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CASE REPORT

Adult Intussusception Caused by Cecal Lymphangioma: A Case Report

Çekal Lenfanjioma Bağlı Erişkin İnvajinasyon: Olgu Sunumu

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ABSTRACT

A 23-year-old female was admitted to our hospital with abdominal pain and diarrhea. Preoperative abdominal ultrasonography and computed tomography revealed ileocecal intussusception. Emergency laparotomy was performed due to escalation of the patient's abdominal pain and development of acute abdomen. During the laparotomy, we found an ileocecal intussusception and solid mass in the cecum, and an ileocecal resection was performed. Histopathological examination showed cystic lymphangioma in the cecum. This case emphasizes that lymphangioma, a rare benign tumor, should be considered in the differential diagnosis of intussusception.
Keywords: Cecum, lymphangioma, intussusception

ÖZ

Yirmi üç yaşında kadın hasta karın ağrısı ve ishal şikayeti ile hastanemize başvurdu. Preoperatif abdomen ultrasonografi ve bilgisayarlı tomografide ileoçekal invajinasyon saptandı. Hastanın karın ağrısı şiddetlenmesi ve akut batın gelişmesi üzerine acil laparotomi yapıldı. Laparotomide ileoçekal invajinasyon ve çekumda sert kitle saptanması üzerine ileoçekal rezeksiyon yapıldı. Histopatolojik incelemede çekumda kistik lenfanjiom saptandı. Bu olgu, nadir görülen ve benign bir tümör olan lenfanjiomun invajinasyon olgularında ayırıcı tanıda göz önünde bulundurulması gerektiğini vurgulamaktadır.
Anahtar Kelimeler: Çekum, lenfanjiom, invajinasyon

Introduction

Lymphangioma is a benign congenital tumor in the lymphatic system arising from a rare developmental anomaly. It is usually seen in childhood, with 80-90% of cases detected in the first five years of life, and rarely occurs in adulthood. They are most commonly located in the head, neck, and axilla. In rare cases they may also be found in the abdomen.[1,2] Gastrointestinal involvement occurs in less than 1% of cases and can cause bowel invagination. Intussusception is rare in adults, accounting for approximately 1-3% of all bowel obstructions.[3] In this report, we present a rare case of ileocecal intussusception due to intestinal lymphangioma in an adult patient.

Case Report

A 23-year-old woman presented to our emergency department with complaints of abdominal pain and diarrhea starting one week earlier. Her medical and family histories were unremarkable. On physical examination, she exhibited right lower quadrant abdominal tenderness. Her blood pressure was 100/60 mmHg, pulse was 96/min, and body temperature was 36.7 °C. Laboratory tests were normal except for elevated C-reactive protein level (2.9 mg/dL). Air-fluid level was observed in the small intestine on standing anteroposterior X-ray. Abdominal ultrasound and computed tomography showed that the terminal ileum was invaginated into the cecum (Figure 1). Emergency laparotomy was performed due to escalation

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of the patient's abdominal pain and development of acute abdomen. The ileocecal intussusception and a solid mass in the cecum were detected during the laparotomy. After manual reduction, we observed that the invaginated small intestine loop had diminished perfusion, and ileocecal resection was performed. On macroscopic examination of the resected material, a 4x4x2 cm polypoidal lesion was observed at the cecal base, ulcerating the mucosa but limited to the submucosa (Figure 2). Microscopic examination revealed an endothelium-lined cyst divided by fibrous septa under a normal mucosal membrane (Figure 3). Histopathological diagnosis was reported as cystic lymphangioma.

The patient was discharged on postoperative day 6. No recurrence was detected in colonoscopy or computed tomography in follow-up at postoperative 3 and 9 months.

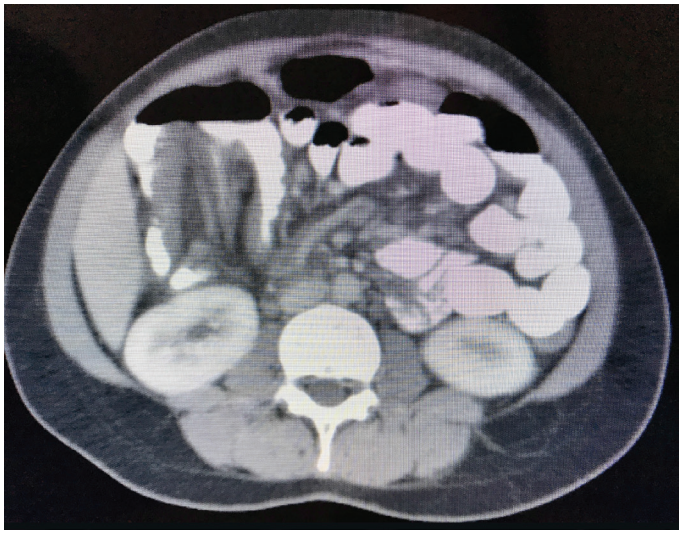

Figure 1. Abdominal computed tomography axial image shows ileocecal intussusception

