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# **Case Report**

# Cystic lymphangioma of the omental bursa in adult: A rare case report\*

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### ABSTRACT

Cystic lymphangioma is a benign lymphatic malformation that primarily affects children, with rare occurrences in adults. These malformations are most commonly found in the head and neck region, though their presence in the abdominal cavity is infrequent. In this report, we present the case of 71-year-old women with a cystic lymphangioma located in the omental bursa. The rarity of this condition in adults, combined with its unusual abdominal location, highlights the unique aspects of this case. This report explores the clinical presentation, diagnostic challenges, and management strategies for these uncommon lymphatic malformations.

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# Introduction

Cystic lymphangioma is a rare congenital malformation of the developing lymphatic system [1]. It primarily occurs in children, with over 90% of cases diagnosed in patients under 2 years old, and only 7% found in adults [2,3]. These malformations typically occur in the head and neck region, accounting for the majority of cases. Intraabdominal lymphangiomas in adults are particularly uncommon. Among these, cystic lymphangioma of the omental bursa is exceedingly rare, with only a few cases reported in the literature [4]. The omental

bursa, also known as the lesser sac, is a compartment within the abdominal cavity, and its involvement in lymphangiomas presents unique diagnostic and therapeutic challenges.

The clinical presentation of cystic lymphangioma can vary widely, often leading to misleading or nonspecific symptoms. This variability necessitates the use of complex imaging studies to accurately evaluate and diagnose the condition. Techniques such as MRI (magnetic resonance imaging), CT (computed tomography) scans, and ultrasound are critical in providing detailed images that help differentiate cystic lymphangiomas from other abdominal masses and guide appropriate management strategies.

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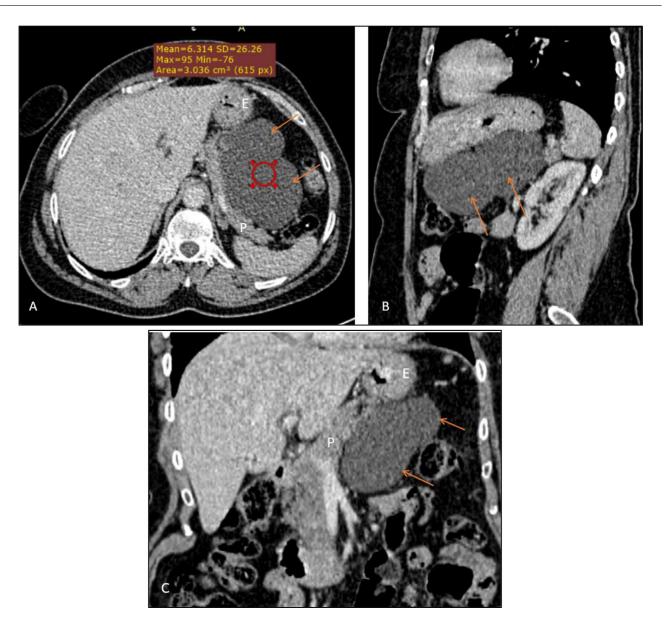


Fig. 1 – Axial (A), sagittal (B) and coronal (C) CT images of the abdomen with intravenous contrast show a well-circumscribed cystic mass (orange arrows) with a thin wall and lobulated margins, situated within the omental bursa. The lesion displaces the stomach (E) anteriorly and exerts external compression on the body of the pancreas (P).

# Case report

A 71-year-old woman with no significant medical history presented with chronic epigastric pain that had progressively worsened over the past few years, while maintaining generally good overall health. Clinical examination revealed slight tenderness in the epigastric region, but no palpable mass was detected. Routine blood tests were normal, and upper gastrointestinal endoscopy showed no abnormalities.

Abdominal ultrasound revealed a large cystic lesion with a few internal septa and thin wall located superior to the pancreas. The remainder of the abdomen appeared unremarkable. Contrast-enhanced computed tomography (CT) (Fig. 1) identi-

fied the lesion as a large, well-circumscribed cystic mass with a thin well and lobulated margins, measuring  $9\times5\times6$  cm, situated within the omental bursa. The lesion displaced the stomach anteriorly and exerted external compression on the body of the pancreas. It did not show any pathological enhancement following contrast agent injection. Biological tests and tumor markers were normal, and hydatid serology was negative. Based on these findings, a diagnosis of cystic lymphangioma was suggested. A complete surgical resection was carried out, and the pathological examination of the specimen confirmed the diagnosis of cystic lymphangioma (Fig. 2). The postoperative course was uneventful, and the patient's symptoms resolved completely following surgery.

