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SCIENTIFIC LETTER

# Lymphangioma of the pancreas diagnosed through endoscopic ultrasound-guided fine-needle aspiration biopsy: A case report

## Linfangioma de páncreas diagnosticado mediante biopsia con aguja fina guiada por ultrasonido endoscópico: reporte de caso

Lymphangiomas are malformations of the lymphatic system secondary to the failure of the lymphatic channels to communicate with the larger lymphatic vessels, causing the formation of lymph-filled cysts. The macrocysts (cysts larger than 2 cm) are also known as hygromas. They tend to predominantly affect the pediatric population (80-90%) and are rare in adults.[1] Ninety-five percent of cases are usually located in the neck and axillary region and the remaining 5% at the level of the mediastinum and abdomen.[2] Lymphangiomas of the pancreas are extremely rare, accounting for less than 1% of abdominal lymphangiomas and less than 0.2% of pancreatic tumors.[3]

A 32-year-old woman, diagnosed with dextrocardia, had a history of resection of a nonspecified thoracic lesion in childhood. Due to the presence of abdominal discomfort and epigastric pain, she underwent a tomography scan of the chest and abdomen with intravenous contrast. It revealed a diffuse increase of mediastinal fat and a 53 x 33 mm cystic image in the body and tail of the pancreas that did not infiltrate adjacent and vascular anatomic structures. There was no contrast medium enhancement, and the spleen had a heterogeneous parenchyma due to the presence of multiple hypodense ovoid images (Fig. 1a). The laboratory work-up reported the following results: hemoglobin 11.7 g/dl, leukocytes 5,320 cell/mm$^3$, platelets 192,000/mm$^3$, INR 1.16, glucose 82 mg/dl, serum creatinine 0.64 mg/dl, total bilirubin 0.82 mg/dl, ALT 14 IU/l, AST 22 IU/l, GGT 17 IU/l, ALP 40 IU/l, and HbA1c 5.0%.

Endoscopic ultrasound (EUS) was carried out to evaluate the cystic lesion of the body and tail of the pancreas, as well as the splenic lesions, visualizing a considerably enlarged spleen with multiple multiloculated, anechoic, nonvascularized lesions, with diameters of 20-30 mm and thin septa (1-2 mm); qualitative elastography showed no stiffness (Fig. 1b). Adjacent to the spleen, at the level of the tail of the pancreas, there was a cystic, multiloculated 54 x 40 mm lesion with regular borders and septa of up to 2 mm; qualitative elastography showed no stiffness and the lesion did not cause pancreatic duct dilatation or involve vascular structures Fig. 1c). Biopsies were taken from the walls of the cysts at the tail of the pancreas, utilizing a 25 G Acquire™ fine needle (FNB) and transgastric access (3 passes with the fanning method and dry suction technique). The procedure was completed with no immediate complications. The fluid from the cystic lesions could not be sent for cytochemical and cytologic analyses due to the thickness of the fluid and the diameter of the needle (25G). The histopathologic report was consistent with lymphangioma (Fig. 2a and b). The patient was evaluated at the oncologic surgery service, and surveillance was decided upon, once malignancy of the pancreatic lesion was ruled out.

Lymphangiomas of the pancreas are rare cystic lesions, with around 100 cases reported in the literature.[4] A greater prevalence has been described in females. The most frequent location tends to be at the level of the tail of the pancreas, followed by the head of the pancreas. The lesions are usually multicystic, as in the present case.[5] The most frequent symptoms are abdominal pain and tumor palpation.[2] The diagnosis of this type of pancreatic lesion can be difficult, given that there are no pathognomonic characteristics in imaging studies or biomarkers that enable their identification without a histologic study. In computed tomography, as well as magnetic resonance imaging, the most frequent finding is a multiloculated lesion in 74% of cases and a uniloculated one in 19%.[5] The differential diagnosis of lymphangioma of the pancreas is mainly other cystic lesions. The main ones are serous and mucinous cystadenomas, pancreatic pseudocysts, congenital cysts, and cystic ductal carcinomas.[6] EUS, with or without tissue acquisition, is a useful tool in the diagnosis of the different pancreatic lesions. Only 10 cases of pancreatic lymphangioma diagnosed by EUS have been reported in the literature, 9 of which were through fine-needle aspiration (FNA) and one through biopsy with microforceps.[5,7–9] The biochemical and histologic characteristics tend to be a whitish viscous fluid, with elevated triglyceride levels and abundant lymphocytes and macrophages.[8,9] To the best of our knowledge, this is the first report of a lymphangioma of the pancreas diagnosed with endoscopic ultrasound-guided fine-needle aspiration biopsy (EUS-FNAB). The patient presented with no adverse events, suggesting that EUS-FNAB could be a safe diagnostic method for this type of lesion. Given the usually benign behavior of pancreatic lymphangiomas, surgical resection is unnecessary, if making a definitive diagnosis is possible and there are