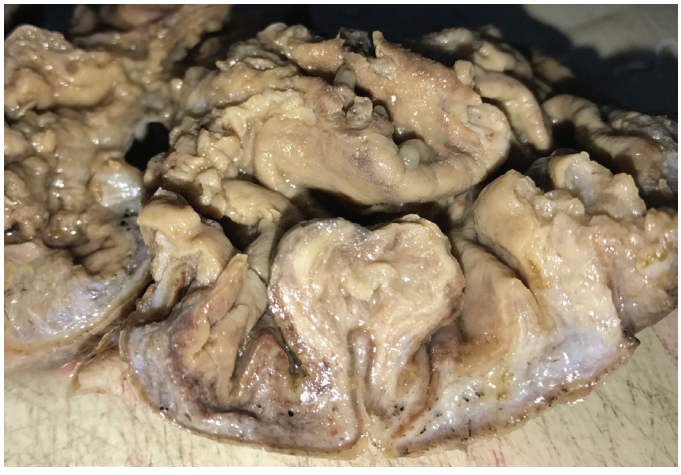

Figure 2. Macroscopic examination of the resected material

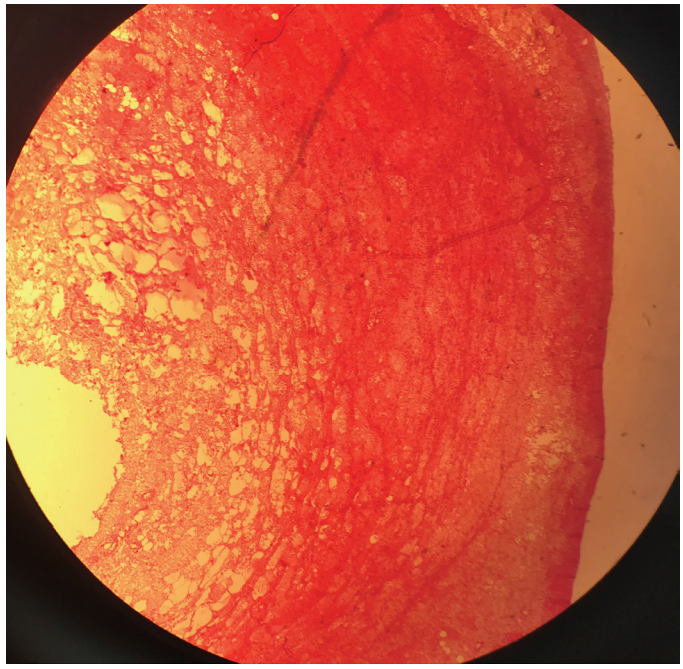

Figure 3. Microscopic examination of the resected material

Discussion

Lymphangiomas are benign congenital tumors of the lymphatic system and are divided into three classes: capillary, cystic, and cavernous.[4] Cystic lymphangiomas are the most common. Cystic lymphangiomas have large endothelium-lined cystic spaces, often with thick walls, and collagen and smooth muscle are present at various proportions. Patients with lymphangioma are usually asymptomatic. Symptomatic patients may experience abdominal pain, vomiting, diarrhea, constipation, obstruction, invagination, lower gastrointestinal hemorrhage, and protein-losing enteropathy.[5] The diagnosis of intraabdominal lymphangioma can be facilitated by barium enema, colonoscopy, endoscopic ultrasonography, abdominal computed tomography and magnetic resonance imaging. In cases where a diagnosis cannot be established with imaging methods, diagnostic laparoscopy should be considered despite its invasive nature. Especially in female patients, diagnostic laparoscopy can be used both for diagnosis and treatment. We used abdominal ultrasonography and computed tomography for diagnostic purposes in the present case. Regression of lymphangiomas is unlikely. The recommended treatment is endoscopic polypectomy and mucosal resection for lesions smaller than 2 cm intraluminal, and surgical resection for lesions larger than 2 cm.[6,7,8,9] Due to the development of intussusception and acute abdomen in our case, we performed laparotomy and surgical resection. Postoperative examination revealed the cecal lymphangioma was 4 cm in size, making our treatment

approach consistent with the literature. In the literature, the first colonic lymphangioma was described by Chisholm and Hillkowits[10]. Only six cases of intussusception due to colonic lymphangioma have been reported in adults; five of them were located in the ascending colon and one was in the transverse colon.[11,12,13,14,15,16] As in our case, all six of those patients were female and had lesions over 4 cm in size, and all were treated with laparotomy and surgical resection. Lymphangioma, which is rare in adulthood, is generally detected by colonoscopy and endoscopic ultrasonography and treated when small. However, it must be kept in mind that in the rare cases where lymphangiomas reach large sizes, they may cause obstruction and intussusception and require treatment by surgical resection.

Ethics

Informed Consent: Consent form was filled out by all participants.

Peer-review: Internally peer-reviewed.

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

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