CASE REPORT

TJG
The Turkish Journal of Gastroenterology

Huge cystic lymphangioma mimicking ovarian malignancy: A case report

Çağlar YILDIZ[1], Kürşat KARADAYI[2], Cihat ŞARKIŞ[3], Ali ÇETİN[1]

Departments of [1]Obstetrics and Gynecology, [2]General Surgery and [3]Internal Medicine, Cumhuriyet University School of Medicine, Sivas

Lymphangiomas are rare benign congenital malformations of the lymphatic system. Clinical presentation varies from asymptomatic masses to acute abdominal pain. A 25-year-old female who presented with acute onset of abdominal distension and severe pelvic pain is presented. As a palpable mass was found on the pelvic examination and ultrasonography demonstrated a hypoechoic cystic mass, an ovarian malignancy was suspected. Exploratory laparotomy was performed and revealed cystic lymphangioma of the sigmoid colon. The clinical, radiological and pathological findings of the patient are discussed with a brief review of the literature.

Key words: Cystic lymphangioma, sigmoid colon, pelvic mass

Ovaryan malignensiyi taklit eden dev kistik lenfanjiyom: Bir olgu sunumu

Lenfanjiyomlar lenfatik sistemin benign konjenital malformasyonlarıdır. Klinik prezentasyon asemptomatik kitleden akut abdominal ağrıya kadar değişkenlik gösterir. Çalışmamızda akut başlangıçlı abdominal distansiyon ve şiddetli pelvik ağrı ile başvuran 25 yaşında kadın olgu sunulmaktadır. Pelvik muayenede palpe edilebilen bir kitle saptandığı ve ultrasonografide hipoekoik kistik bir kitle görüldüğü için ovaryan bir malignensiden şüphelenildi. Eksploratif laparatomi uygulandı ve sigmoid kolona ait kistik lenfanjiyom saptandı. Olgunun klinik, radyolojik ve patolojik bulguları literatürün kısaca gözden geçirilmesi eşliğinde tartışılmaktadır.

Anahtar kelimeler: Kistik lenfanjiyom, sigmoid kolon, pelvik kitle

INTRODUCTION

Lymphangiomas are rare benign congenital malformations of the lymphatic system. They are thought to occur due to obstruction of the local lymph flow (1). Approximately 95% of lymphangiomas are found in the head, neck and axilla, and 5% appear in other parts of the body such as the lungs, pleura, pericardium, esophagus, stomach, jejunum, colon, pancreas, liver, gallbladder, kidney, and the mesentery. Gastrointestinal lymphangiomas are extremely rare in adults (2-4). Clinical manifestations vary from asymptomatic masses to acute abdominal pain. Ultrasonography (US), computed tomography (CT), and magnetic resonance imaging (MRI) are the useful imaging modalities for the diagnosis of lymphangiomas. The typical imaging finding is a partially septated, cystic mass. We present an adult patient with a huge cystic lymphangioma of the sigmoid colon, together with a brief review of the literature.

CASE REPORT

A 25-year-old female presented with acute-onset abdominal distension and severe pelvic pain after a one-month history of pelvic fullness and discomfort. The abdomen was uniformly distended with tenderness. An irregularly shaped, soft, immobile, and palpable mass in the left adnexal region was found on pelvic examination. US demonstrated a hypoechoic cystic mass, measuring 10 x 9 x 4 cm, located in the left adnexal region. We suspected an ovarian malignancy based on these findings. CT

Address for correspondence: Çağlar YILDIZ
Cumhuriyet University School of Medicine,
Department of Obstetrics and Gynecology, Sivas, Turkey
E-mail: dr_caglaryildiz@yahoo.com

Manuscript received: 10.02.2010 **Accepted:** 26.04.2010

Turk J Gastroenterol 2011; 22 (3): 344-346
doi: 10.4318/tjg.2011.0225

images revealed a multicystic mass located at the sigmoid colon and the left adnexal region (Figure 1). Complete blood count analysis showed a hemoglobin level of 8.5 g/dl (normal range: 12-16 g/dl).

Exploratory laparotomy was performed and revealed a heterogenous mass of the sigmoid colon, measuring 10 x 7 x 6 cm (Figure 2). There was no adnexal pathology. Pathological examination confirmed the diagnosis of cystic lymphangioma of the sigmoid colon, measuring 9 x 7 x 1 cm. No other pathology was found, and the postoperative recovery of the patient was uneventful.

