of the gastrointestinal tract. It occurs more commonly in males and stomach is the favored site followed by ileum, colon and duodenum. Multiple inflammatory fibroid polyps (IFPs) is a rare occurrence, so we thought of presenting a case of a 40 years old female with multiple inflammatory fibroid polyps (IFPs). She presented with abdominal pain and abdominal fullness. Her C.T. abdomen showed multiple lobulated soft tissue masses causing intussusception. Surgical resection was done, and histopathological examination of these polyps showed features of IFPs. A literature review of 9 cases (including ours) between 2000-2022 is also presented. To our knowledge this is the third female patient with multiple IFPs.

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Introduction

The inflammatory fibroid polyp (IFP) is a benign lesion that arises from submucosa of the gastrointestinal tract most commonly in the antrum (70 %), ileum (20 %) and only occasionally in the duodenum and jejunum.[1,2] Vanek was the first to describe it as eosinophilic submucosal granuloma followed by Helwing who established the term inflammatory fibroid polyp in 1953.[3] These polyps occur more commonly in males, and stomach is the favored site followed by ileum, colon and duodenum.[1]

Morphologically, IFP shows spindle cell proliferation with marked eosinophilia and prominent small blood vessels. A characteristic feature is “onion skin” arrangement of spindle cells around blood vessels.[5] Immunohistochemically IFP shows reactivity for CD34, Smooth muscle actin, and CD68 and shows negative expression for CD117, DOG1, S100 and cytokeratin.[1]

The exact etiopathogenesis of IFP is still controversial. An autoimmune etiology superimposed with an environmental trigger, such as local infection stimulating an allergic reaction or an exaggerated host response to unknown stimulus is suggested.[5] Recent research has shown mutations in exon 12 or 18 and, less frequently, in exon 14 of

platelet derived growth factor receptor alpha gene indicating a possible neoplastic etiopathogenesis of the lesion.[3] There are numerous case reports of solitary IFP, but there are very few case reports of patients presenting with multiple inflammatory fibroid polyps (IFPs).We are presenting our case report of multiple IFPs in a female patient with the review of literature.

Case Report

A 40 years old non-diabetic and non-hypertensive lady presented with complaints of abdominal pain, vomiting indigestion, anorexia and abdominal fullness for 15-20 days. X-ray abdomen demonstrated air fluid levels. Her abdominal CT scan report showed multiple lobulated soft tissue masses serving as a lead point and causing intussusception. She had a past history of cholecystectomy appendectomy, two caesarean sections and laparotomy for intestinal obstruction. She had a family history of intestinal obstruction in father, grandfather, aunt from father side and sister for which they underwent laparotomy. Histopathology report of sister was available in which diagnosis of inflammatory fibroid polyp was given. However, histopathology reports of father, grandfather and aunt from father side are not available. Laparotomy was planned for this patient. Intraoperative findings included multiple variable sized masses related to bowel along mesentery at various foci. Small gut resection was done and resection specimen was sent to us. Gross examination of small gut resection specimen revealed, iatrogenically incised bowel specimen. Opening of bowel revealed multiple polyps in the lumen ranging in size from 5 to 100 mm. Further slicing of these polyps revealed a fleshy cut surface (Figure 1 and Figure 2).

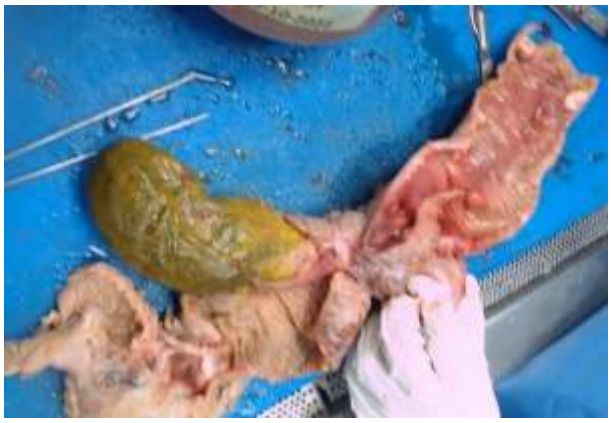

Figure 1: Small Gut Resection Specimen showing multiple polyps

Figure 2: Cut Section of Polyp showing Fleshy Appearance

Histological examination revealed exuberant, non-encapsulated, submucosal proliferation of mononuclear spindle shaped cells with an inflammatory infiltrate dominated by eosinophils. A network of variably sized blood vessels in background noted. Arrangement of spindle shaped cells was concentric giving “onion-skin” like appearance (Figure3 and Figure 4).

To differentiate this lesion from gastrointestinal stroma tumor, CD117 and DOG1 were performed which were negative. Hence the diagnosis of multiple inflammatory fibroid polyps was made.

