Giant cystic lymphangioma in the mesoileum: A case report and literature review

Ling Liu, Ke Ge, Wei-Min Shen, Chang-Ku Jia

Department of General Surgery, Affiliated Hangzhou First People's Hospital, Zhejiang University School of Medicine, Zhejiang Province, China

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Dear Editor,

Mesenteric lymphangioma is a rare, benign tumor derived from the lymphatic vessels that accounts for less than 1% of all the lymphangiomas (1). It is rarely diagnosed, when an intra-abdominal mass is found, because of its low incidence and vague symptoms (2). Imaging assessments, including abdominal ultrasonography, computed tomography (CT), and magnetic resonance imaging, are very important when diagnosing the patients (3, 4). Optimal treatment mainly involves surgical resection (5, 6). We report a rare case of a giant cystic lymphangioma in an older man who underwent laparotomy.

A 64-year-old man with progressive abdominal pain was transferred and admitted to our hospital. He sought medical attention for dull abdominal pain and distension 5 days before presentation. He did not report about having nausea, vomiting, or any other significant constitutional symptom. CT imaging revealed a giant abdominal mass with a predominantly low-grade malignant tumor (Figure 1). Physical examination showed a slightly distended abdomen, sensitivity, and no tenderness without a dominant mass; other features were unremarkable. The initial diagnosis was mesenchymoma, and he was treated with oral antitumor drugs. After 4 days, the patient was examined via ultrasound-guided biopsy to make a definite pathologic diagnosis, but the report was not useful. He was brought to our hospital the next day with progressive abdominal pain and rapid abdominal distension. Physical examination revealed a typically distended abdomen with abdominal tenderness from slight pressure. Laboratory examination showed no abnormal results. Therefore, we performed resection to reach a definitive diagnosis and treatment. During the laparotomy (Figure 2), we found a giant mass in the mesentery of the ileum, and the mass and partial small bowel were surgically removed. Neoplasm examination revealed a gray mass of approximately 20 cm×15 cm in size, consisting of solid and cystic components that adhered to the mesentery of the ileum. There were 3 needle eyes on the tumor surface with oozing blood. Histological examination revealed that the neoplasm was composed of reticular lymphatic vessels with clusters of lymphocytes between the lymph vessels, smooth muscle, and endothelial cells, which are characteristic of cystic lymphangioma (Figure 3). The patient was discharged from our hospital on postoperative day 10, and there was no recurrence at the 1-year follow-up.

Cystic lymphangiomas in the mesentery are very rare in the English literature published in PubMed. However, the detailed number of cases is uncertain owing to the incomplete pathologic differentiation between the mesenteric cysts. The etiology of lymphangioma is still unclear. Several studies have reported that it may be a result of inflammation, abdominal trauma, abdominal surgery, or lymphatic obstruction (6, 7). Lymphangiomas are predominantly asymptomatic, but large intra-abdominal tumors usually cause abdominal pain, secondary infection, rupture-related hemorrhage, intestinal obstruction, or volvulus (8, 9). The tumor's increasing size sometimes causes acute clinical symptoms (10). In our case, the patient presented to the emergency department before being transferred to our institution.

Lymphangiomas are classified into 3 main pathological categories: capillary (simple), cavernous, and cystic (11). The capillary type is usually found in the skin and consists of tiny dilated lymphatic spaces with endothelial cells. The cavernous type has the following characteristics: dilated lymphatic spaces of various sizes, lymphoid stroma,

Corresponding Author: ***Chang-Ku Jia; jiachk@126.com***
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and wall thickness. The cystic type is composed of dilated lymphatic spaces with collagen and smooth muscle bundles in the stroma as well as a cystic wall with a monolayer of endothelial cells (1, 12). In this case, the condition was classified as cystic lymphangioma because of the following characteristic signs: dilated lymphatic spaces with smooth muscle and endothelial cells.

A differential diagnosis should be made between multicystic mesothelioma and lymphangiomyoma (12, 13). The former is a benign tumor that relapses easily, is more common in mid-aged women, and the tumor capsule is lined with cuboidal or flattened mesothelial cells. The latter results from the lymphatic systems compensating for multisystem disease, including lymphangion dilation and hyperplasia of smooth muscle epithelial cells, which is also more common in women.

