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Ileo-Ileal Intussusception Caused by an Inflammatory Fibroid Polyp: A Case Report

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Abstract

Inflammatory fibroid polyps (IFP) are a rare benign entity that arises from the submucosa of the gastrointestinal tract and protrudes into the lumen. The clinical presentation depends on the size and localization. They are particularly found in the ileum, where they are frequently present as an ileal intussusception. We report the case of a 56-year-old female patient who presented with periumbilical pain along with multiple episodes of vomiting and chronic constipation. An abdominal CT scan revealed an ileo-ileal intussusception of an endoluminal hypodense lesion with mesenteric lymphadenopathy. Surgical treatment consisted of segmental ileal resection with primary anastomosis. The histopathological analysis revealed an inflammatory fibroid polyp in the ileum. Although these tumors have no malignant potential, surgery is always indicated in ileo-ileal intussusception to ascertain the histological nature of the lesion.

Keywords

Intussusception, Inflammatory Fibroid Polyp, Surgical Resection

1. Introduction

Inflammatory fibroid polyps are a rare entity among the benign pathologies of the submucosa of the digestive tract. They are mainly observed in adults and occasionally in children.

The symptomatology depends on the location of the polyp. In the case of intestinal location, invagination is often the mode of revelation reported in adults [1].

Intestinal intussusception, or invagination, is a condition in which a segment of the bowel slides into an immediately adjacent segment.

We report a case of a patient who was admitted to the emergency department with a bowel obstruction attributed to intestinal invagination.

2. Clinical Case

2.1. Patient History

B.D. was a female patient, age 56, with a medical history of hypertension treated with calcium channel blockers as a monotherapy, and she was admitted to the emergency department for acute bowel obstruction evolving for 5 days.

2.2. Clinical Finding

A physical exam revealed a distended abdomen with tenderness in the periumbilical region, without any palpable mass or organomegaly. Examination of hernial orifices revealed no abnormality.

A digital rectal examination found an empty rectum.

The rest of the physical exam was without abnormalities, and the patient was hemodynamically stable.

The patient's blood tests revealed hyponatremia at 130 mEq/l, potassium level at 4.20 mEq/l, and chloride level at 100 mEq/l. The rest of her biochemical parameters as well as her hemogram were within normal limits.

The abdominal X-ray showed no abnormalities.

A CT scan of the abdomen showed an ileo-ileal intussusception, an endoluminal hypodense lesion as a lead point, and mesenteric lymphadenopathy.

After medical preparation and resuscitative measures, the patient underwent surgery under general anesthesia.

A midline abdominal laparotomy was performed. Operative exploration found an ileo-ileal intussusception at 100 cm from the ileocecal valve without any signs of necrosis (**Figure 1**).

Segmental ileal resection was performed without releasing the intussusception, followed by a termino-terminal anastomosis using the 3-0 absorbable monofilament suture.

The gross examination of the specimen showed an ovoid, white, light-obstructing mass with regular contours, measuring 4 × 3 cm (**Figure 2**). The adjacent mucosal folds appeared normal.

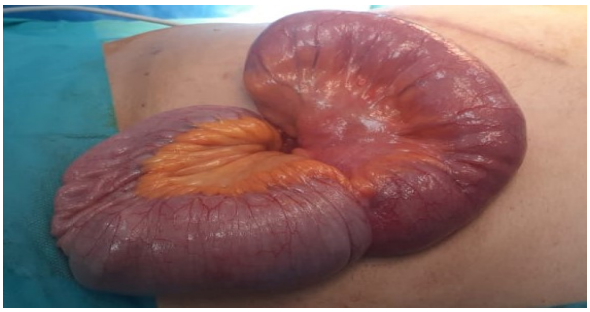

Figure 1. Per operative view of the intussusceptions.

Pathology showed a proliferation of spindle-shaped cells that were arranged in short bundles with no cytonuclear atypia. The stroma was fibromyxoid, highly vascularized, with an eosinophil-rich inflammatory infiltrate (Figure 3).

