Filiform Polyposis: Just an Ibd’s Shade or More?

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1. Abstract

1.1. Introduction

Postinflammatory polyps (PIPs) are non-neoplastic lesions originating from the mucosa after repeated periods of inflammation and ulceration connected with excessive healing processes.

This pattern is usually associated with Inflammatory Bowel Diseases (IBD), but rarely they can be related to different clinical conditions.

PIPs can be classified into: localized, multiple, giant, generalized and filiform polyposis (FP).

FP is characterized by numerous finger-like inflammatory polyps, lined by normal colonic mucosa. If they reach up to 100 mm forming bridges and tumor-like mass, they can be defined Giant Polyposis (GP).

To date, FP has not the tumoral transforming potentiality.

1.2. Results

The first presented case is a 52-year-old man with a past clinical history of Ulcerative Colitis (UC) who underwent rectosigmoidoscopy for distal sigmoid obstruction and an Hartmann’s procedure was performed; the histological findings suggested a pattern consistent with an active fase of UC with a lumen obstructive FP.

The other patient is a 40-year-old man who suffered from colonic substenotic episodes; the colonscopy revealed an increase in number and dimension of mucosal polyps with bridge formation. The patient underwent resection of ileum and recto-sigmoid junction with an ileocolic anastomosis T-T; the surgical specimen showed a large polyposus tumor-like mass with finger-like projections with non-specific inflammatory changes and without epithelial dysplasia or neoplastic lesion.

1.3. Conclusions

This short case series points out how eterogenous the presentation of FP could be: there is no certain link between IBD and FP; in addiction, these polyps can form in a generalized condition of bowel inflammation.

2. Introduction

FP is a rare cause of non-neoplastic, non-syndromic polyposis of uncertain pathogenesis. FP is most frequently secondary to a post-inflammatory reparative process and may occur in 10–20% of the cases of IBD: it is believed that chronic inflammation of the large bowel mucosa with repeated ulceration and healing may lead to the formation of worm-like polyoid projections. FP typically presents as one to hundreds of uniform, slender, arborizing, vermiform projections of the bowel mucosa and submucosa lined by normal or inflamed mucosa. In rare cases, polyps coalesce and a large tumor mass which may measure over 15 mm, known as Giant Filiform Polyposis (GFP), is found. The mucosa surrounding the polyps may be normal or show acute or chronic inflammatory changes, depending on the context in which it arises [1, 2, 19-24].

Non-IBD associated GFP is extremely rare, for instance the overall evidences about FP describe few cases related to Histiocitosis X.
Tuberculosis and Diverticular Disease [3, 4, 5]. In literature, there are only four cases of FP associated with the findings of colon dysplasia or adenocarcinoma, but immunohistochemical and genetic analysis don’t support the theory that inflammation could trigger the occurrence of adenocarcinoma in FP [6, 7].

FP onset can be characterized by a variety of symptoms including obstruction [7, 8]. Our case series, indeed, displays two cases of FP manifesting as colon obstruction and the histological examination confirmed an inflammatory framework linked with these polyps, supporting the idea of a flogistic pathogenesis of FP.

3. Results

3.1. Case 1: 52-Year-Old Man with A Known History of UC Comes to Medical Attention for Disease Flare-Up with Impassable Distal Sigmoid Stenosis

A 52-years-old man with a 20 years history of UC was admitted in April 2021 for reacutization. Prior surgery consisted in right inguinal hernioplasty. Colonoscopy and Computed Tomography (CT) of the chest and abdomen showed a pseudo-mass-like complete stenosis in the sigmoid colon and biopsies revealed active Crohn-like UC: hence, it was not possible to exclude the malignancy of the lesion. Pre-operative laboratory tests indicated anemia (Haemoglobin = 10.9 mg/dl, Haematocrit = 35.1%). Due to the dubious diagnosis of the bowel stenosis, various surgical strategies were evaluated and discussed with the patient: because of the Crohn-like features of the biopsies, total colectomy with J-pouch seemed to be not safe enough relating to the long-term follow-up of the IBD; on the other hand, the incomplete pre-operative endoscopic evaluation of the large bowel related to the stenosis didn’t make sigmoidectomy with direct (upfront) anastomosis a valuable option.

As a result, in May 2021 a laparotomic Hartmann’s Procedure was executed, proving to be the safest, the most effective procedure and the right balance for this specific pathological condition. The surgical sample was made up of a 13 cm-long resection of the sigma. The lumen of the bowel for the entire extension was occupied by numerous large worm-like polyps. The wall appeared thickened with abundant fibrin-hematic material stratified on the serosa (Figure 1-A).