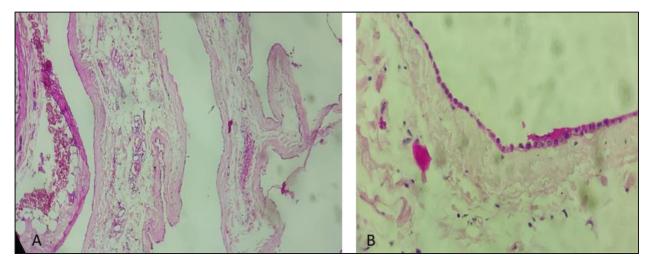


Fig. 2 – Histology of cystic lymphangioma. (A) Multiple large irregular cystic luminal spaces (H-E stain: original magnification x40). (B) Cystic lumen lined by widely spaced, thin endothelial cells without atypia (H-E stain: original magnification x200).

## Discussion

Cystic lymphangioma is a rare benign malformation of the lymphatic vessels that primarily manifests during childhood, and is rarely seen in adulthood. It most commonly affects the head and the neck (75%), and axillary region (20%) [5]. However, its occurrence in the abdominal cavity, including mesenterial and retroperitoneal locations, is extremely rare, representing less than 5% of all cases [6]. Additionally, only a few instances of lymphangiomas located in the omental bursa have been reported [4].

Abdominal cystic lymphangiomas occur most commonly in the mesentery, followed by the greater omentum, the mesocolon, and the retroperitoneum [7].

The etiology of cystic lymphangioma remains unclear. However, most researchers regard it as a congenital anomaly, caused by an obstruction in lymphatic tissue due to insufficient communication with the lymphatic drainage system during embryonic development. In adults, other proposed causes include abdominal trauma, lymphatic obstruction resulting from inflammation or cancer, as well as prior surgeries and radiation therapy [8].

Clinically, most cystic lymphangiomas are asymptomatic and are often discovered incidentally. The clinical manifestations of abdominal cystic lymphangiomas vary widely depending on the size and location of the lesion. In symptomatic cases, patients may present with chronic abdominal pain, nausea, vomiting, weight loss and abdominal distension. Acute abdominal pain requiring surgery is not common [9].

Abdominal ultrasound is the initial examination performed to determine whether the mass is cystic or solid. The characteristic ultrasound appearance of cystic lymphangioma is a well-defined, multilocular cystic mass with multiple thin septa. It is usually anechoic but may appear hyperechoic if it contains debris, a high lipid concentration, infection, or hemorrhage [10].

On CT, lesions appear as cystic masses with homogeneous fluid density and no internal contrast enhancement after intravenous injection. It is rare for CT to demonstrate intrinsic septations.

MRI is an essential diagnostic tool that provides a detailed evaluation of the extent of involvement with surrounding tissues, which is valuable for surgical planning. On MRI lymphangiomas appear hypointense or isointense compared to muscle on T1-weighted images and hyperintense on T2-weighted images [6] suggesting fluid content, with regular margins, thin walls, and internal septa; after gadolinium administration, the walls and the internal septa may show a contrast enhancement.

Recognizing characteristic imaging features can enable early diagnosis and timely treatment of abdominal cystic lymphangioma [11].

The diagnosis of cystic lymphangioma is definitively confirmed through histopathological examination. Pathologically, cystic lymphangiomas are characterized by dilated, thinwalled lymphatic channels with flat endothelial cells. It may show prominent stromal lymphoid tissue and proteinaceous fluid [12].

The differential diagnosis for cystic lymphangioma of the omental bursa includes:

- Teratoma: On CT and MRI, teratomas appear as cystic lesions with significant fat content and possible calcifications [13].
- Primary Malignant Neoplasms: This category includes liposarcoma, leiomyosarcoma, and malignant gastrointestinal stromal tumors [14,15]. While their imaging characteristics are nonspecific, CT and MRI can reveal features suggestive of malignancy, such as invasion, hemorrhage, or necrosis.
- Lymphadenopathy: Additional localizations of lymphadenopathy can assist in diagnosis.
- Enteric Duplication Cyst: Imaging features of duplication cysts are nonspecific due to their variable locations [13].

Complete surgical resection is considered the gold standard for managing intra-abdominal cystic lymphangiomas [11]. This approach is advised even in asymptomatic cases, as it helps to minimize the risk of complications and recurrence. The surgery can be performed either through laparotomy or laparoscopy, depending on the case specifics and surgeon's preference.