Immunohistochemistry analysis confirmed the diagnosis of inflammatory fibroid polyp (Figure 4).

Figure 2. Macroscopic appearance of the polyp as a whitish tumor.

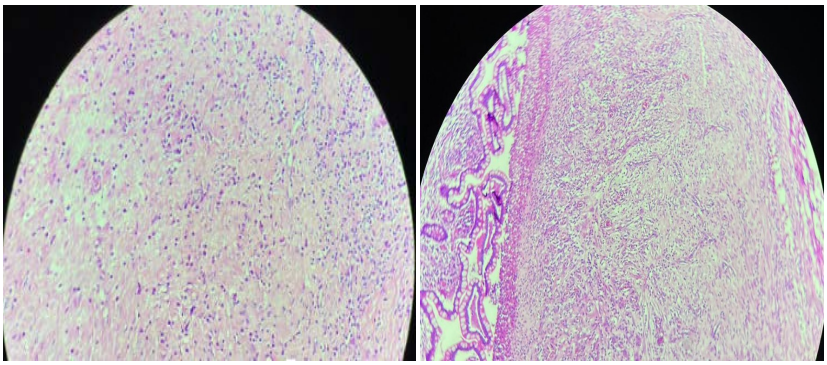

Figure 3. Microscopic study with onion bulb appearance of our case report, Ibn Sina Rabat pathology laboratory.

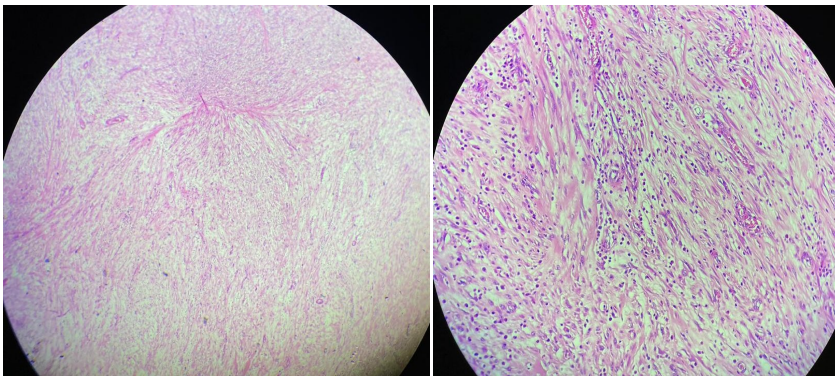

Figure 4. Additional microscopic and immunohistochemical studies of our case report, Ibn Sina Rabat pathology laboratory.

2.3. Follow-Up and Outcome

The patient had an uneventful postoperative recovery with no specific complica-

tions.

Oral intake was authorized on the third postoperative day (POD) and the patient was discharged on POD 6.

There were no recurrences at the 10-month follow-up. A total colonoscopy and esophagogastroduodenoscopy showed no other locations or abnormalities.

3. Discussion

Inflammatory fibroid polyps (IFP) are a rare and benign anatomopathological entity of the gastrointestinal tract [1]. They were initially called "Vanek's tumor" or "eosinophilic granuloma" by Vanek in 1949. The term "inflammatory fibroid polyps" was proposed by Helwing in 1953 [2] [3].

IFPs can occur anywhere in the gastrointestinal (GI) tract, but they are most common in gastric localization followed by the small intestines, especially the jejunum and colon [4] [5]. When located in the small intestines, IFP can manifest an intestinal obstruction attributed to intestinal invagination, which is the case in our patient [6].

These lesions arise from the submucosa of the GI tract and present as small nodules protruding into the lumen; they may be sessile or pedunculated.

The clinical presentation of PFAs is variable. Patients are usually asymptomatic, and polyps are discovered incidentally on prior radiologic or endoscopic examination.

When present, symptoms are polymorphic and vary according to the size, location, and the number of polyps. They may include generalized abdominal pain over several months with or without alteration of the general condition, upper or lower digestive bleeding, and a sub-occlusive or occlusive syndrome attributed to intestinal invagination when the localization is the small bowel, as in our patient [7].

