References

1. Martínez D, Busto B, Cerezo C. Gastritis inducida por nivolumab en un paciente con melanoma metastásico. Revista de Gastroenterología de México. 2021;86:90–1.
2. Alhatem A, Patel K, Eriksen B Bukhari S, Liu C. Nivolumab-induced concomitant severe upper and lower gastrointestinal immune-related adverse effects. ACG Case Rep J. 2019;6:e00249, http://dx.doi.org/10.14309/crj.0000000000000249.
3. Iranzo I, Huguet J, Suárez P, Ferrer-Barceló L, Iranzo V, Sempere J. Endoscopic evaluation of immunotherapy-induced gastrointestinal toxicity. World J Gastrointest Endosc. 2018;10:392–9.
4. Obeidat A, Silangcruz K, Kozai L Wien E, Fujiwara Y, Nishimura Y. Clinical characteristics and outcomes of gastritis associated with immune checkpoint inhibitors: scoping review. J Immunother. 2022;45:363–9.
5. Vindum H, Agnholt J, Nielsen A Nielsen MB, Schmidt H. Severe steroid refractory gastritis induced by Nivolumab: a case report. World J Gastroenterol. 2020;26:1971–8.

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Peripancreatic lymphangioma: When fine needle aspiration changes the diagnosis—Case report and literary review

Linfangioma peripancreático: cuando la aspiración con aguja fina cambia el diagnóstico. Reporte de caso y revisión de la literatura

Lymphangiomas are rare, slow-growing, benign lesions of vascular origin with lymphatic differentiation.[1,2] They uncommonly involve the abdomen and rarely the pancreas.[1–4] Pancreatic lymphangiomas were first described by Koch in 1913 and are extremely rare, accounting for only 1% of abdominal lymphangiomas and less than 0.2% of all pancreatic lesions.[1–3,5] Lymphangiomas are considered to be of pancreatic origin if they occur adjacent to the pancreas, within the pancreatic parenchyma, or connected to the pancreas via a pedicle.[1,3]

A 44-year-old white male was referred to a Gastroenterology consultation after an abdominal computed tomography (CT) showed a 22 mm hypodense nodular image located between the inferior vena cava (IVC) and the second part of the duodenum externally to the splenomesaic confluent, whose density suggested a liquid/cystic nature. The physical examination and laboratory test results were unremarkable. An endoscopic ultrasound (EUS) was requested and showed a 23 mm cystic lesion with a well-defined and regular wall located between the IVC and the cephalic portion of the pancreas; the lesion was next to the pancreatic parenchyma, raising the hypothesis of a pancreatic cystic lesion. Fine needle aspiration (FNA) biopsy revealed a milky fluid with a yellowish color that might correspond to lymph, suggesting the possibility of a lymphangioma (Fig. 1). Cytological and biochemical exam of the fluid revealed 18,000 nucleated cells/μL with a lymphocyte predominance and a background of proteinaceous material; an elevated triglyceride value (2953.3 mg/dL); amylase (351 U/L) and carcinoembryonic antigen (CEA) (8.84 ng/mL) within normal limits. All these aspects support the diagnosis of peripancreatic lymphangioma. As the patient was asymptomatic, a conservative approach with imaging surveillance was chosen. The patient will be re-evaluated in six months.

