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Acute Intestinal Infarction Due to Diffuse Jejunoileal and Mesenteric Lipomatosis in a 39-Year-Old Woman

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Study Design A
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Patient: Female, 39-year-old
Final Diagnosis: Acute intestinal infarction
Symptoms: Painful
Medication: —
Clinical Procedure: Intestinal resection
Specialty: Surgery

Objective: Unusual clinical course

Background: Although lipomas are common benign tumors of adipose tissue, diffuse lipomas involving the small bowel, large bowel, and mesentery are rare. Multiple non-encapsulated lipomas characterize diffuse intestinal and mesenteric lipomatosis. Intestinal lipomatosis can be asymptomatic or may result in complications such as intussusception, volvulus, intestinal obstruction, or hemorrhage due to mucosal ulceration. A rare case is presented of intestinal infarction due to diffuse segmental jejunoileal and mesenteric lipomatosis in a 39-year-old woman.

Case Report: A 39-year-old woman was admitted to the emergency department with a 12-hour history of diffuse abdominal pain, nausea, vomiting, and absent bowel movements. She had a known history of intestinal lipomatosis, diagnosed two years previously on abdominal computed tomography (CT) imaging. At surgery, segmental jejunoileal and mesenteric lipomatosis was identified associated with acute intestinal infarction. She underwent ileal resection with side-to-side enterocolic anastomosis.

Conclusions: Diffuse intestinal and mesenteric lipomatosis is a rare condition that can be associated with complications. To our knowledge, this is the first reported case to present with acute small bowel infarction.

MeSH Keywords: Lipoma • Lipomatosis • Mesenteric Vascular Occlusion**Full-text PDF:** <https://www.amjcaserep.com/abstract/index/idArt/922830>

Background

Intestinal lipomatosis is a rare condition that was first described by Hellström in 1906 as ‘multiple lipomatous formations’ [1]. However, intestinal lipomatosis is a poorly defined condition that consists of a diffuse overgrowth and infiltration of well-differentiated fatty tissue in the submucosal layer of the small and large bowel [2]. The condition has also been defined as the occurrence of more than four encapsulated submucosal lipomas [3].

Intestinal lipomatosis is usually asymptomatic, and no treatment is required unless it is complicated with intussusception, bleeding, obstruction, or bowel perforation [4]. A rare case is presented of intestinal infarction due to diffuse intestinal and mesenteric lipomatosis in a 39-year-old woman, treated by ileal resection and side-to-side enterocolic anastomosis.

Case Report

A 39-year-old woman was admitted to the emergency department with a 12-hour history of diffuse abdominal pain, nausea, vomiting, and absent bowel movements. She had a known history of intestinal lipomatosis, diagnosed two years previously on abdominal computed tomography (CT) imaging (Figure 1), confirmed by video capsule endoscopy and biopsies of the terminal ileum. Her past medical history also included mild chronic anemia of unknown origin since childhood. The patient described postprandial colic and diarrhea in the previous six months and 10 kg weight loss.

On physical examination, the patient was anxious, with a pulse rate of 110 beats per minute. Her abdomen was distended, with diffuse tenderness and guarding, and was painful on palpation. Digital rectal examination detected a lack of stool in the rectal vault. Laboratory tests showed a white blood cell (WBC) count of 27,000/ μ l and a hemoglobin level of 12 g/dl. Plain abdominal radiography (Figure 2) was performed with the patient in a sitting position due to her intolerance to being upright. Abdominal X-ray showed multiple jejunoileal and colonic air-fluid levels and minimal intramural intestinal gas in the inferior half of abdominal cavity.

Emergency laparotomy was performed due to the severity of the symptoms. The intraoperative findings showed approximately 250 cm of grossly enlarged and thickened jejunoileal loops with diffuse yellow deposits of fatty tissue and an infarcted intestinal loop of approximately 30 cm (Figures 3, 4). The mesentery contained multiple diffuse nodules measuring between 5–15 cm in diameter.

Because the patient was relatively young and there were parts of the bowel that appeared healthy with no signs of ischemia,



Figure 1. Abdominal computed tomography (CT) imaging shows lipomatous infiltration of the small bowel wall. The white arrows indicate the areas of lipomatous change.



