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GI IMAGE

# A Rare, Giant, Cystic, and Cavernous Lymphangioma Originated from the Stomach in a Young Woman

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## Introduction

Lymphangioma is a congenital benign tumor which originates from abnormal lymphatic tissue. Over 90% of lymphangiomas involve the head and neck and occur in children less than 5 years of age.[1] Abdominal lymphangiomas are extremely rare. According to the histopathology, lymphangiomas have been classified into three subtypes, including capillary lymphangiomas, cavernous lymphangiomas, and cystic lymphangiomas.[2] Lymphangiomas involving the gastrointestinal system are not common, and mixed lymphangiomas of two pathological types are actually rare. The present study describes a relatively complete diagnosis and treatment of a giant lymphangioma containing both cystic and cavernous components, which originated from the lymph tissue of gastric wall and occurred in a 24-year-old female. To our acknowledgement, this might be the largest lesion that has ever been reported. This study may provide further clinical experience for treatment of gastrointestinal lymphangiomas.

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## Report

A 24-year-old female with gastroenteric syndrome presenting as consistent abdominal distension in the middle-lower abdomen, nausea, and vomiting. After hospitalization, physical examination revealed a 12 × 12-cm palpable mass in the middle abdomen. The palpable mass shared a relatively soft touch with poor mobility. Abdominal tenderness was positive, whereas Murphy sign and shifting dullness were negative. The laboratory analysis showed normal. Further abdominal routine examination and enhanced computed tomography (CT) showed a 8 × 15 × 22-cm giant irregular cystic mass in the left peritoneal cavity, which originated from the stomach. Multilocular compartment and small vessels were observed in the mass via enhanced CT (Fig. 1, both in longitudinal section and cross section). Hydatid fluid from the cystic mass was used for laboratory analysis. Several biochemical index showed relatively high expression, with red blood cell (RBC) of $3 \times 10^9$/L, white blood cell (WBC) of $1.39 \times 10^8$/L, and hyaline leukocyte of 77%. Also, Rivalta test was positive according to the data.

Since the cystic mass has a large size, further treatment for this patient was surgical resection by laparoscopic partial gastrectomy. Pathology showed that the lymphangioma originated from the serosal layer of the stomach with a typical feature of cystic and cavernous lymphangioma (Fig. 2a). It also presented as multilocular compartment with pavement epithelium cells and accumulation of leukomonocyte and vessels was also observed in leukomonocyte (Fig. 2b, c). Immunohistochemistry (IHC) study for D2-40 (specific lymphangioma marker) was

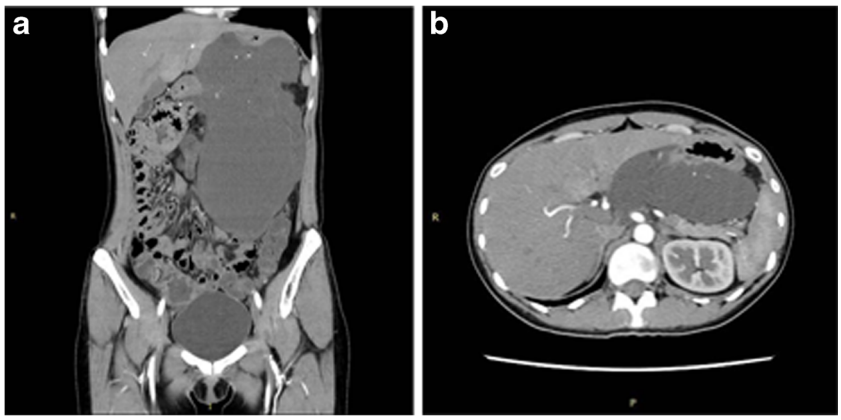


**Fig. 1** **a** Abdomen routine and enhanced computed tomography (CT) showed a giant cystic mass in the left peritoneal cavity in longitudinal section. **b** A lucid image to illustrate the origination of lymphangioma from corpora ventriculi of the stomach (cross section performed)

positive, whereas HMA-45 was negative (Fig. 2d, e). Based on follow-up evaluation, the patient showed no more symptoms within the next half year after surgical treatment.

