ISSN 2689-8268 | Volume 8

American Journal of surgery and Clinical Case Reports

Case Report Open Access

Presumed Lymphoma Revealed to Be a Rare Small Intestinal Lymphangioma: A Case Report

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Received: 10 May 2025
Accepted: 19 May 2025
Published: 22 May 2025
J Short Name: AJSCCR

Keywords: Case Report; Lymphoma; Lymphangioma; Digestive Surgery

Citation: AM Lucchese. Presumed Lymphoma Revealed to Be a Rare Small Intestinal Lymphangioma: A Case Report. Ame J Surg Clin Case Rep. 2025; 8(16): 1-3

1. Abstract

Lymphangioma is a benign tumor that typically occurs in children, most commonly affecting the head, neck, and axillary regions. This report describes the rare case of an adult patient initially suspected to have abdominal lymphoma. However, surgical exploration revealed the true diagnosis to be a lymphangioma of the small intestine and mesentery-an exceedingly uncommon presentation. The patient also exhibited an unusual clinical picture, characterized by chronic refractory iron-deficiency anemia and persistent diarrhea. Data for this case report were collected through a review of the patient's medical records, direct interviews, photographic documentation of diagnostic procedures, and a literature review.

2. Introduction

Lymphangioma is a non-epithelial tumor characterized by an aggregation of dilated lymphatic vessels. Most cases are diagnosed in early childhood, particularly before the age of two, with only about 10% identified in adulthood [1]. Intra-abdominal presentation is rare, as the most common anatomical sites of involvement are cranio-cervical and axillary regions [2, 3]. When located within the abdominal cavity, the mesentery, omentum, mesocolon, and retroperitoneum are the most frequently affected areas [3]. Involvement of gastrointestinal tract organs is even more uncommon, accounting for less than 0.05% of all cases [4].The clinical manifestations of lymphangioma are diverse and often nonspecific, which poses a diagnostic challenge. Intra-abdominal lymphangiomas may present with symptoms that vary according to the lesion's size and anatomical location. Case reports describe presentations mimicking acute abdomen-whether obstructive, subocclusive, ischemic, or inflammatory in nature [5, 6]. Some lesions are discovered incidentally [7], while others present with subtle, insidious symptoms, such as chronic refractory iron-deficiency anemia [8,9].Here, we report an unusual clinical presentation of a small intestinal and mesenteric lymphangioma, manifesting as chronic iron-deficiency anemia refractory to iron supplementation, associated with non-bloody mucous diarrhea, in the presence of normal upper gastrointestinal endoscopy and colonoscopy findings. Although lymphangiomas typically follow a benign course, their size and location may lead to significant complications. Surgical resection is generally curative when complete excision is achieved, although rare cases of recurrence have been reported [10].

3. Case Report

A 52-year-old male presented to a tertiary care hospital in southern Brazil with a 10-day history of non-bloody, non-mucous diarrhea, occurring up to 15 times per day. He had previously used antiparasitic agents and antibiotics (ciprofloxacin and metronidazole), without clinical improvement. Additionally, he reported a history of iron-deficiency anemia diagnosed three months earlier, refractory to intravenous iron supplementation.Apart from anemia, laboratory investigations were within normal limits, and tumor markers were not assessed at that time. Upper gastrointestinal endoscopy and colonoscopy revealed no abnormalities.The patient also reported intentional weight loss over the previous six months due to dietary changes. He denied fever or vomiting, and his physical examination was unremarkable.Abdominal computed tomography (CT) revealed a segment of small bowel in the mesogastric region with subtle irregular parietal thickening, associated with adjacent mesenteric infiltration. There was also a large area of mesenteric density with a nodular appearance measuring 9.6 × 7.0 cm, in contact with distal mesenteric vessels, but without signs of vascular obstruction. Additional findings included simple renal and pancreatic cysts, two indeterminate hypodense splenic lesions, and a hepatic hemangioma.Subsequent abdominal magnetic resonance imaging (MRI) identified multiple nodular formations, the largest located at the root of the mesenteric vessels, suggestive of lymph node conglomerates, raising suspicion for lymphoproliferative disease. The exam also revealed irregular thickening of a jejunoileal loop spanning approximately 6 cm.Given the imaging findings and differential diagnosis of abdominal neoplasia—particularly lymphoproliferative disease or carcinoid tumour-a diagnostic video laparoscopy was performed. Intraoperative findings included thickened omental tissue with mucinous-like changes, a segment of small bowel with tumoral areas characterized by reddish vesicular lesions,