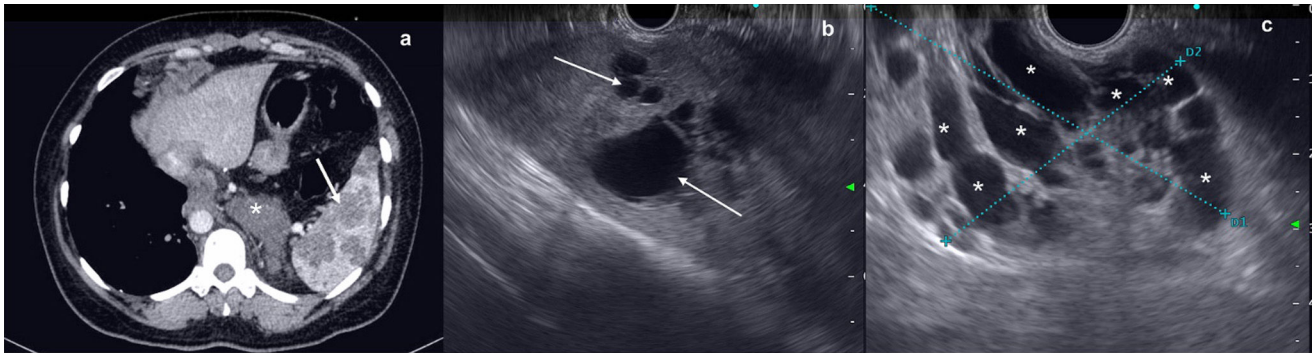


**Figure 1** a) Abdominal CT scan showing multiple rounded lesions in the spleen (arrow), as well as a lesion at the level of the body and tail of the pancreas (asterisk), all with a cystic aspect. b) EUS showing rounded anechoic lesions in the spleen (arrows), consistent with cysts. c) EUS showing cystic lesions in the tail and body of the pancreas (asterisks).

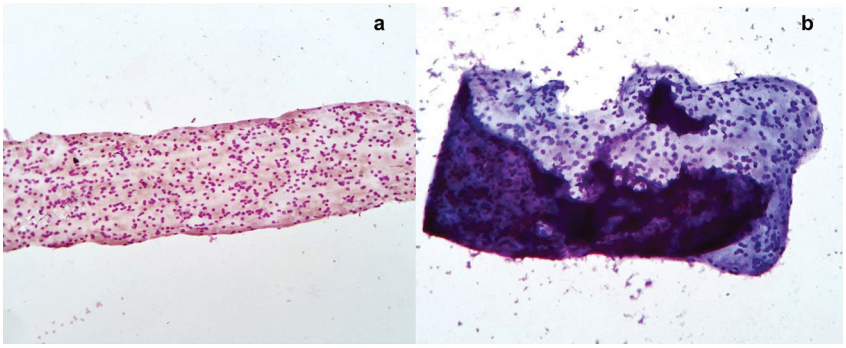


**Figure 2** Lymphocytes immersed in dense proteinaceous material, a finding consistent with lymphangioma. a) Papanicolaou stain, cell block, x400. b) Hemacolor stain, cell block, x400.

no significant symptoms attributed to the lymphangioma. In such cases, patients can be closely monitored with imaging studies.

## Ethical considerations

The authors declare that they requested informed consent from the patient to undergo the procedure described herein. Because this is a case report, authorization by a bioethics committee was not needed. The article contains no personal information that could identify the patient, maintaining his/her anonymity.

## Financial disclosure

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## Conflict of interest

The authors declare that there is no conflict of interest.

## Author contributions

The authors equally contributed to carrying out the present work. All the authors read and approved the final manuscript.

