

Cystic lymphangioma of the lesser sac (omental bursa)

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Citation

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Abstract

Cystic lymphangioma is a benign, slow-growing tumour that usually occurs in children and the most frequent location is the head and neck. Intra-abdominal cystic lymphangiomas are rare. We present herein a case of cystic lymphangioma of the lesser sac (omental bursa) in a 58-year-old woman. The patient presented with nonspecific abdominal symptoms. Abdominal ultrasound and computerized tomography revealed a cyst within the lesser sac. Laparotomy was performed and the tumor was removed completely and intact. Histologic examination showed cavernous lymphangioma. The postoperative course was uneventful and the patient was discharged on the 5th postoperative day. During a 1-year follow-up there was no radiologic sign of recurrence.

INTRODUCTION

Cystic lymphangioma is a benign, slow-growing tumour derived from the lymphatic vessels. It usually occurs in children and the most frequent location is the head and neck, while Intra-abdominal location is an extremely rare entity. We report a case of cystic lymphangioma of the lesser sac (omental bursa) in a 58-year-old woman.

CASE REPORT

A 58-year-old woman presented with a 6-month history of mild epigastric pain, aggravated immediately after food intake, early satiation and anorexia. A 6kg weight loss during this period was also mentioned. In physical examination, mild epigastric tenderness was noted with no other physical signs. Abdominal ultrasound revealed a multiseptated elliptic cyst anterior to the pancreas, measuring 125x130mm. An abdominal contrast-enhanced computed tomogram showed a multilocular elliptic cyst measuring 12.5x10x15cm within the lesser sac. Displacement of the stomach anteriorly and external compression of the corpus of the pancreas was obvious.

Figure 1

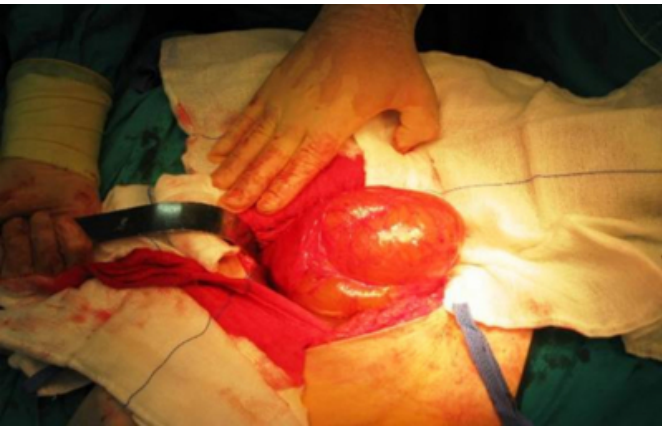
Figure 1: On CT, a well-defined, hypodense lesion is seen at the lesser curvature of the stomach and anterior to the head of the pancreas.



Routine laboratory tests as also tumor markers (Carcinoembryonic antigen, alpha-fetoprotein and carbohydrate antigen 19-9) were within normal range (Table 1). Laparotomy was decided upon for estimation of definitive diagnosis. The cystic lesion was located in the lesser sac with an intimate attachment to the lesser curvature of stomach and celiac axis, containing clear fluid (Figure 2).

Figure 2

Figure 2: Appearance of the cyst during operation.



The tumor was removed completely and intact (Figure 3).

Figure 3

Figure 3: A cystic lesion, containing clear fluid, was removed completely and intact.

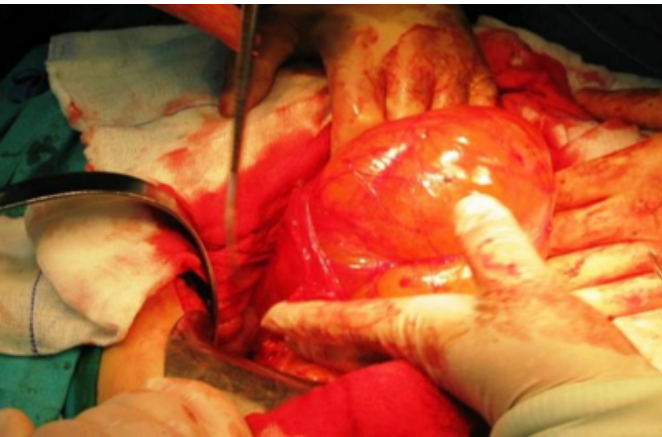


Figure 4

Table 1: Biochemical analysis of the cyst fluid.