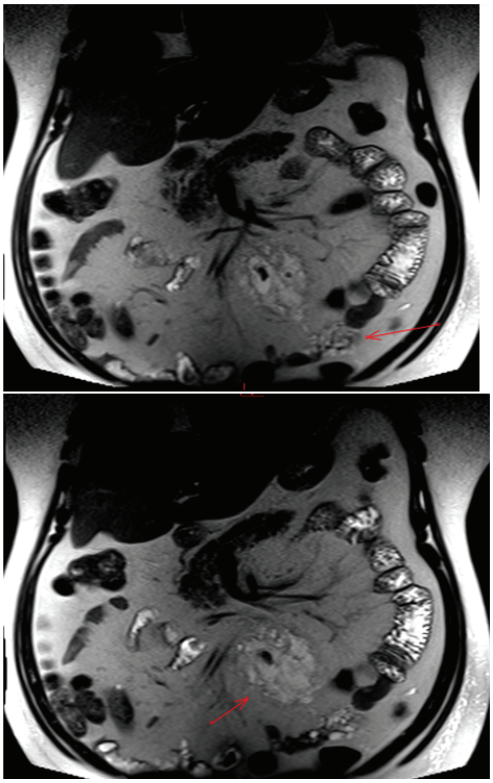

Figure 1: T2-weighted nuclear magnetic resonance image: red arrows indicate thickening of the small intestine in the first image and nodular formations in the root of the mesentery in the second.

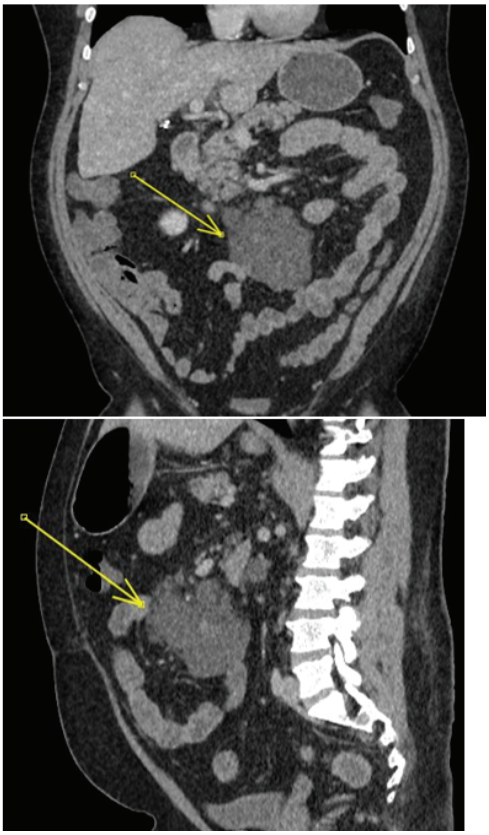

Figure 2: CT Scan: the yellow arrows show a segment of small intestine with slight irregular parietal thickening, in the mesogastric region, associated with infiltration and thickening of the adjacent mesentery.

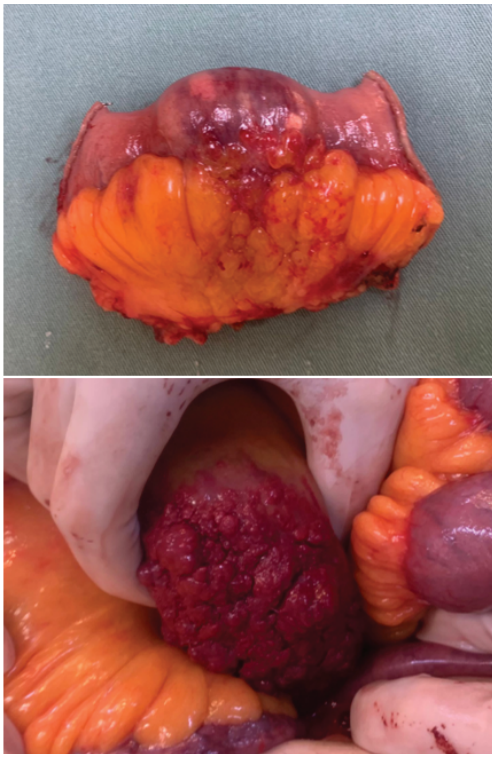

Figure 3: The first image shows a surgical piece from the enterectomy and image 2 shows a nodular-shaped conglomerate biopsied at the root of the mesentery.