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J.M. Jiménez-Gutiérrez[a,*], J.O. Alonso-Lárraga[a], L.F. Villegas-González[b], A.Y. Ortega-Caballero[a,c]

[a] *Servicio de Endoscopia Gastrointestinal, Instituto Nacional de Cancerología, Ciudad de México, Mexico*
[b] *Servicio de Patología Quirúrgica, Instituto Nacional de Cancerología, Ciudad de México, Mexico*
[c] *Facultad Mexicana de Medicina de la Universidad La Salle, Ciudad de México, Mexico*

* Corresponding author. Servicio de Endoscopia, Instituto Nacional de Cancerología, Av. San Fernando 22, Col. Sección XVI, Alcaldía Tlalpan, CP 14080, Ciudad de México, Mexico. Tel.:+52 5534560265.
*E-mail address:* miguel.jimenezg@comunidad.unam.mx (J.M. Jiménez-Gutiérrez).

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# Chylous ascites secondary to pancreatic pseudocyst: A case report

## Ascitis quilosa secundaria a seudoquiste pancreático: reporte de un caso

The abnormal intraperitoneal accumulation of lymph resulting from extravasation into the abdominal cavity is defined as ''chylous ascites'' (CA) or ''chyloperitoneum''.[1] It is produced after the disruption or obstruction of thoracic or abdominal lymphatic circulation. Considered a rare entity, it has an incidence of approximately one case per 20,000 patients and is related to malignant (lymphomas), congenital, and inflammatory (acute pancreatitis) diseases.

A 61-year-old man had presented with severe acute pancreatitis complicated with a pancreatic pseudocyst one year earlier. He sought medical attention due to nausea and vomiting 5-6 times per day, increase in abdominal perimeter, and weight loss of 12 kg, of 4-month progression. He stated he had no dyspnea, orthopnea, or previous surgeries. Physical examination revealed poor general status; the patient was wasting (weight: 50 kg, height: 1.65 m, BMI: 18.3), dehydrated, with mild pallor, a nonpainful distended abdomen, a positive fluid wave, and no hepatic stigmata.

Laboratory tests showed mild anemia, hypoalbuminemia, and no alterations in blood chemistry, electrolytes, or liver function tests. In addition, fecal elastase was measured and found to be low (130 μg/g), and so treatment with pancreatic enzymes and a proton pump inhibitor was started in the context of a patient with exocrine pancreatic insufficiency.

Paracentesis revealed a whitish, milky fluid (Fig. 1) and its analysis (Table 1) showed a hyperintense serum ascites albumin gradient, with elevated proteins and triglycerides of more than 200 mg/dL, confirming the diagnosis of chylous ascites. Elastography was not carried out due to the presence of ascites, and the echocardiogram showed no alterations.

Magnetic resonance imaging of the abdomen identified a 95 x 110 x 95 mm collection (volume: 496 mL), dependent on the body and tail of the pancreas, and abundant free fluid. The liver and portal venous system appeared normal, and there was no adenomegaly (Fig. 2).

Parenteral nutrition was started with a special regimen and enteral tube feeding with a low-fat diet and medium-chain triglyceride supplementation. Octreotide 100 mcg was administered subcutaneously every 8 h, and the milky peritoneal fluid became clearer, turning to citrine yellow.

The patient underwent percutaneous drainage of the pancreatic pseudocyst due to the lack of access to endoscopic ultrasound (EUS) at our hospital. A total of 500 mL of the pancreatic collection was extracted, leaving catheters in the pancreatic bed and peritoneal cavity, with progressively descending flows. The patient did not undergo surgery. His evolution was favorable, with improvement in tolerating oral intake and weight gain (a current IBM of 22), as well as tomographic evidence of reduced volume of the pancreatic collection and free fluid (Fig. 3).

The diagnosis of CA was made by consensus, with a concentration of triglycerides > 200 mg/dL in the ascitic fluid.[2] Its origin is more frequently associated with portal hypertension, endothelial involvement, and rupture of dilated lymph channels. In causes not associated with portal hypertension (congenital, inflammatory, infectious, neoplastic, postoperative, and traumatic), lymphatic fluid is released by the dilated retroperitoneal vessels into the abdominal cavity through a fistula with the peritoneum.[2,3]

Inflammatory causes are reported to be associated with acute and chronic pancreatitis. Two mechanisms are reported in both: filtration through the lymph vessels damaged by pancreatic enzymes or lymph exudation due to flow obstruction secondary to inflammatory changes in the retroperitoneum adjacent to the pancreas. This

**Figure 1** Milky-appearing ascitic fluid.