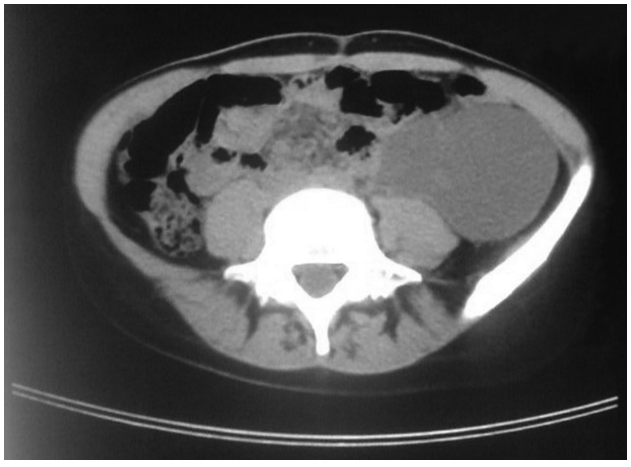

Figure 1. Computed tomography image of the cystic lymphangioma.

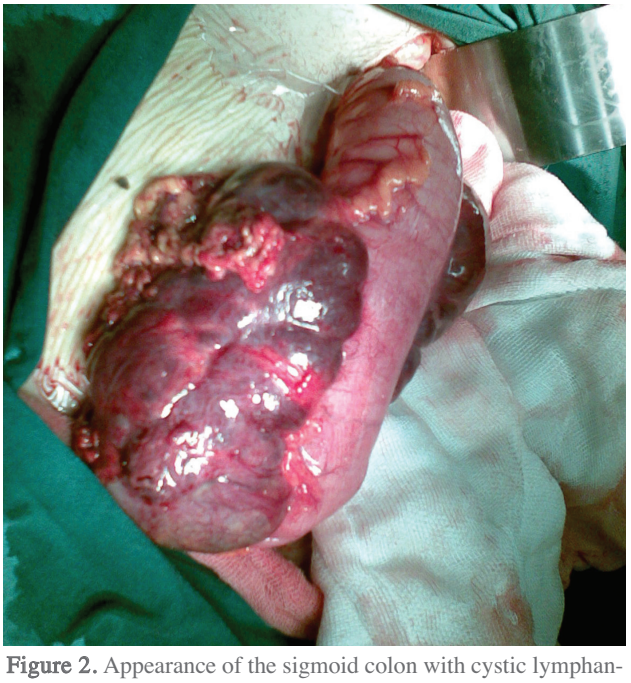

Figure 2. Appearance of the sigmoid colon with cystic lymphangioma.

DISCUSSION

The etiology of lymphangiomas remains unclear. Congenital malformation of the lymphatic vessels leading to sequestrations of lymphatic tissue seems to be the cause of this condition (5). Several forms of lymphangiomas, including simple capillary, cavernous and cystic, have been described. The classification of lymphangiomas is mainly based on the size of the lymphatic space and the nature of the lymphatic wall (6,7). The cystic form is characterized by a thin irregular wall covered by endothelium, smooth muscle, foam cells, and lymphatic tissue; it appears as a multiseptated, cystic mass with or without intracystic debris and has no connection with the adjacent normal lymphatics (8,9). Thin-walled lymphatic vessels situated superficially in the skin characterize the simple type. The cavernous type is composed of dilated lymphatic vessels and lymphoid stroma and has connections with normal adjacent lymphatics (10).

The size and location of the lymphangiomas are the determinants of the clinical presentation. The clinical manifestations vary from an asymptomatic state to acute onset of severe abdominal pain (11-13). Abdominal pain and distension are the most common symptoms of intraabdominal lymphangiomas (14). US evaluation is the first step of the diagnostic procedures. A cystic lesion with multiple thin septa is the typical US finding of a mesenteric lymphangioma (10). Cavernous lymphangiomas appear as solid masses on CT. MRI is more useful than CT for detecting fluid-filled cystic lesions like cavernous lymphangiomas (15).

