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CASE REPORT / OLGU SUNUMU

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Intestinal Lipomatosis of the Ileum: A Rare Cause of Small Bowel Perforation

İleumun İntestinal Lipomatozisi: İnce Bağırsak Perforasyonunun Nadir Bir Nedeni

Ömer Atmış[1], Semin Ayhan[1], Peyker Temiz[1], Hanife Seda Mavili[1], Halil İbrahim Tanrıverdi[2]

[1]Manisa Celal Bayar University Faculty of Medicine, Department of Pathology, Manisa, Türkiye
[2]Manisa Celal Bayar University Faculty of Medicine, Department of Pediatric Surgery, Manisa, Türkiye

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Abstract

Intestinal lipomatosis is characterized by submucosal infiltration of mature adipose tissue, which is usually asymptomatic and can be detected incidentally. However, it may cause complications, such as intussusception, bleeding, and perforation. We describe a 12-year-old female patient who underwent surgery after the detection of intestinal perforation on abdominal computed tomography. Histopathologic examination of segmentary small bowel resection revealed a lesion predominantly located in the submucosa, occasionally causing atrophy in the muscle tissue and extending to the serosa. The lesion consisting of mature lipocytes was completely unencapsulated. Based on these findings, the patient was diagnosed with intestinal lipomatosis. Intestinal lipomatosis is usually asymptomatic and can be detected incidentally. However, it can lead to complications, such as intussusception, bleeding, and perforation, and these clinical conditions may be the first signs of the lesion, as in our case.

Keywords: Intestinal lipomatosis, ileum, intestinal perforation, intussusception

Öz

İntestinal lipomatozis, genellikle asemptomatik olan ve insidental olarak saptanan, tümör oluşumu olmaksızın matür adipöz dokunun submukozal infiltrasyonu ile karakterize lezyonlardır. Ancak invajinasyon, kanama ve perforasyon gibi komplikasyonlara neden olabilir. Bu olgu sunumunda, batın bilgisayarlı tomografisinde bağırsak perforasyonu saptanması üzerine opere edilen 12 yaşında kız hasta sunulmaktadır. Segmenter ince bağırsak rezeksiyonunun histopatolojik incelemesinde ağırlıklı olarak submukoza yerleşimli, yer yer kas dokuda atrofiye neden olan ve serozaya uzanım gösteren lezyon gözlenmiştir. Matür lipositlerden oluşan lezyon bütünüyle kapsülsüz görünümdedir. Bu bulgularla olguya intestinal lipomatozis tanısı konulmuştur. İntestinal lipomatozis genellikle asemptomatiktir ve rastlantısal olarak tespit edilir. Ancak invajinasyon, kanama, perforasyon gibi komplikasyonlara yol açabilmekte ve bu klinik durumlar bizim olgumuzda olduğu gibi lezyonun ilk belirtisi olabilmektedir.

Anahtar Kelimeler: İntestinal lipomatozis, ileum, intestinal perforasyon, intussusepsiyon

Introduction

Intestinal lipomatosis refers to lesions characterized by submucosal infiltration of mature adipose tissue without the formation of a tumor[1]. Unlike lipoma, intestinal lipomatosis is not encapsulated lesions[2]. Adipose tissue proliferation and deposition are usually limited to the submucosa but may also extend to the serosa and mesenteric adipose tissue[3]. The cause of fat deposition is unclear[4]. Intestinal lipomatosis does not show sex predominance and usually occurs after the fourth decade of life[5]. The most common

Address for Correspondence/Yazışma Adresi: Ömer Atmış MD, Manisa Celal Bayar University, Faculty of Medicine, Department of Pathology, Manisa, Türkiye
E-mail: omeratmis@hotmail.com
ORCID ID: orcid.org/0000-0003-4789-0875

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location of lipoma is the colon (65-75%), while the ileum is the second most common location[6]. In the literature, no predilection site for lipomatosis has been described[1]. Intestinal lipomatosis is usually asymptomatic and can be detected incidentally. However, it may cause complications, such as intussusception, bleeding, and perforation, and these clinical manifestations may be the first signs of the lesion[7]. Lipomatosis of the gastrointestinal tract is rare, and only few cases have been reported[1]. Because it is a rare entity and, as in our case, complications, such as perforation, may be the first presentation finding, we herein report a case of intestinal lipomatosis.

Case Report

A 12-year-old girl presented to the emergency department with complaints of vomiting and abdominal pain that persisted for 2 days. It was explained that the patient had previously been followed up for chronic constipation. The examination revealed no findings except minimal tenderness in the umbilical region. The patient was operated upon the detection of intestinal perforation on abdominal computed tomography. Segmentary small bowel resection was performed because of the large perforation area in the middle part of the ileum and stool in the abdomen during the operation. In the gross examination of the small bowel resection, a perforation area of 7.5 cm in diameter was observed, and the segment adjacent to the perforation was found to have a dilated appearance. The mucosa of the dilated segment was hyperemic in places, with an area of approximately 3 cm in diameter, paler color, and flattening of the mucosa. In the cross-section of this area, the intestinal wall was yellow under the mucosa (Figure 1). Microscopic examination revealed mucosal ischemic changes in the dilated segment. In areas adjacent to the perforation, where the mucosa was flattened and pale in appearance, a lesion was observed predominantly located in the submucosa, occasionally causing atrophy in the muscle tissue and extending to the serosa. The lesion, consisting of mature lipocytes, including congested vessels and connective tissue areas, was completely unencapsulated (Figure 2).