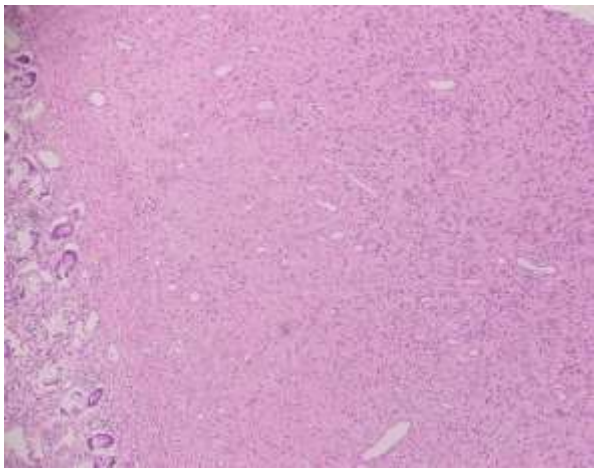

Figure 3: IFP at 10x showing spindle cell proliferation in a background of inflammation and vascular proliferation

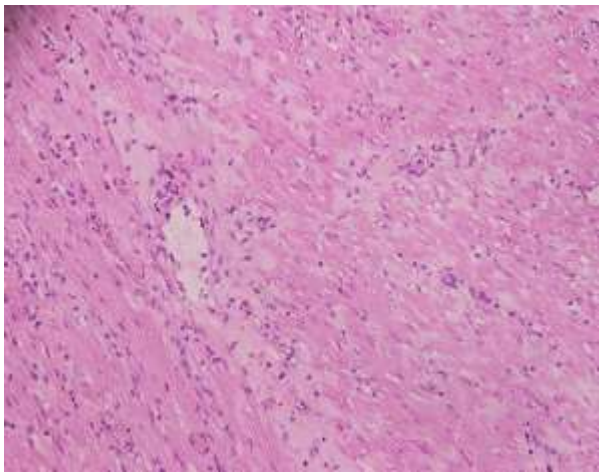

Figure 4: IFP at 40x showing spindle cells with prominent eosinophils

Table I: Comparison of IFP cases reported during 2000-2022

Sr. No.	Author	Year/Ref	Sex	Age	Clinical Presentation	Family Hx	No of polyps	Location
1	M hussain	2008 [1]	F	30 years	Small bowel obstruction	+ve	3	Mid ileum
2	J.Fazzio	2015 [3]	F	70 years	Nausea, vomiting bloody stool	-ve	1	Antrum
3	WysocKi A.P	2007 [4]	F	53 years	Right upper quadrant pain radiating to back	-ve	1	Duodenum
4	Afroditi Kourti	2022 5	F	12 months	Rectal bleeding and polypoidal mass protruding from anus	-ve	1	Anorectal junction
5	Srikantaiah	2015 [6]	M	46	Fever, constipation and abdominal distension	-ve	1	Distal ileum
6	Jun Sang Bae	2014 [7]	F	48	Abdominal pain	-ve	1	Ileum
7	Yifan MD	2022 [8]	F	41	Abdominal pain, distension	-ve	3	Esophagus, gastric horn, duodenal bulb
8	Ki Bun Park	2022 [9]	M	23	Abdominal pain, squeezing epigastric pain, fever, constipation	-ve	1	Jejunum
9	Iftikhar A et al	2022	F	40	Abdominal fullness, anorexia pain	+ve	Multiple	Ileum

Discussion

Multiple IFPs is a rare entity. Its incidence is 0-2 %. IFP was first described in 1920 as "Polypoid fibroma". In 1949, Vanek described the term "Submucosal granuloma" and described six cases. Later it became recognized by different names; eosinophilic granuloma, inflammatory pseudotumor and Vanek polyps.[3] IFPs occur most commonly in stomach followed by small intestine leading to intestinal obstruction with a slight predominance in males.[1] IFP lesion causes intussusception which is the main cause of intestinal obstruction, however the sign and symptoms differ according to the site of origin.

Macroscopically IFPs are either sessile or pedunculated, usually solitary and non-encapsulated.[4] Histologically these lesions are composed of spindle cell proliferation, small blood vessels, inflammatory infiltrate comprising of lymphocytes, plasma cells and predominantly eosinophils and a myxoid edematous stroma. Four distinct histological patterns have been described, i.e., classical fibrovascular, nodular, sclerotic, and oedematous.[4] Differential diagnoses include gastrointestinal stromal tumor, inflammatory myofibroblastic tumor, schwannoma, solitary fibrous tumor and inflammatory pseudotumor.[3,4] Immnonohistochemically spindle cells of IFPs are positive for CD34, Vimentin and Cyclin D1 (suggesting a defect in cell cycle regulation) hence, concluding proliferating stromal cells are of dendiritic cell origin with myofibroblastic differentiation and negative for smooth muscle markers, neural markers (S100) and GIST markers (CD117, DOG1).[5-7]

There are numerous case reports of solitary IFP, but very rare case reports of multiple IFPs. Our case is reported with multiple IFPs. When we reviewed the literature, we found that there are only 2 cases of multiple IFPs reported between 2000-2022. The first case reported by M Hussain et al in 2008 was of 30-year-old female patient who presented with small bowel obstruction.[1] She had three polyps in mid ileum with a positive family history. This case differs from our case in that aspect our female patient was ten years older than this patient and more than three polyps were present in ileum. However, our patient had family history of IFP as the patient reported by M Hussain et al. Second case compared was reported by Yifan MD in 2022, who reported a case of 41-year-old female who presented with abdominal pain. She had three polyps in esophagus and stomach with negative family history.[8] This case differs from our in that aspect, the location of polyps in our case is ileum and a there is a positive family history. Other cases reported have solitary IFP with negative family history (table 1)

Conclusion

Multiple IFPs is a rare entity. As it is a spindle cell lesion, it can be misdiagnosed as other spindle cell lesions like gastrointestinal stromal tumor, inflammatory myofibroblast tumor etc. Knowledge of this lesion is therefore important as treatment and prognosis of all these lesions differ from each other. Correct diagnosis of inflammatory fibroid polyp is essential to avoid over or undertreatment and better patient outcome.

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