Figure 2. Abdominal X-ray image with the patient in the seated position. Multiple jejunoileal and colonic air-fluid levels.

jejunoileal enterectomy of the whole lipomatous bowel (250 cm) was performed with anastomosis of remaining small intestine to the ascending colon. Macroscopic examination showed 30 cm of the intestine that was brown-black with white focal deposits on the serosa. Histopathology of the infarcted segment showed a fibrinous acute inflammatory exudate, with focal necrosis and transmural hemorrhage, edema, neutrophil infiltrate. The remaining part of the resected bowel segment (approximately 220 cm) showed diffuse proliferation of mature

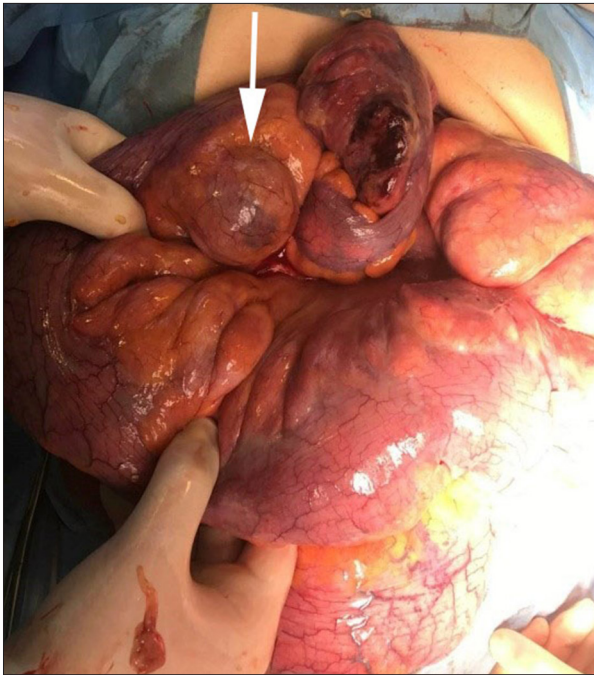


Figure 3. The findings during surgery in a 39-year-old woman with acute intestinal infarction due to diffuse intestinal and mesenteric lipomatosis. At surgery, dilated and thickened bowel loops are shown. The white arrow indicates a diverticulum.

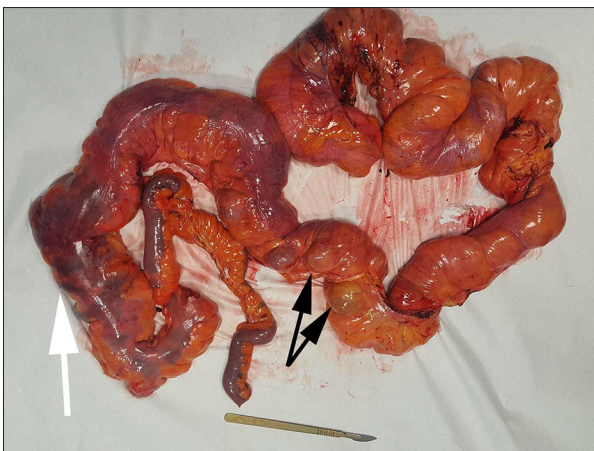


Figure 4. The surgical resection specimen in a 39-year-old woman with acute intestinal infarction due to diffuse intestinal and mesenteric lipomatosis. The surgical specimen of resected ileum is shown following ileal resection and side-to-side enterocolic anastomosis. The white arrow indicates an ischemic intestinal segment. The black arrows indicate intestinal diverticula.

fat cells, without encapsulation, within the submucosa and subserosa (Figure 5). There were four diverticula on the mesenteric border of the small bowel and multiple lipomas in the mesentery. Postoperatively, the patient experienced frequent