Patients with mesenteric lymphangioma usually demonstrate the following symptoms: abdominal pain, distension, vomiting, and a palpable abdominal mass (14). However, because of its low incidence and vague symptoms, intra-abdominal lymphangioma is frequently difficult to diagnose, and in most cases, laparotomy has to be performed to diagnose and treat the patients (2, 15). Imaging methods including, B-mode ultrasonography, CT, and magnetic resonance, provide little useful information. They can confirm the presence of an abdominal mass but cannot help accurately diagnose it. The final diagnosis needs to be verified via surgical resection and histopathological investigation (7, 16).

Several studies have reported that fine-needle aspiration may be useful in diagnosing the patients with lymphangioma (17, 18). In this case, the patient underwent ultrasound-guided biopsy prior to presentation; however, we did not acquire the available information for further measure. The patient actually reported continuous abdominal pain after undergoing fine-needle aspiration and was then immediately transferred to our hospital for emergency surgery. We also found 3 needle eyes in the tumor surface with oozing blood in the anatomopathological study of the specimen that we retrieved. It is possible that the patient's pain and peritonitis were biopsy-related hemorrhagic infarction. Given the giant size of the tumor and its proximity to the vessels, it must be treated with extreme caution. Laparotomy or laparoscopic excision is more efficacious and safe in similar cases.

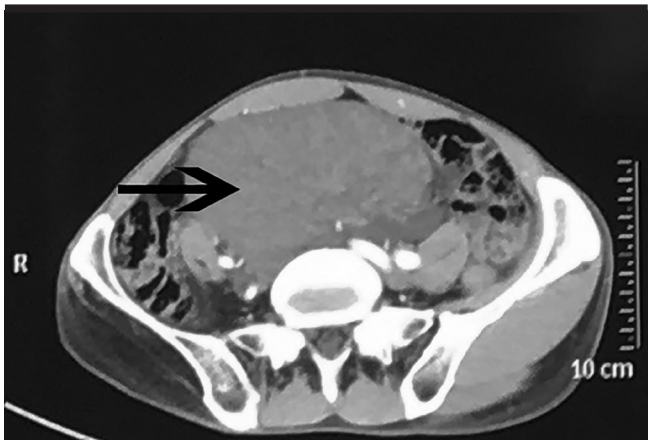

Figure 1. Abdominal computed tomography scan showing a huge cystic tumor

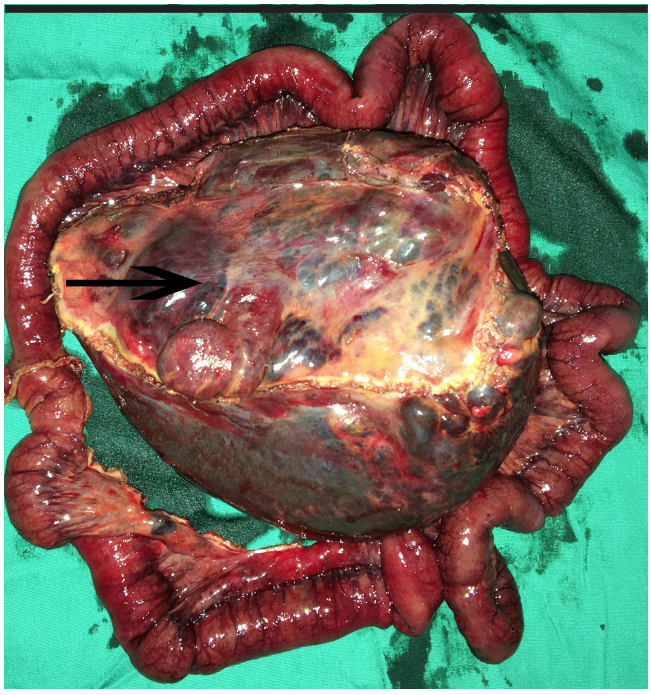

Figure 2. A huge tumor occupied the peritoneal cavity and there were three needle holes on the tumor surface with oozing blood.