and a 9 cm cystic mass at the mesenteric root resembling the jejunal wall thickening.Due to the complexity of the lesions, conversion to an open approach was undertaken. Segmental resection of the affected small intestine and omental biopsy were performed. Histopathological analysis of both specimens revealed findings consistent with enteric lymphangioma, which was subsequently confirmed by immunohistochemical staining.

4. Discussion

Lymphangioma is a benign congenital malformation of the lymphatic system, most frequently diagnosed in childhood. The most common anatomical sites are the cervical, axillary, and craniofacial regions [11,12]. Although rare, lymphangiomas can also occur in the abdominal cavity, particularly among pediatric patients. The literature describes a wide range of abdominal presentations in children, including abdominal distension, pain, signs of intestinal obstruction, and lower limb lymphedema. For instance, Kondo et al. [13] reported multiple intestinal lymphangiomas associated with episodic gastrointestinal bleeding in pediatric patients [9], while Protopapas et al. (2005) described a case of mesenteric lymphangioma mimicking adnexal torsion [7]. Other studies emphasize the importance of early diagnosis and appropriate management to avoid complications [7].In contrast, the clinical presentation of small intestinal lymphangioma in adults is highly variable and often nonspecific, making diagnosis more challenging. According to a study by Chin et al. [14], abdominal pain is the most frequent symptom, occurring in 77.78% of adult cases. Additionally, 27.78% of patients presented with a palpable abdominal mass, and 16.67% experienced intestinal bleeding, manifesting as melena or hematochezia [3].Many adult patients are asymptomatic, with lymphangiomas

being discovered incidentally during imaging or endoscopic evaluations. Wang et al. [4]. Performed a retrospective study of 15 patients in China and found that most were asymptomatic, with lesions predominantly located in the duodenum. These were identified by endoscopy showing characteristic white spots [4].In rarer cases, systemic symptoms such as dizziness, fatigue, and anemia may occur, suggesting protein-losing enteropathy. This was described by Safatle-Ribeiro et al. [5] in a Brazilian case [5]. Intussusception, although less frequent, can present with acute abdominal pain, vomiting, and a palpable mass, as reported by Zhao et al. [6]. More severe complications, such as volvulus, may cause rapid abdominal distension and vomiting, sometimes requiring emergency surgery, as seen in a case described by in Brazil [10].Imaging plays a critical role in the evaluation of abdominal lymphangiomas. Gorelick et al. [11], in an analysis involving seven pediatric cases across three university hospitals, highlighted the importance of ultrasonography (US) and computed tomography (CT) in preoperative assessment. US revealed multiloculated abdominal cystic masses in five of six cases and ascites in three. CT consistently showed septate cystic masses of various sizes, with serial imaging demonstrating progressive enlargement and signs of complications requiring urgent intervention [9]. Given the wide spectrum of presentations-both in children and adults-lymphangiomas may mimic other gastrointestinal disorders. Thus, imaging and histopathological evaluation are essential for definitive diagnosis. Surgical resection remains the primary treatment strategy in symptomatic patients or when diagnostic uncertainty exists, providing a definitive solution in most cases [1-9].

5. Conclusion

Lymphangioma is an extremely rare condition in the adult population, with involvement of gastrointestinal tract organs being even more uncommon. The lack of specific symptoms makes diagnosis particularly challenging, underscoring the importance of imaging studies in the diagnostic process.
In cases where imaging is inconclusive or raises suspicion of malignancy, biopsy or diagnostic laparoscopy should be considered to achieve a more accurate diagnosis.Although lymphangioma is a benign entity, the decision to proceed with surgical resection should be individualized, taking into account the patient's clinical presentation and the presence of symptoms.

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