The paraclinical diagnosis of intestinal obstruction is often radiological, allowing the diagnosis of intestinal invagination. Abdominal radiography without preparation shows images of hydro-aeros levels often localized topographically according to the site of the intussusception or sometimes a rounded opacity of hydric tone circumscribed on one side by a clear crescent and which may contain within it clear arciform images (a "coil spring" appearance) [8]. (**Figure 5**)

Abdominal CT with contrast injection or enteroscanner is an examination that allows a global study of the digestive tract. In the non-emergency context, the PFI appears as a regular protrusion in the digestive lumen without any thickening of the wall opposite.

In the case of intestinal invagination, as in the case of the observation described above, a cocooned tissue mass is visualized at the exact site of the occlusion, and the severity is assessed by looking for signs of complications, essentially digestive distress or perforation [9] [10].

On the other hand, the CT scan in the diagnosis of PFI cannot determine which layer develops, hence the interest in performing an echo-endoscopy outside of the emergency context, which allows for making the exact topographic diagnosis.

Abdominal MRI or entero-MRI remains more efficient than CT in diagnosis

of PFI, which appears as a hypointense endoluminal tumor on the T1 sequence and intermediate on T2 [11]. (**Figure 6**)

Digestive endoscopy remains a reference examination in the diagnosis of these polyps, allowing us to visualize the site of the tumor, the size of the polyp, which can go from a few millimeters to several centimeters, and the macroscopic aspect (generally, the millimetric polyps are sessile whereas those that are voluminous are pedicled), and to take biopsies to confirm the histological diagnosis. Sometimes it also allows us to make a therapeutic gesture in exactly the right situations.

Macroscopically, the polyp develops from the submucosal layer, and it is manifested at high fibroscopy by an endoluminal expansion localized to the antral or pre-pyloric region, which is covered by normal mucosa with an ulcerated central depression for polyps that exceed 10 mm [14].

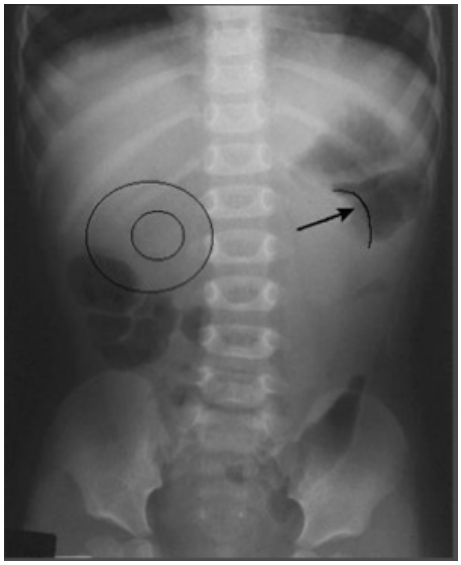

Figure 5. Unprepared abdomen showing an intussusception with an impression of the bladder [12].

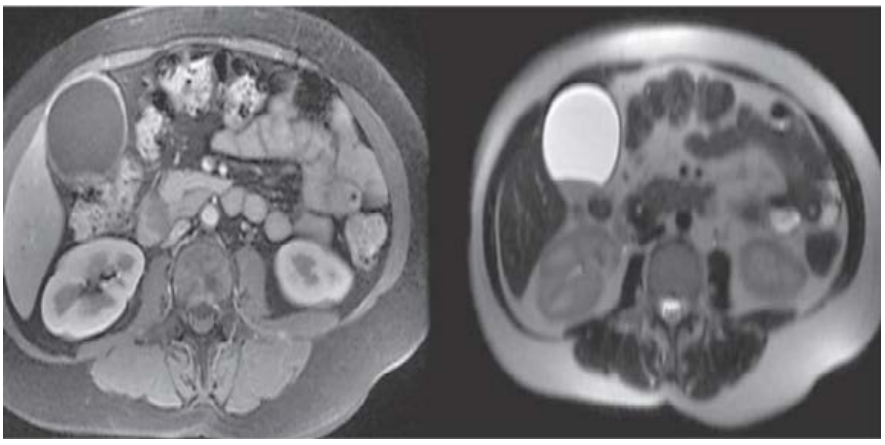

Figure 6. Magnetic resonance imaging shows a hypointense 26 mm image on the T1 sequence (left), and intermediate intensity on the T2 sequence (right) in favor of the PFI of the duodenum [13].

