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Case Report
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Surgical Resection of a Symptomatic Mesenteric Cystic Lymphangioma

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Illustrative Case Report

A 76-year-old female with onset of intense epigastric pain is admitted to the emergency room.

Lab values showed a neutrophilic leukocytosis (21.50 × 10,000/mm^3), and an increase in inflammatory markers including CRP (344.5 mg/dl).

Abdominal CT scan with and without contrast medium highlighted a voluminous oval formation, homogeneously hypodense, with regular margins, an ample surface contact with the greater curvature of the stomach and without an apparent cleavage plane with the left lobe of the liver. The mass occupies the hepatogastric recess, abolishing it, the axial diameter of the lesion is 104 mm and extends inferiorly along the longitudinal line for 105 mm, with a mean an axial diameter of 120 mm, resulting in contiguity with the margin of the antimesenteric transverse colon.

The patient was electively hospitalized to undergo a CT-guided percutaneous biopsy procedure, during which serosanguinous fluid was aspirated and sent for cytological examination. Intracystic LDH was increased and no amylases or atypical cells were found.

Esophagogastroduodenoscopy showed an ab extrinsic compression of the gastric wall at the level of the small curvature. Colonoscopy showed no pathological findings.

Due to the compressing symptoms and the discomfort, surgical resection was planned. Considering the adhesion of the neoplasia to the right and proximal transverse colon and to the Glissonean capsule of the left hemiliver, an extended hemicolectomy with omentectomy and an atypical resection of the III segment of the liver was performed with open technique. The postoperative course was uneventful and the patient was discharged postoperative day 12.

The histopathological findings were suggestive for a mesenteric cystic lymphangioma (D2-40+, CD34-, calretinin-).

Three months of follow-up revealed complete resolution of the abdominal pain and no diagnostic features of recurrence at CT imaging.

Etiopathogenesis

Cystic tumors of the mesentery are relatively rare, but lymphangiomas account for the majority of them. Lymphangiomas are rare benign lesions originating from the lymphatic system following a dysembryogenic factors, abdominal trauma, lymphatic obstructions, or a local degeneration of some lymphatic tissues due to radiation, inflammation, infection or previous surgical resection. The main etiological hypothesis seems to be related to a congenital abnormality of the lymphatic system [1-11], and this explains the increased incidence in children [1,2], with an 80% to 90% diagnosis in the first year of life [2]. Most reports show a female predominance.

Any part of the body can be involved, except the brain, although head and neck, and axilla are more commonly affected [11]. Abdominal lymphangiomas, especially retroperitoneal forms, are found to be rare. Most of all abdominal lymphangiomas originate from the small bowel mesentery, followed by the mesocolon and the retroperitoneum [3].

They are slow-progressing tumors and only few cases of transformation have been reported. They are most frequently single, but multiple lymphangiomatous cysts can affect a single organ.

Only a small proportion of cystic lymphangiomas are multilocular, and most contain a single cavity [11]. Pathologically, lymphangiomas are subdivided into three main types: Capillary, cavernous, and cystic [11]. The first two are predominantly cutaneous lesions. Cystic lymphangiomas

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are generally found in the abdomen and they typically occur in spaces surrounded by loose connective tissue such as the mesentery and retroperitoneum [3]. Mesenteric lymphangiomas have an incidence of 1/20000 in children and 1/100000 in adults [12].

The cystic spaces are lined with a single layer of endothelium; there are small lymphoid aggregates in the cyst wall that aid in distinguishing lymphangiomas from simple cysts of the mesentery. Lymphangiomas may contain serous, serosanguinous, or chylous fluid, less frequently the fluid can be purulent because of infection [3]. The variability in fluid content might be explained by different degrees of lymph stasis and the cyst fluid's protein content. The presence of blood into the cyst may lead to a misdiagnosis. Different nature of hemorrhagic lymphangiomas and hemangiomas must be established with immunohistochemical analysis [3].

MCL could be classified into 4 types: 1) pedunculated, 2) sessile, 3) with retroperitoneal extension and 4) multicentric. Type 1 is often responsible for volvuli. Type 2 often originates from the mesenteric border. Type 3 and 4 are the ones which can easily become unresectable due to invasion [14].

Clinical Presentation

MCL is a specific and depends on the site of development and on size of tumor. [13] Some patients may present abdominal pain that is not localized, a palpable mass, abdominal distension or an acute abdomen. These symptoms may result from compression, perforation, torsion, or rupture of the adjacent structures, such as the intestine [2].

This neoplasm may grow till reaching huge dimensions or showing infiltrating features.[10]

Diagnosis

Imaging of mesenteric lymphangiomas is not characteristic.

Computed Tomography (CT), and Magnetic Resonance Imaging (MRI) are the most frequently used methods for evaluation of these tumors, therefore the diagnostic accuracy of imaging is not sufficient to differentiate these lesions from other cystic or solid-cystic lesions [1].

Several authors have reported that fine-needle aspiration is useful for confirming a preoperative diagnosis of lymphangioma [2]. Histologic confirmation and immunohistochemical analysis are necessary to make a definitive diagnosis [1].

Treatment

The treatment of choice is radical excision because incomplete resection may lead to recurrence [2]. If the tumor cannot be safely resected because of its extension, systemic treatment with mTOR inhibitors can be considered. Activation of the mTOR pathway must be proven on a sample obtained from a needle biopsy. This scenario is typically found in vascular malformation and congenital tumors [9].

Laparoscopic cyst resection has been performed in many centers and it is considered a safe procedure [4]. Its feasibility depends on the size and site of mesenteric cyst. For bigger lesions, a preoperative fluid aspiration may be taken into consideration [4]. Recurrence after complete and incomplete resection of the lymphangioma has been reported to be about 7% and 50%, respectively [1].

Conclusion

CLM is a rare tumor which rarely originates from the mesentery. Its clinical presentation in a specific, with symptoms varying from mild abdominal pain to acute abdomen. Diagnosis can be done with CT or MRI, but confirmation can only be obtained with histopathological examination. Treatment has the goal of radical surgical excision, to avoid recurrence, and could be performed traditionally or minimally invasive depending on the tumor characteristics.

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