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Case Report

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ABSTRACT

Omental lymphangioma is a rare pathology presented in the abdomen, and it can mimic several clinical conditions. Owing to its variable signs and symptoms, making a correct diagnosis by clinicians may be difficult. Surgical excision is the gold standard treatment. In this article, we report a case who presented with nonspecific abdominal complaints, and underwent surgery. Final pathologic diagnosis revealed omental cystic lymphangioma.

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Introduction

Lymphangiomas, also known as "lymphatic malformations," are benign vascular neoplasms believed to result from a developmental failure of lymphatic system or inflammation of lymphatics causing obstruction. The majority of cases occur in the children, especially in the neck or axilla. Less frequently, they are found in the adults, and are even rarely located in the abdomen.

Clinical symptoms depend on the size and location of cyst, ranging from mild nausea, abdominal distention, to acute abdominal pain. Accurate diagnosis may be difficult even with imaging studies and eventually requires tissue proof.

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Fig. 1 – Contrast enhanced abdominal CT, axial view (A) and coronal reconstruction (B), revealed a lobulated thin-walled cystic mass abutting the greater curvature of the gastric body (straight arrows), with mild fatty infiltration of the adjacent omentum. A suspicious uterine myoma in the lower abdomen (curved arrow in B).



Fig. 2 – (A) Laparoscopy revealed a 13 cm omental hemorrhagic cystic tumor. (B) The excised tumor after drainage of 200 cc. cystic content.

Case report

A 46-year-old female patient with medical history of hepatitis B and Helicobacter pylori infection presented to the emergency room due to abdominal pain for 2 days. She also mentioned a palpable epigastric mass for a period of time. On arrival, initial examination revealed stable vital signs but elevated blood pressure(152/99 mm Hg), and an unremarkable laboratory study. Abdominal computed tomography (CT) disclosed a cystic mass with thin-walled enhancement closely abutting the greater curvature of the gastric body (Fig. 1). The differential diagnosis at that time included cystic gastrointestinal stromal tumor(GIST), abscess, and gastric duplication cyst. She was admitted for further evaluation and treatment.

During hospitalization, the patient received operation for tumor excision. A 13 cm omental hemorrhagic cystic tumor was found (Fig. 2). Histologic examination (Fig. 3) of the tumor revealed large lymphatic channels with focally disorganized smooth muscle wall, chronic inflammatory cell infiltration in the connective tissue stroma, granulation tissue, fibrosis, fibrinous exudate, blood cells, and focal fat necrosis. Immunohistochemically, the lining cells of channels were positive for CD34, and negative for calretinin immunostain. Final pathologic diagnosis of omental cystic lymphangioma was confirmed.

The patient recovered uneventfully after the surgery. No local recurrence is noted so far during follow-up.

Discussion

Lymphangiomas are benign vascular neoplasms. They are also known as the common synonym "lymphatic malformations." According to the 2018 ISSVA (International Society for the Study of Vascular Anomalies) classification of vascular anomalies, they can be further divided into 3 subtypes based on the size of the cystic lesions: macrocystic, microcystic, and mixed cystic lymphatic malformations. The case presented in this article belongs to the macrocystic category.

Although the exact etiology of lymphangiomas remains unclear, it is believed to result from a developmental failure of lymphatic system or inflammation of lymphatics causing



Fig. 3 – Histology of cystic lymphangioma. Cystic space with thin wall, endothelial lining, and proteinaceous content (A: H&E 20x; B: H&E 400x). Immunohistochemical staining disclosed positive for CD34 (C) and negative for calretinin (D).

obstruction [1–6]. They usually occur in children, and more than 95 percent involve the head and neck regions [7]. Less than 1% of cases affect the mesentery, greater omentum, and retroperitoneum [5].

The clinical presentations of lymphangiomas can be diverse and nonspecific. They are usually found incidentally during imaging studies or laparotomy done for other reasons [8]. They can cause subtle abdominal discomfort such as mild nausea, abdominal distention, and may also result in acute abdomen with abdominal pain, small bowel obstruction, volvulus, rupture, or hemoperitoneum, depending on the size, and location [9].

Most lymphangiomas appear homogeneous and cystic on imaging. The attenuation of the fluid varies, depending on internal contents. However, an accurate diagnosis cannot be made in many cases owing to their positions related to organs. Differential diagnosis includes other abdominal cystic lesions such as mesenteric cysts, cystic teratomas, mucinous cystadenomas, ovarian cysts, nonpancreatic pseudocysts, intestinal duplication cysts, cystic tumors, and complicated ascites [10].