MAIN POINTS

- *Mesenteric lymphangioma is a rare, benign tumor derived from the lymphatic vessels.*
- *Lymphangiomas are classified into 3 main pathological categories: capillary (simple), cavernous, and cystic*
- *Intra-abdominal lymphangioma is frequently difficult to diagnose.*
- *Laparotomy or laparoscopic excision is more efficacious and safer in similar cases.*

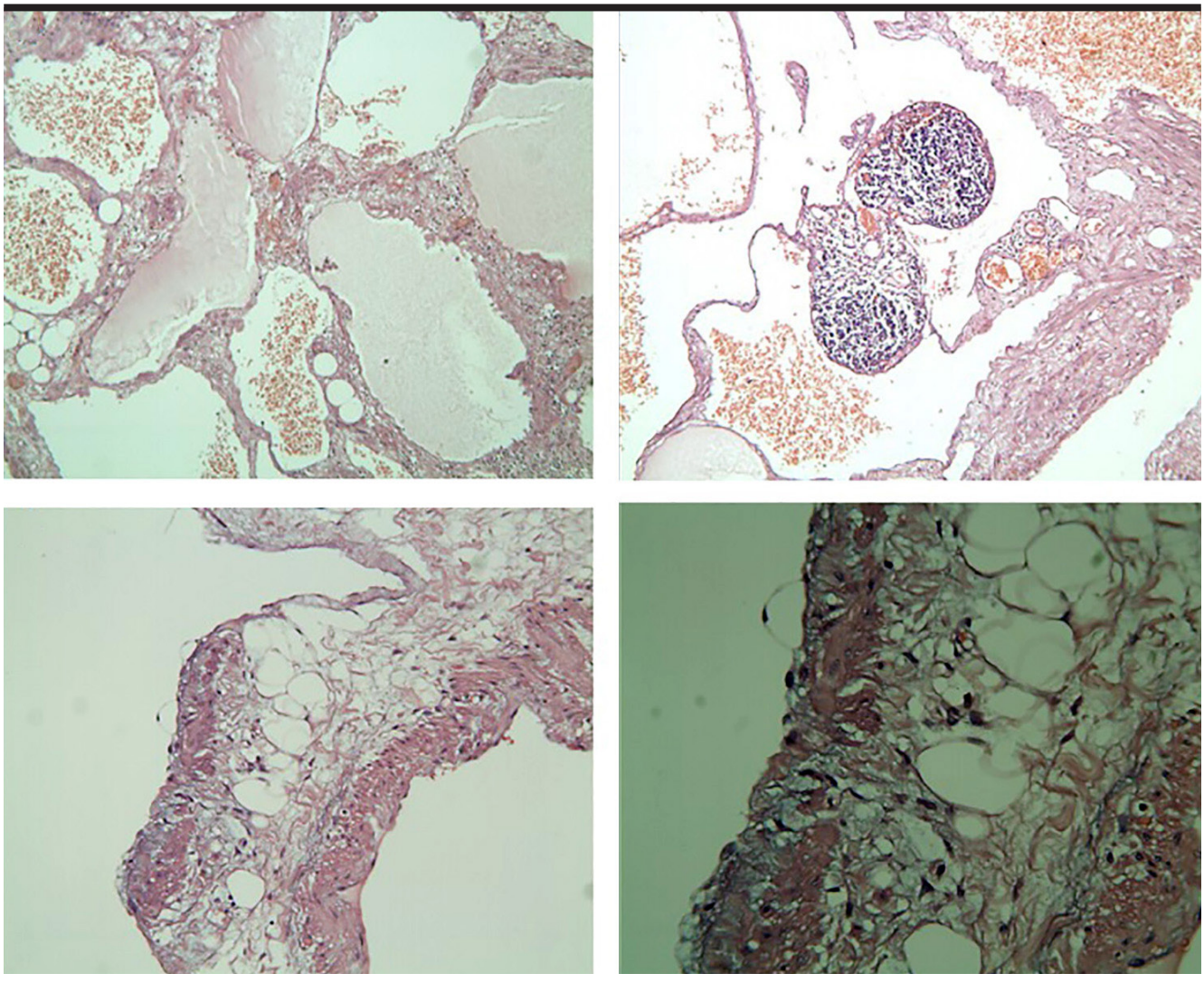

Figure 3. Histopathology (Hematoxylin and eosin). A: Numerous dilatated lymphatic vessels (×100); B: Lymphocytic aggregates in the cyst walls(×100).

Informed Consent: Informed consent was obtained from the patient who participated in this study.

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