Ulcerated intussuscepted jejunal lipoma-uncommon cause of obscure gastrointestinal bleeding: A case report

Tudor Cuciureanu, Laura Huiban, Stefan Chiriac, Ana-Maria Singeap, Mihai Danciu, Florin Mihai, Carol Stanciu, Anca Trifan, Nutu Vlad

Abstract

BACKGROUND

Intestinal lipomas are rare benign gastrointestinal (GI) tumors, usually asymptomatic, but may become symptomatic as the result of some complications such as intussusception, intestinal obstruction, volvulus or bleeding. They can occur at any site along the entire GI tract, more frequent in colon and rarely in small intestine. The patient reported here is a very rare case of jejunal lipoma, ulcerated and intussuscepted, diagnosed in an adult investigated for a chronic iron deficiency anemia (IDA), and successfully managed by segmental jejunal resection.

CASE SUMMARY

A 63-year-old male was referred to “St. Spiridon” Hospital, Institute of Gastroenterology and Hepatology, Iasi, to investigate an obscure GI bleeding with an IDA. After upper GI endoscopy and colonoscopy were performed, excluding potentially bleeding lesions, videocapsule endoscopy was then carried out, revealing fresh blood and a protruding lesion in proximal jejunum, findings confirmed by a single-balloon enteroscopy. Multiple biopsies were taken from the lesion, but histological results were inconclusive. Then, contrast - enhanced computed tomography was performed revealing the intussuscepted jejunal segment which was resected en
INTRODUCTION

Lipomas are benign, fatty gastrointestinal (GI) tumors, more commonly located in the colon (64%)\[1\], the small intestine being the second site, and very rarely in the jejunum (< 2%)\[2\]. Usually, small bowel lipomas are asymptomatic, uncomplicated and discovered incidentally during investigation for other abdominal pathologies or when cases present a complication such as intussusception, intestinal obstruction or bleeding. The intussusception is defined as the telescoping of a proximal segment of bowel into the lumen of an adjacent segment of bowel (usually resulting in obstruction), and it is more common in children, where is generally idiopathic; it seldom occurs in adults (with 5% of all cases of intussusception)\[3\] and it is often due to a malignancy (70% of the colonic and 30% of small bowel intussusceptions are attributable to malignancy)\[4\]. In a retrospective study including all adult patients diagnosed with intussusception at Mayo Clinic, Rochester from 1983 to 2008, among all 196 patients, only 10 presented with small bowel lipomas\[5\]. We report a very rare case of an ulcerated intussuscepted jejunal lipoma-uncommon cause of obscure gastrointestinal bleeding: A case report. World J Clin Cases 2019; 7(22): 3765-3771 URL: https://www.wjgnet.com/2307-8960/full/v7/i22/3765.htm DOI: https://dx.doi.org/10.12998/wjcc.v7.i22.3765

CASE PRESENTATION

Chief complaints
A 63-year-old man with personal history of hypertension was admitted to our department of Gastroenterology and Hepatology to investigate an obscure GI bleeding with iron deficiency anemia (IDA).

History of present illness
He complained of intermittent abdominal pain accompanied by nausea.

History of past illness
He had a prior history of hypertension well controlled by treatment with angiotensin-converting enzyme inhibitors. He had no abdominal surgery.

**Personal and family history**
He was a construct engineer and a current smoker (15 cigarettes/d for the past 20 years). He had no serious family history.

**Physical examination**
Physical examination showed pale teguments, and the abdomen was soft and tender in the umbilical and right flank area, without any palpable abdominal masses.

**Laboratory examinations**
Laboratory data showed IDA (hemoglobin 9.5 g/dL, serum iron 45 µg/dL, ferritin 10 µg/L).

**Imaging examinations**
An upper GI endoscopy and colonoscopy were performed, excluding lesions with potential for bleeding. Then, videocapsule endoscopy was performed, revealing fresh blood in the proximal jejunum, and a protruding lesion, with discolored covering mucosa (Figure 1A and B). Next, a single-balloon enteroscopy was carried out, which showed a polypoid mass with ulceration, situated in the proximal segment of jejunum (Figure 2A and B). Multiple biopsies were taken from the lesion, but the histological result was inconclusive, as it frequently occurs in submucosal GI benign tumors including lipomas, due to depth factor—the amount of submucosal tissue required in biopsies of the lesion. Then, contrast-enhanced abdominal computed tomography was performed which showed a 6 centimeters elongated structure inside the intestinal lumen with homogenous fat density and smooth well-defined contour, suggestive for an intestinal lipoma (Figure 3). Within the next week the patient complained of abdominal pain, nausea and several episodes of vomiting. A laparotomy was performed revealing jejuno-jejunal intussusception.

**FINAL DIAGNOSIS**
Intra-operative macroscopic observation identified six centimeters intussuscepted yellowish mass suggestive for lipoma (Figure 4). The histological examination revealed in the submucosa a nodular mesenchymal tumor consisting in mature adipocytes, with no pleomorphism and no mitotic activity. These findings were compatible with a diagnosis of lipoma (Figure 5A and B).