Here, we reported a giant lymphangioma containing both cystic and cavernous compartments, which originated from lymph tissue of gastric wall and occurred in a young female. After reviewing previous researches in this field, we find that the present case we report might be the largest lymphangioma that has ever been reported, with the size of 8 × 15 × 22 cm.

With the development of medical imaging, routine CT and MRI scannings have been major examinations for diagnosis of lymphangiomas. Generally speaking, lymphangioma may serve as a non-enhancing extramucosal mass with homogeneous attenuation, allowing evaluation of the lesion extent before the treatment commences. Of note, medical imaging scannings help identifying other abdomen lesions, such as ovarian cyst, abscess, and some gastrointestinal tumors. Pathological diagnosis provides exact classification for lymphangioma. Combining use of CD31 and D2-40 staining as specific markers for both blood vessels and lymphatic endothelium is helpful.[3]

Up to date, main treatment for lymphangioma is surgical resection. Some studies also shared a method of endoscopic submucosal dissection (ESD) when facing a relative small lesion within the diameter of 2 cm. Otherwise, laparoscopy has been the first choice of surgical resection in large lesions, which is safer and has reduced wound than laparotomy. Further follow-up observation is also needed to evaluate the curative effect after surgical treatment.

In sum, our study shared an integrated diagnostic and treating process of a giant gastric cystic and cavernous lymphangioma in a young female. To our knowledge, this may be the largest size of gastrointestinal mixed lymphangioma that has ever been reported. This study may provide more clinical experience for diagnosis of gastrointestinal mixed lymphangioma.

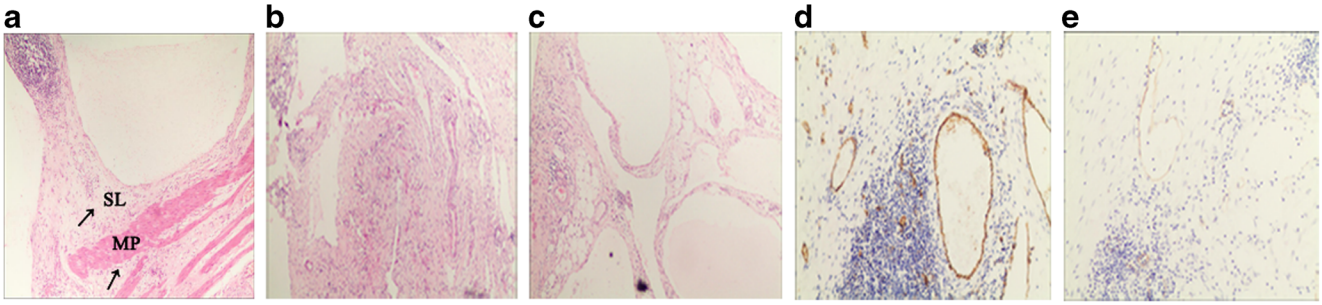


**Fig. 2** **a** Histopathological findings showed that the lymphangioma originated from the serosal layer of the stomach with a typical feature of cystic and cavernous lymphangioma. **b** Multilocular compartment with pavement epithelium cells were found in the cystic mass. **c** **c** Accumulation of leukomonocyte and vessels were also observed in leukomonocyte. Positive immunohistochemical (IHC) staining of CD31 and D2-40, which were specific markers of blood vessels and lymphatic endothelium, were also showed in **d** and **e**. *SL* serosal layer, *MP* muscularis propria

**Author Contributions** Kangmin Zhuang collected the patient's clinical data and wrote the report; Xiangwu Jiang edited, reviewed, and approved the manuscript. Simin Huang revised the manuscript.

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## Compliance with Ethical Standards

**Informed Consent Statement** Written informed consent was obtained from the patient for publication of the case report and accompanying images.

**Conflict of Interest** The authors declare that they have no conflict of interest.

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