

# Malnutrition Due to Sub-occlusive Intestinal Syndrome Secondary to Extraluminal Jejunal Leiomyoma

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## Abstract

Leiomyomas comprise approximately one fourth of benign gastrointestinal tumors and are the most common symptomatic benign tumors of the small bowel. Diagnosis is difficult because of their rarity and the absence of specific symptoms.

We present a case of malnutrition due to subocclusive intestinal syndrome secondary to extraluminal jejunal leiomyoma. The patient experienced recurrent attacks of abdominal pain and distention after ingestion of food, with significant weight loss. Abdominal CT scan showed a small intestine compressive tumoral mass. Laparotomy revealed a proximal jejunal tumor. The tumor was resected and bowel continuity was restored. Histopathological study showed jejunal leiomyoma.

Usually, intestinal leiomyoma presents with gastrointestinal bleeding, or small bowel occlusion; however, abdominal pain after ingestion of food, weight loss and malnutrition constitute an exceptional presentation. We have not found another similar case published in the revised literature.

Epidemiology, clinical presentation, diagnosis and surgical management of small intestinal leiomyoma are reviewed.

## INTRODUCTION

The small intestine accounts for approximately 75% of the length and 90% of the mucosal surface of the gastrointestinal tract; despite this large surface, small-bowel tumors are rare and account for less than 2% of the gastrointestinal tumors. The most frequent neoplasms found have been the gastrointestinal stromal tumors (GIST) (36%), followed by lymphomas (24%) and adenocarcinomas (18%). Most tumors (65%) have been located in the ileum<sup>1</sup>. Leiomyomas represent one subgroup of the GIST<sup>2</sup>.

Leiomyomas are the most common of all symptomatic small intestinal tumors, comprising 25-30% of all benign gastrointestinal tumors. Sixty-five percent of the tumors are located in the stomach, with 23% in either the jejunum, ileum or duodenum<sup>3,4,5</sup>.

Leiomyomas are mostly benign and are uncommonly seen in clinical practice. They are difficult to diagnose due of their

rarity. Therefore, misdiagnosis can occur if careful history and good examination are not performed. The two most frequently presenting symptoms are intermittent gastrointestinal bleeding<sup>6,7,8</sup> and gastrointestinal obstruction<sup>1,9,10</sup>, in that order. Less commonly, one may see intestinal invagination, volvulus, intestinal perforation, recurrent abdominal pain, weight loss, nausea and vomiting, abdominal mass, jejunal infarction, and chronic diarrhea<sup>11,12,13,14,15,16,17</sup>.

We report a case of malnutrition secondary to intermittent attacks of abdominal pain and distention, after ingestion of food, of 8 months of evolution, due to sub-occlusive extraluminal jejunal leiomyoma.

## CASE REPORT

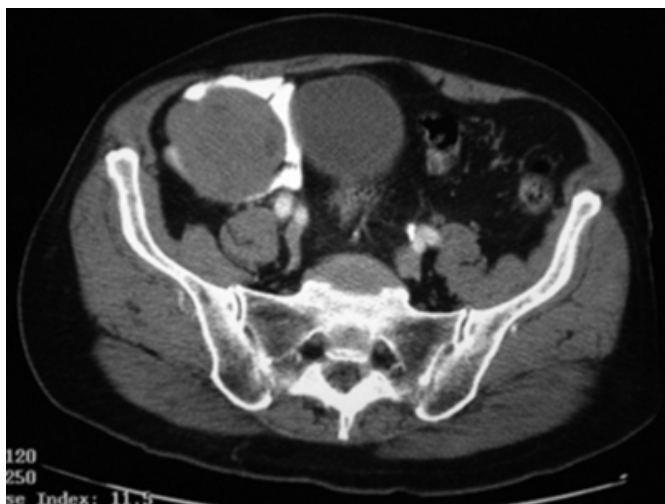
A 78-year-old male patient was admitted to our hospital with an 8-month history of intermittent attacks of abdominal pain, abdominal distention, recurrent nausea, and constipation.

Moreover, the patient referred inappetence, significant weight loss and progressive deterioration of his general state. The patient experienced increasing colics after ingestion of food. He had a history of a cerebrovascular accident without sequels.

Physical examination showed skin and mucous paleness, malnutrition signs, cardiopulmonary auscultation without alterations, abdominal palpation without mass and soft edema of the lower extremities. Rectal examination was normal. Laboratory analysis: Total leukocyte count  $2.3 \times 10^3 / \mu\text{L}$  (marked shift to the left), erythrocytes  $3.5 \times 10^6 / \mu\text{L}$ , hemoglobin 9.1g/dL, hematocrit 28%, platelets  $156 \times 10^3$ , glucose 123mg/dL, urea 45mg/dL, total protein 5.1g/dL, serum albumine 2.7g/dL, iron 17 $\mu\text{g/dL}$ , ferritin 234ng/dL, prothrombin time 79.3%, cephalin time 30 seconds. Hepatic and thyroid function were normal. Carcinoembryonic antigen, alpha-fetoprotein and prostatic antigen were normal. The thorax x-ray revealed no alterations. Abdominal computed tomography (CT) scan showed a proximal jejunal extraluminal mass of soft-tissue density causing partial obstruction of the small intestine in the right upper quadrant (fig. 1). Colonoscopy was negative.