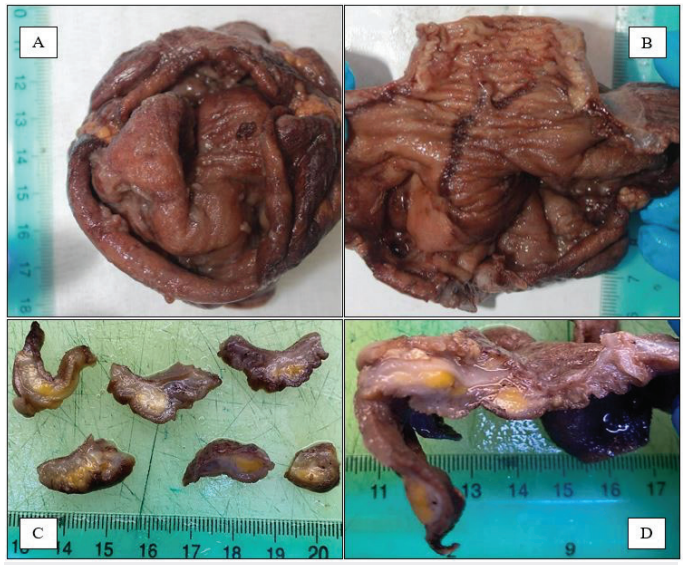

Figure 1. Perforation area with a diameter of 7.5 cm (A), hyperemic mucosa in the dilated segment and the adjacent pale-looking area (B), and cross-section of the pale-looking area (C, D)

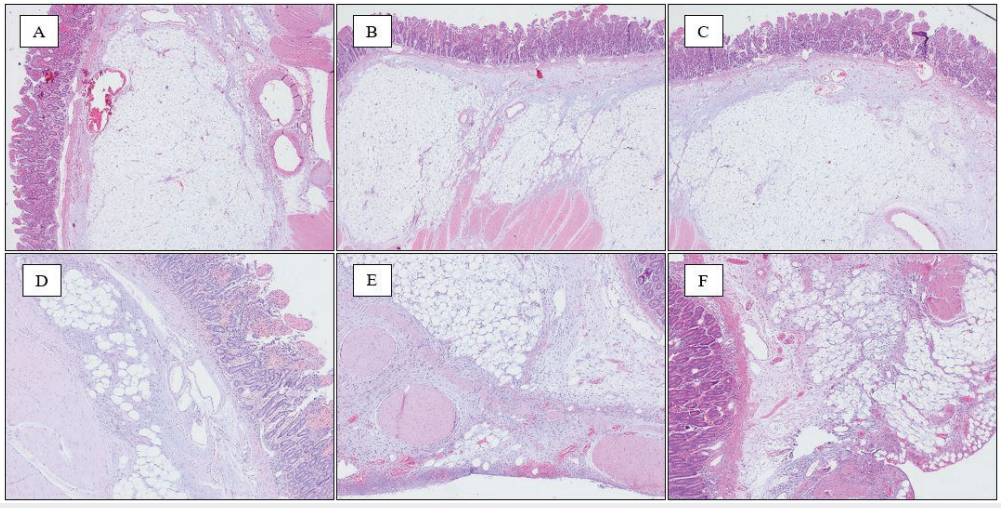

Figure 2. A lesion consisting of mature lipocytes in the submucosa (A, B; x20), unencapsulated lesion, and ischemic changes in the superficial mucosa (C, D; x20, x40), areas where the lesion causes muscle tissue atrophy and extends to the serosa (E, F; x40)

In many samples of the material, submucosal adipose tissue was observed only in this segment. Based on these findings, the patient was diagnosed with intestinal lipomatosis. Informed consent was obtained.

Discussion

Abnormal deposition of mature adipose tissue in the intestine occurs in four different ways. The first category includes single and well-circumscribed lipomas, while the second category includes cases that present as multiple well-circumscribed lipomas. The third type of abnormal adipose tissue deposition is diffuse nodular lipomatosis, which is characterized by numerous nodules growing into the lumen leading to an increase in the irregular intestinal wall thickness. The fourth form, called intestinal lipomatosis, refers to lesions characterized by submucosal infiltration of mature adipose tissue without tumor formation[8]. Although intestinal lipomatosis is usually asymptomatic and can be detected incidentally, it can lead to complications, such as intussusception, bleeding, and perforation, and these clinical conditions may be the first signs of the lesion[7]. The most common symptoms in symptomatic patients are non-specific abdominal pain, constipation, diarrhea, and bleeding[2]. Although our case was followed up with a complaint of chronic constipation, surgical treatment was applied because of ileum perforation, which was only diagnosed in this way. Due to its rarity, there are no current guidelines for the selection of imaging modalities for intestinal lipomatosis. Surgical treatment may be required in patients who present with complications and do not respond to conservative measures[9].

The term "intestinal lipomatosis" is used by some authors to describe cases of multiple lipomas[6,10,11]. However, in some publications, this refers to unencapsulated lesions characterized by submucosal infiltration of mature adipose tissue without tumor formation, unlike lipomas[2]. We propose that these two distinct morphological appearances may represent separate entities with different pathogenesis: a neoplastic process and a developmental abnormality. This terminological confusion could be clarified through the reporting of new cases and a better understanding of the pathological and molecular features distinguishing the two groups.

Ethics

Informed Consent: Informed consent was obtained.

Footnotes

Authorship Contributions

Surgical and Medical Practices: H.İ.T., Concept: Ö.A., S.A., Design: Ö.A., S.A., Data Collection or Processing: Ö.A., S.A., P.T., H.S.M., Analysis or Interpretation: Ö.A., S.A., P.T., Literature Search: Ö.A., P.T., H.S.M., Writing: Ö.A., S.A., P.T.

Conflict of Interest: No conflict of interest was declared by the authors.

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