SGOT	22 IU/L
SGPT	2 IU/L
Glucose	87 mg/dL
Urea	51 mg/dL
Kreatinine	0.85 mg/dL
Potassium	3.7 mmol/L
Sodium	142 mmol/L
Albumin	3 g/dL
Bilirubin (total)	0.4 mg/dL
Bilirubin (direct)	0.0 mg/dL
γ-GT	8 U/L
Amylase	33 U/L
Alkaline phosphatase	4 IU/L

SGOT: Serum glutamic oxaloacetic transaminase

SGPT: Serum glutamic pyruvic transaminase

γ-GT: γ-glutamyl transpeptidase

Diagnosis was made by light microscopy supported by immunohistochemistry. Histologic examination showed cavernous lymphangioma with irregularly shaped dilated lymphatic vessels with endothelial lining embedded with fibrofatty tissue (Figures 4,5,6).

Figure 5

Figure 4 : Dilated spaces lined by flattened cells, filled with amorphous eosinophilic material.

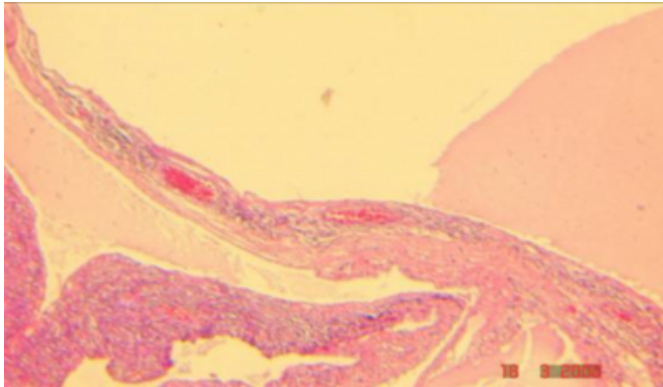


Figure 6

Figure 5: Collections of lymphocytes in loose connective tissue.

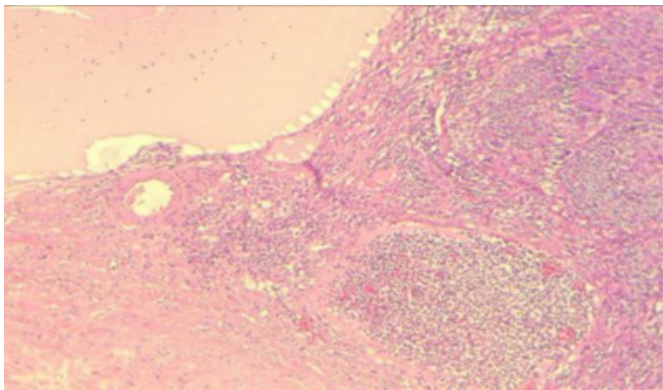
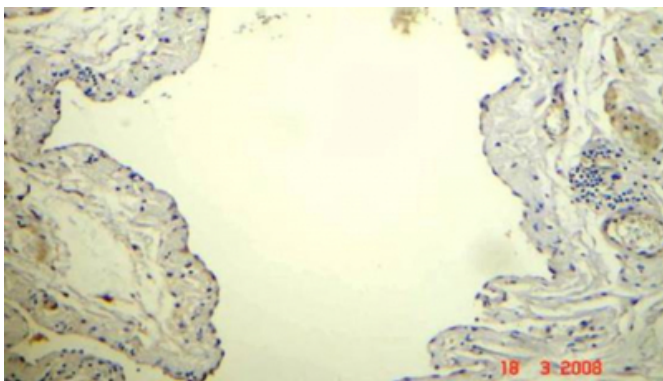


Figure 7

Figure 6: The cells that lined cystic spaces were not immunoreactive for cytokeratins 7/1/3.