Treatment options are either resection or sclerotherapy. Complete surgical excision remains the gold standard treatment either through laparoscopy or by conventional open surgery. Although lymphangiomas are benign lesions, they tend to invade adjacent structures [1], and the involved organ must also be resected. Nevertheless, radical resection is sometimes technically impossible, because of local invasiveness with infiltration of adjacent organs or the main arterial branches. Incomplete resection has a 10% postoperative recurrence rate. Histologically, lymphangioma is characterized by thinwalled cystic structure with endothelial lining and contains lymphoid tissue, small lymphatic spaces, smooth muscles, and foam cells [5].

In our case, the patient initially presented a palpable epigastric mass with epigastralgia. CT revealed a thin-walled cystic mass abutting the greater curvature of the gastric body with intrinsic density, thus gastric tumor such as cystic gastrointestinal stromal tumor cannot be excluded with confidence. Abdominal abscess was also suspected due to adjacent fatty infiltration. Furthermore, gastric duplication cyst was also considered because of the thin wall inseparable from the stomach.

Owing to the clinical symptoms and diagnostic uncertainty, the patient underwent surgery for tumor removal. Pathologic diagnosis disclosed an omental cystic lymphangioma with typical features of thin-walled endothelial lining cystic spaces, stromal aggregations of lymphocytes, and internal proteinaceous content. Because mesenteric cyst grossly resembles cystic lymphangioma, immunohistochemical staining was performed for further confirmation. Based on the results of positive CD34 and negative calretinin staining, final diagnosis of cystic lymphangioma was made.

Conclusion

Omental cystic lymphangioma is a rare benign vascular neoplasm. However, it can mimic several disease entities, and has nonspecific clinical symptoms. Because the tumor is essentially benign, it can be watched for a period if there are no significant symptoms. Rarely, malignant degeneration to lowgrade sarcoma has been reported in the literature. Surgery remains the gold standard treatment and is rational to relieve symptoms and confirm diagnosis.

Patient consent

Entirely anonymized images were used.

REFERENCE

- Takiff H, Calabria R, Yin L, Stabile BE. Mesenteric cysts and intraabdominal cystic lymphangiomas. Arch Surg 1985;120:1266–9.
- [2] Roisman I, Manny J, Fields S, Shiloni E. Intra-abdominal lymphangioma. Br J Surg 1989;76:485–9.
- [3] Fonkalsrud EW. Congenital malformations of the lymphatic system. Semin Pediatr Surg 1994;3:62–9.

- [4] de Perrot M, Rostan O, Morel P, Le Coultre C. Abdominal lymphangioma in adults and children. Br J Surg 1998;85:395–7.
- [5] Losanoff JE, Richman BW, El-Sherif A, Rider KD, Jones JW. Mesenteric cystic lymphangioma. J Am Coll Surg 2003;196:598–603.
- [6] Kim Jong Han, Ryu Woo Sang, Min Byung Wook, Song Tae Jin, Son Gil Soo, Kim Seung Joo, et al. Acquired omental cystic lymphangioma after subtotal gastrectomy: a case report. J Korean Med Sci 2009;24:1212–15.
- [7] Alameri Aws, Museedi Abdulrahman, Nashawi Mouhamed, Ghalli Abdullah, Nathanson Robert. A rare presentation of omental lymphangioma masquerading as malignancy in an adult patient. ACG Case Rep J 2020;7 Page 2 (Discussion).
- [8] Kumar S, Agrawal N, Khanna R, Khanna AK. Giant lymphatic cyst of omentum: a case report. Cases Journal 2009;2(1):23.
- [9] Oudheusden Thijs Ralf van, Nienhuijs Simon Willem, Demeyere Thomas Bernard Joseph, Luyer Michael Derek Philip, Hingh Ignace Hubertus Johannes Theodorus de. Giant cystic lymphangioma originating from the lesser curvature of the stomach. World J Gastrointest Surg 2013;5(10):264–7.
- [10] Wilson SR, Bohrer S, Losada R, Price AP. Retroperitoneal lymphangioma: an unusual location and presentation. J Pediatr Surg 2006;41(3):603–5.