#### Figure 1

Figure 1: Abdominal CT scan showing a tumor of 10x8cm in intimate contact with small bowel



Laparotomy revealed a jejunal stenosis caused by a proximal small-bowel tumor of 12x10cm diameter. The neoplasia almost completely obstructed the intestinal lumen.

Jejunio-jejunal anastomosis was performed after resection of the tumoral segment.

Histopathological examination showed a jejunal extraluminal leiomyoma (fig. 2). No complication occurred in

the early postoperative period, and the patient was discharged on the 9<sup>th</sup> postoperative day. After ten months of follow-up the patient is asymptomatic.

#### Figure 2

Figure 2: Macroscopic appearance of the extraluminal leiomyoma causing external compression of the jejunum



#### DISCUSSION

Leiomyomas comprise approximately one fourth of the benign gastrointestinal tumors and are the most common symptomatic benign tumors of the small bowel. They are found most commonly in the jejunum (as our case), but also in the ileum and duodenum. These tumors show three different growth patterns: intraluminal (33%), extraluminal (50%), as our case, and bidirectional (dumb-bell shaped) (17%) (18). The tumor is usually single, firm, grayish-white, well-defined and encapsulated. It originates from the mesenchyma and arises from spindle cells of the muscular layer of the intestine (1). An accurate diagnosis should include good medical history, physical exam, adequate laboratory analysis and appropriate imaging.

Symptoms and signs of small-bowel tumors are vague, unspecific and frequently are confused with other gastrointestinal manifestations, as a consequence these lesions often go untreated for prolonged periods, because they are notoriously difficult to diagnose. In some occasions these lesions can be incidental findings during unrelated surgery (19,20,21).

Jejunioileal leiomyomas can affect any age (22), have a peak incidence between 50-60 years and have a male to female ratio of 2:1 (1). Although most remain asymptomatic, non-specific complaints such as acute gastrointestinal bleeding (most common symptom with 65% frequency) (6), chronic

anemia due to occult blood loss ( 7,8 ), intermittent abdominal pain, or weight loss may be seen ( 19,20,21 ). Intermittent intestinal obstruction is the second most common complication, with an incidence of 25-45%, especially in tumors located in the ileum ( 1,9,10 ). Intussusception may also cause subocclusive syndrome or gastrointestinal bleeding due to ischemia and necrosis of the tumor ( 11,12,13,14 ). Anemia with iron deficiency and obstructive symptoms should alert the physician to examine the small intestine. Sometimes the tumor may enlarge enough to be palpable in an asymptomatic patient.

Our patient had anemia and malnutrition signs. His main symptoms were abdominal pain after ingestion of food with important weight loss; therefore, initially we suspected chronic mesenteric ischemia or a malignant tumor, although the colonoscopy was negative.

CT scan, magnetic resonance (MR), barium studies, endoscopy, endoscopic ultrasound, angiography and capsule endoscopy can be complementary tests of great utility for the diagnosis of this pathology. CT scan can show 90% of leiomyomas. They are sharply defined spheric or ovoid masses of homogeneous density and show contrast enhancement. Sometimes central necrosis can be seen, making the differentiation more difficult ( 23 ). Leiomyomas are seen as oval or round filling defects on barium studies, except extraluminal lesions, which may not be detected unless they are quite large ( 18,24 ). However, only 29.3% of primary tumors of the small intestine could be demonstrated by barium x-ray examination ( 3 ). Capsule endoscopy has increased the preoperative diagnosis of these tumors; as a consequence, they are currently diagnosed early and operated electively with increased frequency. Nonetheless, this procedure is contraindicated in suspected intestinal obstruction ( 25 ).

In most patients, the mean period between onset of complaints and diagnosis has been found to be 7 months with a range between 3 months and 5 years ( 5,18,26 ). Our patient was correctly diagnosed after eight months. Among the differential diagnoses there are other GIST, lymphomas, adenocarcinomas, mesenteric cysts and cystic lymphangiomas ( 1,16 ).

Surgical resection is the treatment of choice for gastrointestinal leiomyomas by conventional or laparoscopic approach ( 27,28 ). In patients with intestinal obstruction, complications and survival are related to age, associated morbidity, non-viable strangulation and treatment delay ( 29

). The reported morbidity rate for elective patients operated on for small-bowel tumors was 42%. The most common postoperative complications are surgical wound infection, intestinal fistula, mesenteric ischemia and prolonged postoperative ileus ( 1 ).

Strict follow-up of these tumors should be made due to the uncertain potential of malignancy of GIST. In the case of intestinal leiomyoma, this potential ranges from 10 to 20% ( 30,31 ).

## CONCLUSION

Diagnosis of jejunoileal leiomyoma is difficult because it does not have specific symptoms and frequently is confused with other gastrointestinal manifestations; as a consequence, these lesions often go untreated for prolonged periods. Misdiagnosis can occur if a careful history and good examination are not performed.

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