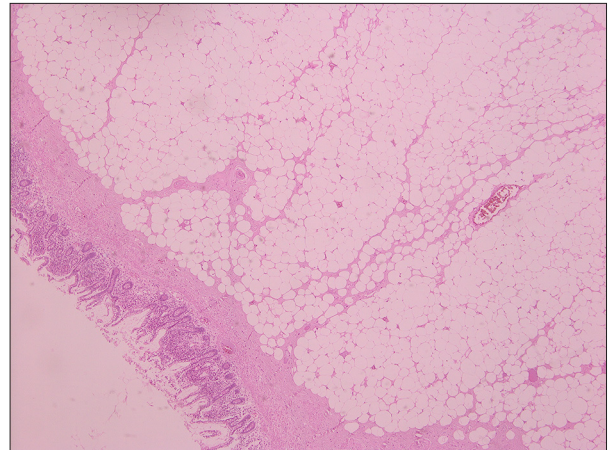


Figure 5. Photomicrograph of the histopathology of the bowel. The histology shows intestinal submucosal infiltration with fatty tissue, consistent with benign lipomatosis. Hematoxylin and eosin (H&E). Magnification $\times 4$.

stools (6–8 stools per day) and was treated with loperamide. After three months of follow-up, her bowel function returned to normal, and she began to gain weight.

Discussion

Abnormal deposits of fat in the intestine are classified morphologically as isolated lipoma, multiple circumscribed lipomas, diffuse nodular lipomatosis, and diffuse adipose tissue infiltration of the submucosa without tumor formation [5]. Solitary lipomas are the second most common form of benign intestinal tumors, with an incidence at autopsy ranging from 0.04–4.5% [6]. The other three forms are rarely encountered, with multiple lipomas in approximately 5% of lipoma cases [2], and diffuse intestinal adipose tissue infiltration is extremely rare. Intestinal lipomatosis shows no gender predilection and occurs at a mean age of 47.3 ± 18.4 years [7]. The lesions of intestinal lipomatosis are usually limited to the submucosa, but may extend to serosa and mesenteric fat [8]. The muscularis propria was reported to be affected in one case [9]. Intestinal lipomatosis usually involves the ileum in 39% of cases, both the ileum and jejunum in 39% of cases and the jejunum alone in 13% of cases [9]. The etiology of intestinal lipomatosis remains unknown. The possible etiological factors include degenerative disease with disturbance of fat metabolism, embryological ectopic adipose tissue, chronic inflammatory bowel disease, low-grade infection [10], fat deposition following chemotherapy [11], and hamartoma syndromes [12].

Intestinal lipomatosis may be associated with the depositions of fat in other tissues and organs and may be a component of genetic syndromes that include Cowden syndrome (multiple mucocutaneous hamartomas and a high risk of developing

tumors), Bannayan–Riley–Ruvalcaba syndrome [13], Proteus syndrome (rapid overgrowth of multiple types of tissue), neurofibromatosis type 1, or Madelung disease (multiple symmetric lipomatosis) [11]. The cells of gastrointestinal stromal tumor (GIST) and lipoma cells may share common genetic abnormalities [14]. Previous reports that describe isolated intestinal lipomatosis, without associated cutaneous or other lipomas, have shown it to be a clinically distinct entity from Madelung disease [15]. However, isolated mesenteric lipomatosis may be a manifestation of abdominal symmetric lipomatosis, a variant of the multiple symmetric lipomatosis [16]. Factors associated with mesenteric lipomatosis have been reported to include steroid treatment [17], alcohol abuse [18], metabolic abnormalities, and polyneuropathy [19].