Although cystic lymphangiomas are benign tumors, they may compress adjacent structures via a mass effect. Other complications, including rupture, secondary infection, volvulus, or intestinal obstruction, have also been reported (16-19). The gold standard treatment modality of cystic lymphangiomas is surgical excision. Treatment with OK-432, steroids, bleomycin, fibrin glue, or Ethibloc has not been established to be superior to surgery (20,21). However, some authors recommend conservative management of asymptomatic cystic lymphangiomas due to the spontaneous regression rate of 10% (8,20). Other treatment modalities including aspiration and injection of sclerosant agents may be performed for emergent decompression; however, these approaches are not recommended for elective therapy due to the high recurrence rates.

REFERENCES

1. Colovic RB, Grubor NM, Micev MT, et al. Cystic lymphangioma of the pancreas. World J Gastroenterol 2008; 28: 6873-5.
2. Vargas-Serrano B, Alegre-Bernal N, Cortina-Moreno B, et al. Abdominal cystic lymphangiomas: US and CT findings. Eur J Radiol 1995; 19: 183-7.
3. Chung JH, Suh YL, Park IA, et al. A pathologic study of abdominal lymphangiomas. J Korean Med Sci 1999; 14: 257-62.
4. Yoshida Y, Okamura T, Ezaki T, et al. Lymphangioma of the oesophagus: a case report and review of the literature. Thorax 1994; 49: 1267-8.
5. Roisman I, Manny J, Fields S, Shiloni E. Intra-abdominal lymphangioma. Br J Surg 1989; 76: 485-9.
6. Chen CW, Hsu SD, Lin CH, et al. Cystic lymphangioma of the jejunal mesentery in an adult: a case report. World J Gastroenterol 2005; 11: 5084-6.
7. Rieker RJ, Quentmeier A, Weiss C, et al. Cystic lymphangioma of the small-bowel mesentery: case report and a review of the literature. Pathol Oncol Res 2000; 6: 146-8.
8. Alqahtani A, Nguyen LT, Flageole H, et al. 25 years' experience with lymphangiomas in children. J Pediatr Surg 1999; 34: 1164-8.
9. Goh BK, Tan YM, Ong HS, et al. Intra-abdominal and retroperitoneal lymphangiomas in pediatric and adult patients. World J Surg 2005; 29: 837-40.
10. Chen CW, Hsu SD, Lin CH, et al. Cystic lymphangioma of the jejunal mesentery in an adult: a case report. World J Gastroenterol 2005; 28: 5084-6.
11. Seki H, Ueda T, Kasuya T, et al. Lymphangioma of the jejunum and mesentery presenting with acute abdomen in an adult. J Gastroenterol 1998; 33: 107-11.
12. de Vries JJ, Vogten JM, de Bruin PC, et al. Mesenterical lymphangiomatosis causing volvulus and intestinal obstruction. Lymphat Res Biol 2007; 5: 269-73.
13. Griffa B, Basilico V, Feltri M, Griffa A. Submucosal jejunal lymphangioma: an unusual case with obscure gastrointestinal bleeding in an adult, detected by videocapsule endoscopy and treated by laparoscopy. Minerva Chir 2006; 61: 529-32.
14. Bliss DP, Coffin CM, Bower RJ, et al. Mesenteric cysts in children. Surgery 1994; 115: 571-7.
15. Gabata T, Terayama N, Yamashiro M, et al. Solid serous cystadenoma of the pancreas: MR imaging with pathologic correlation. Abdom Imaging 2005; 30: 605-9.
16. Ricca RJ. Infected mesenteric lymphangioma. N Y State J Med 1991; 91: 359-61.
17. Porras-Ramirez G, Hernandez-Herrera MH. Hemorrhage into mesenteric cyst following trauma as a cause of acute abdomen. J Pediatr Surg 1991; 26: 847-8.
18. Yoon HK, Han BK. Chronic midgut volvulus with mesenteric lymphangioma: a case report. Pediatr Radiol 1998; 28: 611.
19. Troum S, Solis MM. Mesenteric lymphangioma causing bowel obstruction in a child. Southern Med J 1996; 89: 808-9.
20. Steyaert H, Guitard J, Moscovici J, et al. Abdominal cystic lymphangioma in children: benign lesions that can have a proliferative course. J Pediatr Surg 1996; 31: 677-80.
21. Losanoff JE, Richman BW, El-Sherif A, et al. Mesenteric cystic lymphangioma. J Am Coll Surg 2003; 196: 598-603.