Histologically, at the level of the intestinal wall, there were extensive ulcerations, covered by granulation tissue that extended up to the muscularis, and more superficial aphthoid and fissure-like ulcers (Figure 1-B).

The mucosa surrounding ulcers was the site of intense acute and chronic inflammatory infiltrate with cryptic abscesses and granulomatous reaction around glandular rupture; the crypts glandular were distorted, have basal plasmacytosis and Paneth’s metaplasia. Numerous filiform polyps with vascular axis and inflammatory pseudopolyps were recognizable in the mucosa.

Intense fibrosis associated with lymphoid nodular aggregates in the submucosa, the muscularis propria and the perivisceral adipose tissue was observed.

The set of findings points to an UC in the phase of severe type activity fulminant, with Crohn-like aspects and FP.

Figure 1-A. Resected and cut-open colon presenting worm-like polyps; Figure 1-B. Hematoxylin- and eosin-stained histological section of worm-like polyps with fissure-like ulcers and abundant Chron-like inflammatory infiltrate.

3.2. Case 2: 40-Year-Old Man with No History of IBD

A 40-years-old man was seen for the first time in 2017 for bloody diarrhea. Colonoscopy revealed a stenosing exophytic lesion of the splenic flexure, not allowing the passage of the endoscope; biopsies were in favor of IBD. The patient then underwent robotic right hemicolectomy and histological examination of the specimen (6 cm of ileus and 39 cm of cecum) showed the presence of multiple polypoid lesions with mild chronic inflammation but without any sign of mucosal dysplasia: the final diagnosis was GFP not associated with IBD. Colonoscopy was repeated a year after surgery with both macroscopic and histological evidence of peri-anastomotic recurrence. For persistence of mucorrhea and rectal bleeding, a colonoscopy was repeated in 2019 with evidence of an increase both in number and in dimension of mucosal polyps combined with bridge formation; the diagnosis was steroid associated-IBD. A CT Enterography performed in 2019 showed ileal and anastomotic substenosis and multiple polyposis, so that the patient started a treatment with Verdolizumab in March 2020. The patient was evalua-
ted by our team for a clinical worsening in May 2021: laboratory tests were consistent with thrombocytosis (Platelets = 540000/mm3), leucocytosis (WBC= 17370/mm3), low iron level (Iron= 25 mg/dl). He then underwent laparotomic sub-total colectomy with ileo-rectal T-T anastomosis. His post-operative recovery was uneventful and he was discharged seven days after surgery.

The surgical specimen consisted of a 40 cm ileo-colic resectomy, comprising ileum, descent colon, sigma and superior rectum. The resected specimen revealed a large polyposus tumor-like mass with finger-like projections, which almost occludes the lumen of the colon and sigma, associated to a considerable thickening of the wall with an attached fragment of omentum. The mass was made up of numerous worm-like polyps up to 3 cm in length, whose axis lay lengthwise in the intestine. (Figure 2-A). The lumen of the rectum appeared dilated and the mucosa devoid of macroscopic lesions.

Histologically, the polyps consisted of a central core of submucosal connective tissue with overlying normal mucosa characterized by non-specific inflammatory changes (Figure 2-B).

The mucosa between adjacent polyps was partly ulcerated with fissuring, foci of abscessulation, fibrosis of the submucosa extended to the perivisceral adipose tissue with adherence to the omental tissue. Nodular lymphocytic aggregates with no granulomas were found. No epithelial dysplasia or neoplastic lesion were observed.

4. Discussion

In 1974, Appleman et al first described a characteristic radiographic appearance of numerous, small, elongated filling defects in the colon with normal haustral pattern: they called it “filiform polyposis” [9].

This particular rare aspect was found to be more related to middle-aged patients who suffered from IBD. Although filiform polyps can be located in esophageal, gastric and intestinal wall, sigmoid colon is the most involved site [10].

About radiological description, filiform polyps may appear in miscellaneous ways: thin, straight filling defects resembling the stalk of a polyp without a head, in some cases a radiating pattern of filling defects, a branching pattern particularly at the tip of the polyp [11].

Generally, FP presents as one to hundreds of uniform, slender, worm-like vermiform projections of the bowel mucosa and submucosa lined by normal or inflamed mucosa [1]. If mucosal projections reach up to 100 mm forming tumor-like masses, polyps are defined as GP [19-24].