Clinically, intestinal lipomatosis is characterized by a lack of symptoms. Rarely, patients may present with abdominal pain, nausea, melena, diarrhea, constipation, intermittent vomiting, a palpable mass, and weight loss [20]. The occurrence of both intestinal lipomatosis and diverticulosis has been reported, due to fat infiltration and weakening of the bowel wall. The presence of diverticulosis can trigger bacterial overgrowth in the intestine, which may result in malabsorption [21]. The patient described in this report had four small bowel diverticula. Therefore, the presence of mild anemia in this patient can be a consequence of malabsorption. The diagnosis of intestinal lipomatosis, as well as of other tumors of the small bowel, is difficult to establish. Barium enema alone has a sensibility of 35%, which can increase to 90% if performed using the enteroclysis technique [22]. In cases of extended mesenteric lipomatosis, fat accumulation can simulate ascites and pneumoperitoneum [18]. Ultrasonography of the abdomen may visualize tumors larger than 4 cm [13] that present as hyperechoic lesions, although a reliable ultrasonic characterization of fatty lesions is difficult [23]. Typical manifestations of fatty lesions on CT are symmetrical and well-defined masses with a density ranging from –80 to –120 Hounsfield units (HU), compatible with fatty tissue [24]. The differential diagnosis of a fatty mass includes liposarcoma and lipoblastomatosis [25]. When longstanding symptoms are present, and imaging modalities are negative, capsule endoscopy or balloon-assisted enteroscopy may be performed [26]. The advantages of enteroscopy include real-time observation, the ability to obtain a tissue biopsy, the precise localization of lesions, and the possibility of interventional treatment [27].

Endoscopically, the characteristic features of intestinal lipoma is the ‘pillow’ or ‘cushion’ sign, which is a depression formed in the lipoma by pushing upon it with forceps and the recovery of its original shape upon withdrawal of forceps. Another endoscopic feature is the ‘tenting sign,’ in which the overlying mucosa can be pulled away from submucosa to produce a tent-like appearance [28]. A biopsy is seldom useful because

the lesion lies beneath the normal intestinal mucosa [29]. However, the biopsy may contribute to the enucleation of the lipoma through the damaged mucosa, the so-called phenomenon of ‘self-amputation’ of lipoma [30].

The management of intestinal lipomatosis consists of follow-up alone if the patient is asymptomatic. Endoscopic removal may be performed if the lipoma is smaller than 2.5 cm [28]. If the lipoma is greater than 2.5cm, local open excision and plastic repair [31], or limited resection of the intestine that contains the largest concentration of lipomas may be performed [28].

Endoscopic procedures comprise mucosal or submucosal resection, the use of the unroofing technique, and the application of an endo-loop or endo-clip [32]. In cases of symptomatic diffuse fatty infiltration of the submucosa, surgical resection is the only treatment option. If surgical resection is performed, it is important to conserve at least 120 cm of intestinal loops so that the short bowel syndrome is prevented. The most frequent complications of intestinal lipomatosis are volvulus, intussusception, and perforation [20]. Perforation may occur because of the relative avascularity of the lipomatous lesion, which predisposes the overlying mucosa to ischemia leading to perforation [29].

Acute intestinal infarction of a bowel segment associated with intestinal lipomatosis has not been previously reported. This complication may be the result of volvulus of the affected intestine [33]. However, in this case, the likely cause of intestinal infarction was extrinsic compression of the vascular pedicles by the lipomatous nodules. Furthermore, the first appearance of postprandial colicky pain accompanied by diarrhea occurred six months before the presentation and was followed by weight loss, which may suggest chronic ischemic injury, complicated by an acute episode of infarction. Chronic mesenteric injury may result from extrinsic vascular compression by tumors, which progresses to acute mesenteric ischemia, if untreated [34]. However, it is difficult to exclude other possible causes of intestinal ischemia such as arterial embolism, arterial thrombosis, and mesenteric venous thrombosis, as no CT scan was performed before the intervention. The absence of cardiovascular comorbidities and coagulation disorders, in this case, supports that extrinsic compression was the trigger of intestinal ischemia.

When acute bowel ischemia is diagnosed, the resection of the affected small bowel with a proximal ostomy and secondary restoration should be performed to avoid anastomotic breakdown. In this case, at laparotomy, the remaining intestinal loops appeared viable. Because of the patient’s age and to preserve her quality of life, ileal resection and side-to-side enterocolic anastomosis were performed.

Conclusions

A rare case is presented of intestinal infarction due to diffuse intestinal and mesenteric lipomatosis in a 39-year-old woman. Because this condition is rare, the diagnosis may be missed. In patients with a diagnosis of intestinal lipomatosis following abdominal imaging, follow-up with awareness of the potential complications are recommended. As this case has shown, the treatment of choice in cases with complications is resection of the affected segment of the intestine.

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Conflict of interest

None.