Sometimes these tumors are covered with a whitish exudate on their surfaces, a feature described by Tanaka *et al.* [15].

Concerning the colonic setting, the polyp is in the form of a pedunculated tumor protruding into the lumen covered by healthy mucosa. [16]

An echo-endoscopic complement plays an important role in the characterization of submucosal polyps, particularly gastric or rectal polyps. The appearance is of a heterogeneous hypoechoic mass arising from the submucosa. This appearance can be confused with GIST or submucosal lipomas [17]

The biological workup is not very helpful in the diagnosis of PFI. In some cases, we note the presence of microcytic hypochromic anemia, motivating the realization of a digestive exploration, and finding a polyp with occult bleeding. Two cases reported in the literature by I. ED-DARRAZ *et al.* [18]

Anatomical-pathological examination remains the only means of confirming the diagnosis of Vaneck's polyp. It is performed on a biopsy specimen during endoscopy or on the surgical specimen. The macroscopic appearance of the polyp is in the form of a whitish, firm, and pale mass, sometimes myxoid, not encapsulated. In this section, the submucosal origin is confused, and sometimes it may protrude beyond the muscularis propria, giving an hourglass aspect [19].

Microscopic examination shows a vascularized fibroinflammatory tissue with 3 entities: fibroblastic, inflammatory, and vascular. The cells are made up of elongated or star-shaped monomorphic mesenchymatous elements with amphophilic cytoplasm; mitoses are rare, and the extracellular space contains mucoid material with collagenous and reticulin fibers dispersed between the cells. Spindle cells are often arranged in clusters or concentrically around the vessels, creating an "onion bulb" appearance [20] [21] [22].

The immunohistochemical study plays an important role in the diagnosis of Vaneck's polyp by studying these antibodies which are present in most cases: vimentin, cyclin D1, CD 34, smooth muscle actin, or HFF35, fascin, calponin, and Desmin [4] [8] [23].

Molecular biology shows a genetic abnormality with the existence of a mutation in exon 12 of the PDGFR alpha gene, which predominates in intestinal PFI, and a mutation in exon 18 of the same gene in gastric PFI [24].

The differential diagnosis is essential with gastrointestinal tumors or GIST, which are the most frequent tumors of the digestive tract, hence the interest of the immno-histological study.

The curative treatment consists of the resection of the polyp either by the endoscopic technique of polypectomy or mucosectomy for small polyps; or resection by surgical means depending on the localization, the urgent symptomatology, or not of the polyp.

Generally, small polyps of incidental discovery or asymptomatic with small sizes do not require complete resection. This, as well as the potential for degeneration, has not been demonstrated, so there is no consensus on the pace of monitoring.

The surgical procedure is still being discussed as a curative option. The pro-

cedure depends on the location of the polyp. In symptomatic gastric forms that cannot be resected endoscopically, the procedure consists of partial gastric resection or wedge resection, depending on the location of the polyp. In the case of colonic or gastric forms, and especially in the case of occlusion, tumor resection is performed according to the affected segment with the restoration of digestive continuity during the same surgical procedure if the preoperative conditions allow it.

The evolution is marked by the absence in the majority of cases of recurrences or metastases after complete resection, confirming the benign nature of the polyp.

4. Conclusion

The inflammatory fibroid polyp or Vaneck's polyp is a benign lesion of the digestive tract with submucosal development. It is observed in adults and is usually a fortuitous discovery. The diagnosis is essentially histological on endoscopic resection or the surgical specimen after surgery for occlusion of intestinal intussusception as in the case described above.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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