The postoperative course was uneventful and the patient was discharged on the 5th postoperative day. During a 1-year follow-up there was no radiologic sign of recurrence.

DISCUSSION

Intra-abdominal cystic lymphangiomas are not common. Their location can involve the mesentery, omentum, colon, spleen, pelvis, groin, and retroperitoneum². As far as we could elicit from the literature, four other cases of such location of cystic lymphangioma in the lesser sac are mentioned^{3,4,5,6}. Cystic lymphangiomas are considered to originate from malformed or malpositioned lymphatic tissue, but other factors such as abdominal trauma, lymphatic obstruction, inflammatory process, surgery, or radiation therapy may lead to secondary formation of such tumors⁷. In our case, according to the patient's history and physical examination, the lesion was classified as related to congenital defects of the lymphatics, remaining asymptomatic until late adulthood and discovered when it progressed to produce vague and chronic abdominal symptoms, secondary to a mass effect⁸.

The clinical presentation is variable. Asymptomatic course is usual, but it can also be the cause of nonspecific symptoms such as anorexia, nausea, vomiting, fatigue, and weight loss. Chronic abdominal distension, detected by palpation of a cystic mass may be present. Acute abdominal pain requiring surgery is not common^{7,8}.

The differential diagnosis includes cystic lesions of mesothelial, enteric or urogenital origin, dermoid cysts or teratomas, pseudocysts from trauma or infectious origins, and cystic degeneration of solid tumors^{9,10,11}. The diagnosis is made by means of abdominal ultrasonography (US) and computerized tomography (CT). Cystic lymphangioma usually presents as a large multilocular cystic mass with enhanced walls, in addition to multiple thin septa containing uncomplicated fluid. Acute cases with intracystic hemorrhage are more difficult to diagnose¹².

Management of intra-abdominal cystic tumors depends on the clinical symptoms, size of the cyst, and the degree of clinical suspicion for malignancy. Imaging usually provides useful information for the planning of treatment. The characteristic imaging features of cystic lymphangiomas often provide the radiologist with a specific preoperative diagnosis. However, the definitive diagnosis is always made histologically. In the present case, characteristic imaging features suggested cystic lymphangioma located in the lesser sac. Lymphangiomas do not generally regress¹³. As such, surgical intervention was preferred. Depending on the size and location of the mass, laparoscopic resection has been proposed as a safe and minimally invasive means of surgical

treatment^{14,15}. In our case, the cyst was located in the lesser sac, compression or infiltration of vital structures was encountered and consequently a minimally invasive surgical approach was considered to be difficult, dangerous or impossible. The lesion was removed intact via laparotomy. However, this is not always easy.

Complete removal of the tumor offers an excellent prognosis. On the other hand, recurrence has been reported in 10% of patients in whom primary resection was incomplete^{16,17}. If the stem of the cyst and feeding lymphatics are unsuccessfully ligated, chylous ascites may also occur. Isolation and ligation of the cystic lymphangioma's peduncle as well as ligation of lymph channels can prevent recurrences and chylous ascites¹⁸. Recurrence may necessitate a second operation¹⁷. Management of chylous ascites may include the placement of drains which have been reported to facilitate the injection of fibrinogen and thrombin for effective treatment of persistent lymphatic drainage^{13,19}.

Additionally, patients who refuse surgery or are poor candidates for laparoscopic resection may benefit from a combination of percutaneous image-guided drainage and sclerotherapy with acetic acid as sclerosant. The sclerosant is removed after sclerotherapy. One treatment session is usually enough. With no major complications reported, this approach offers complete resolution in 66% of patients and reduces the lesion size in 25% of patients by more than 50%²⁰.

In conclusion, intra-abdominal cystic lymphangiomas are rare entities. The diagnosis is suggested by means of abdominal US and CT. Early recognition and appropriate treatment of these cysts are associated with a good prognosis. However, long term follow-up is advisable because of the possibility of recurrence.

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