Following complete resection, the prognosis is highly favorable, with recurrence being uncommon.

## Conclusion

In summary, this case report highlights the rare occurrence of cystic lymphangioma in the omental bursa, a location infrequently documented in medical literature. The diagnosis was challenging due to the atypical presentation and the lesion's uncommon location within the abdominal cavity. Advanced imaging techniques, including CT and MRI, played a crucial role in identifying and characterizing the mass, guiding the surgical approach.

#### Patient consent

Written informed consent for the publication of this case report was obtained from the patient.

## REFERENCES

- [1] Wierzbicka E, Herbreteau D, Robert M, Lorette G. Malformations lymphatiques kystiques. Ann Dermatol Venereol. 2006;133:597–601. doi:10.1016/S0151-9638(06)70973-5.
- [2] Singh S, Baboo ML, Pathak IC. Cystic lymphangioma in children: report of 32 cases including lesions at rare sites. Surgery 1971;69:947–51. doi:10.5555/uri:pii:0039606071902492.

- [3] Chen KY, Wu CH, Chen KH, Wu JM. Abdominal cystic lymphangioma presenting fever in an adult. Asian J Surg 2023;46:2899–900. doi:10.1016/j.asjsur.2023.01.103.
- [4] Kumar SS, Das SA, Kate V. Cystic lymhangioma of the lesser sac in adult presenting with features of gastric outlet obstruction: a case report. J Clin Diagn Res 2015;9(11):PD15–16. doi:10.7860/JCDR/2015/14789.6830.
- [5] Bakhshi GD, Subnis BM, Sheikh A, Patnaik A, Algappan C, Kher Y. Cystic lymphangioma of the lesser omentum in an adult. Indian J Gastroenterol 2008;27(1):46.
- [6] Maghrebi H, Yakoubi C, Beji H, Letaief F, Megdich S, Makni A, et al. Intra-abdominal cystic lymphangioma in adults: a case series of 32 patients and literature review. Ann Med Surg (Lond) 2022;81:104460. doi:10.1016/j.amsu.2022.104460.
- [7] Mabrouk MY, Magouri O, Madani A, Guellil A, Rahou FZ, Bouzayan L, et al. Mesenteric cystic lymphangioma in an adult: An unusual case report. Ann Med Surg (Lond) 2022;78:103917. doi:10.1016/j.amsu.2022.103917.
- [8] Chen CW, Hsu SD, Lin CH, Cheng MF, Yu JC. Cystic lymphangioma of the jejunal mesentery in an adult: a case report. World J Gastroenterol 2005;11(32):5084–6. doi:10.3748/wjg.v11.i32.5084.
- [9] C Papanicolaou, G Anthimidis, D Alataki, V Lagopoulos, I Sougas, G Vaios, et al. Cystic lymphangioma of the lesser sac (omental bursa). Internet J Surg. 2008; 18 1.
- [10] Michail O, Michail P, Kyriaki D, Kolindou A, Klonaris C, Griniatsos J. Rapid development of an axillary mass in an adult: a case of cystic hygroma. South Med J 2007;100:845–9. doi:10.1097/SMJ.0b013e3180f60e09.
- [11] Xiao J, Shao Y, Zhu S, He X. Characteristics of adult abdominal cystic lymphangioma: a single-center Chinese cohort of 12 cases. BMC Gastroenterol 2020;20(1):244. doi:10.1186/s12876-020-01388-8.
- [12] Weiss SW, Goldblum JR. Enzinger and Weiss's Soft Tissue Tumors. Philadelphia, PA: Mosby Elsevier; 2008.
- [13] Elmohr MM, Blair KJ, Menias CO, Nada A, Shaaban AM, Sandrasegaran K, et al. The lesser sac and foramen of winslow: anatomy, embryology, and ct appearance of pathologic processes. AJR Am J Roentgenol 2020;215(4):843–51. doi:10.2214/AJR.19.22749.
- [14] Noushad SN, Rajaraman R, Shanmugam S. Primary lesser sac myxoid liposarcoma: a case report. Int J Surg Case Rep 2016;24:10–13.
- [15] Katsoulis IE, Tzortzopoulou A, Tziakou P, Arnogiannaki N, Kostoglou-Athanassiou I, Lypas G, et al. Extragastrointestinal stromal tumour of the lesser omentum: A case report and literature review. Int J Surg Case Rep 2017;37:17–21. doi:10.1016/j.ijscr.2017.05.040.