Microscopically, these polypoidal structures are lined by bland-ap-
paring epithelium with no evidence of dysplasia. Histologically, a polyp is characterized by dilated blood vessels and fibrovascular cores in submucosal tissues and covered with normal mucosa [1, 2].

In patients with IBD, long-term inflammation of the colonic mucosa with alternative periods of ulceration and healing is believed to be a prerequisite for the development of these worm-like polyps. Some authors illustrated rare cases of FP occurring in the setting of unusual inflammatory conditions, for example colonic Tuberculosis and Histiocytosis X; additionally, it seems that inflammatory cytokines and the traction of redundant mucosa by intestinal peristalsis with hyperplasia and fibrosis of the intestinal wall, induced by colon diverticulitis, might relate to the development of FP [3, 4, 5].

Recently, filiform serrated polyp has been described as an uncommon type of serrated adenoma, characterized by dysplastic epithelium with serrated contours: despite the term used to describe this aspect, no association of this type of adenoma with FP has been reported to date. Sometimes, FP are occasionally difficult to distinguish from filiform serrated adenoma based on their endoscopic appearance, hence, a biopsy or polypectomy is necessary to confirm the exact diagnosis [12, 13, 17].

Even if GP may be mistaken for malignancy and in literature few cases of FP coexisting with colon adenocarcinoma have been seen, FP is considered to be a benign polyposis without any risk of dysplasia or neoplasia [14].

Infact, molecular analysis of adenoma and adenocarcinoma revealed microsatellite stable status and absence of BRAF mutation; meanwhile, immunohistochemistry showed abnormal p53 expression in the adenoma and adenocarcinoma, but this was not observed in the mucosa of the polyps [15].

Long-term follow-up are proposed to patients affected by FP, but there are defined clinical conditions which require an active therapeutic intervention: when biopsy proves adenoma or adenocarcinoma or patients with FP experiences symptoms such as bleeding and obstruction, polyps should be removed through a colonoscope or surgical colectomy [7, 8].

Clinically, FP is mostly asymptomatic and incidentally diagnosed on colonoscopy. However, patients may present with a variety of symptoms, including anemia, weight loss, cramping abdominal pain and diarrhea. Sometimes, this pathological situation can also produce obstruction and intussusceptions or it presents as toxic megacolon [7, 8, 18].

We report two cases of FP in patients with deeply different anastomotic background. The first case was a 52-years-old man with a well-known history of UC who presented to our centre with a pseudo-mass-like complete stenosis in the sigmoid colon due to an active Crohn-like UC. The pathological analysis of the specimens after a laparotomic Hartmann’s Procedure confirmed numerous large worm-like polyps with multiple ulcerations, covered by granulation tissue and more superficial aphthoid and fissure-like ulcers; in the mucosal layer surrounding ulcers, severe acute and chronic inflammatory infiltrate with cryptic abscesses and granulomatous reaction were detected. All of these histological findings prove an UC in the phase of severe type activity fulminant, with Crohn-like aspects and filiform polyposis.

The second patient, a 40-years-old man, complained various episodes of bloody diarrhea with a stenosing exophytic lesion of the splenic flexure so that he underwent robotic right hemicolectomy with an histological diagnosis of GFP not associated with IBD. One year later, because of peri-anastomotic recurrence of mucosal polyps increased in dimension and number with ileal and anastomotic substenosis, a laparotomic sub-total colectomy with ileo-rectal T-T anastomosis was performed: a large polyposus tumor-like mass with finger-like projections occluding the lumen of the colon and sigma has been revealed, but the polyps were covered by normal mucosa and nonspecific inflammatory changes without any evidence of epithelial dysplasia or neoplastic lesion.

This report aims to highlight the diversified clinical and, notably, histological presentation of FP: to our knowledge, study of pathogenesis of FP are still ongoing with the result there are not distinct criteria which allow to put a defined tag on FP: the recurring feature is the presence of an unspecified inflammatory substrate.

5. Conclusions

We confirm there is not a clear link between IBD and filiform polyposis, but we intend to suggest the importance of the bowel inflammation as background of FP. Further studies are necessary to investigate deeply the knowledge about this flogistic basal status so as to allow the creation of criteria to define properly the patient who suffers from FP. Accordingly, it would be possible to establish a well-defined surveillance to standardise the diagnostic-therapeutic process of FP. In addiction, analyzing and understanding the pathologic mechanism of FP could lead to a “target therapy”, which would enable to optimize the quality life of patients with FP.

References