**TREATMENT**
The intussuscepted jejunal segment was resected en bloc and the inspection of this segment showed a submucosal firm mass with ulceration of the mucosa. End-to-end anastomosis was performed.

**OUTCOME AND FOLLOW-UP**
The patient made an uneventful recovery and was discharged seven days later, and at six months follow-up he had no complains and his hemoglobin returned to normal value.

**DISCUSSION**
Jejunal lipomas are rare, but they may, nonetheless, represent a diagnostic challenge when complicated by lower gastrointestinal tract hemorrhage or intussusception. Large benign tumors of the small bowel rarely include intussusception in their spectrum, which may become an important risk factor for ischemia and necrosis of the intestinal wall.

The most common sites of GI lipomas reported in the literature are the colon (64%), followed by the small bowel (31%), stomach (3%) and the esophagus (12%)\[^1\]. While most of small bowel lipomas are small in size and asymptomatic, those surpassing 2 cm in size usually manifest through clinical symptoms such as abdominal pain, hemorrhage or bowel obstruction\[^2\].
In adult patients, intussusception is more likely to present progressive misleading symptomatology with diffuse abdominal pain and rarely with classical triad-symptoms such as intense abdominal pain, vomiting and lower gastrointestinal hemorrhage, making the diagnosis complex and requiring further radiological documentation. Intussusception is documented frequently on computed tomography, a method of choice due to its accuracy of virtually staging the lesion\(^8\). Over 90% of intussusception cases found in adults have an organic cause\(^9\).

In order to find similar cases, we have reviewed the literature using Pub Med and found ten cases in the published accounts\(^{[4,10-18]}\). The keywords used were “lipoma”, “intussusception”, “jejunum”, “bleeding”. All ten cases presented jejunal lipoma with intussusception and bleeding (Table 1).

Over the past decade, there has been a constant debate about the appropriate and safe treatment of small bowel benign tumors. Clinical presentation differed probably on account of the different sizes in tumoral mass. Yu et al\(^{[19]}\) reported fifteen cases of gastrointestinal lipomas with different sizes that benefited from endoscopic therapy without important complications. However, it should be noted that endoscopic resection may be associated with a risk of bleeding and perforation. Thus, Raju et al\(^{[20]}\) reported that endoscopic removal of lipomas > 2 cm in diameter was associated with a greater risk of perforation. In our case, the patient presented a tumor over 5 cm in length with a wide base of implantation. Due to its size and vascularization, surgical resection was considered to be the optimal treatment.

CONCLUSION

Jejunal lipomas, very rare benign tumors of the GI tract, are mostly asymptomatic and found incidentally during investigations for other abdominal diseases. However, in some cases, they may lead to complications such as intussusception and hemorrhage. Surgical resection remains the treatment of choice for large and complicated lipomas.
Table 1  Cases of adult patients with jejunal lipoma complicated with intussusception and bleeding

| Ref. | Gender/Age | Diagnostic modality | Size (cm) | Treatment |
|------|------------|---------------------|-----------|-----------|
| Ferrara et al[10], 2011 | F/78 | CT, VCE, Laparoscopy | 3 × 3 × 2.5 | Surgical resection |
| Mouaqit et al[11], 2012 | M/35 | CT, Laparotomy | 4 | Surgical resection |
| Pinto et al[12], 2018 | M/46 | CT, VCE, Enteroscopy | 7.5 | Surgical resection |
| Sarabjit et al[13], 2013 | M/22 | CT, Exploratory laparotomy | 4 | Surgical resection |
| Manouras et al[14], 2007 | M/55 | CT, Laparoscopy | 4 × 4 | Surgical resection |
| Yiğitler et al[15], 2007 | M/76 | US, CT | 4 × 5 | Surgical resection |
| Charalambous et al[16], 2012 | M/46 | CT, Laparotomy | 4 × 4 | Surgical resection |
| Kida et al[17], 2017 | M/67 | CT, Enteroscopy | 4 | Surgical resection |
| Akyüz et al[18], 2018 | F/61 | CT | 1.5 × 1.5 | Surgical resection |
| Baron et al[19], 1996 | F/60 | CT | 5 | Surgical resection |

CT: Computed tomography; US: Ultrasonography; VCE: Video capsule endoscopy.

Figure 2  Images acquired through enteroscopy performed in our patient. A: Ulcerated tumoral mass; B: Tumoral mass with partial bowel obstruction.

Figure 3  Contrast-enhanced abdominal computed tomography scan. Both axial (left) and coronal (right) reformatted images show a large elongated structure inside of intestinal lumen (arrowheads) with homogeneous fat density and smooth, well defined contour.
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Figure 4 Macroscopic appearance of the jejunal lipoma (arrow).

Figure 5 Jejunal submucosal lipoma with ulcerated area of the mucosa. A: Full section (Hematoxylin-eosin staining, × 40); B: Detail (Hematoxylin-eosin staining, × 200).
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