Lymphangiomas result from lymphangiectasia, which may be associated with congenital malformations, inflammatory processes, radiotherapy, surgical intervention or previous abdominal trauma.[1–3] These lesions have a female preponderance (unlike our case), have been reported in all age groups and have a variable size (from 3 to 20 cm).[1,3] They are often asymptomatic and discovered as an incidental finding.[2,5] However, the clinical presentation depends on the size, location, mass effect and complications. Common non-specific clinical manifestations include abdominal pain or mass, nausea, vomiting and weight loss, due to the pressure effect.[1,2] Owing to their rarity, this lesion is difficult to diagnose pre-operatively.[2–4] Most of the imaging studies are non-specific.[2,3] EUS can allow a preoperative diagnosis, particularly because of EUS-FNA of the cystic fluid and its consequent cytological and biochemical studies.[2,4,5] Lymphangiomas can be filled with serous, serohematic, or lymphatic fluid.[1] Milky white fluid with a triglyceride level greater than 5000 mg/dL is diagnostic of lymphangioma. Amylase and CEA should be measured to rule out epithelial lesions and pseudocysts. Cytological examination shows a high population of small mature lymphocytes.[2,3] There have been no reports of malignant transformation.[5] As so, when there is a definitive diagnosis, the patient is asymptomatic and the lesion is not locally invasive, a conservative approach with imaging surveillance can be undertaken, as was the choice in our case.[1,2,4,5] Although lymphangiomas are benign neoplasms, they can be locally invasive and may involve adjacent organs.

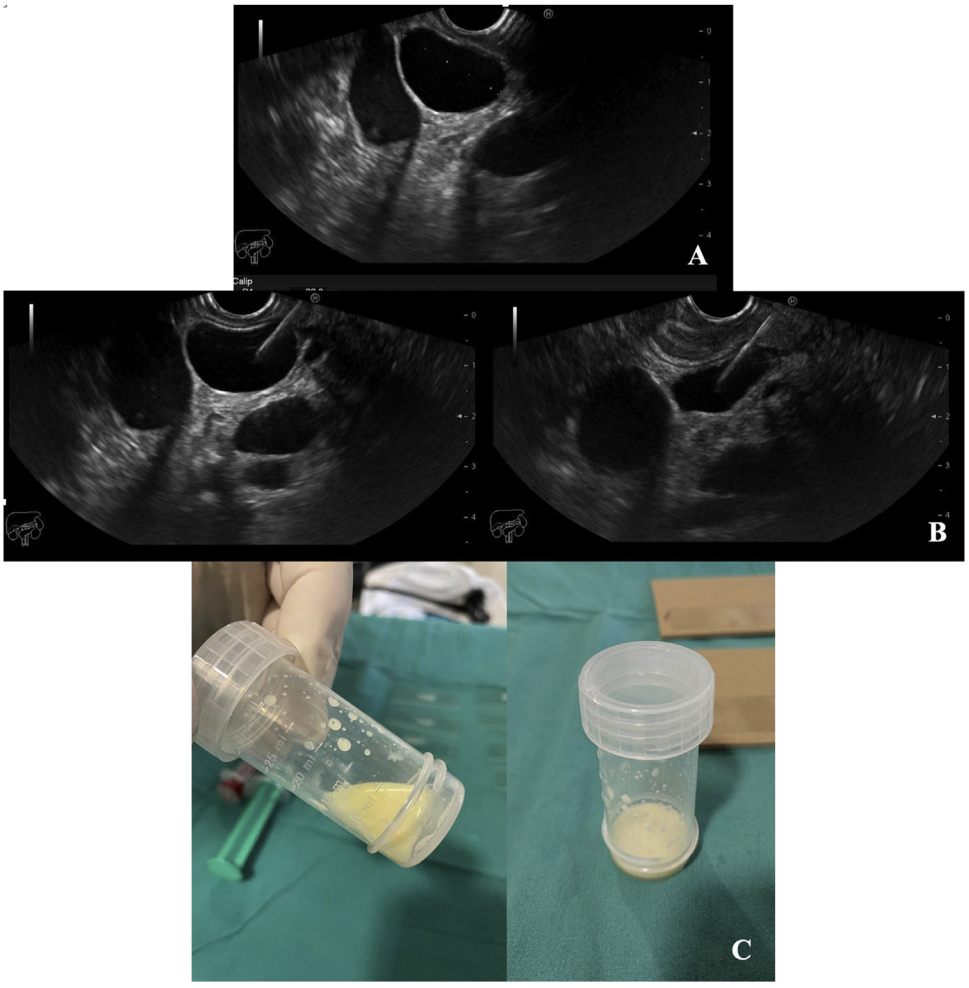

Figure 1 (A) Peripancreatic cystic lesion (peripancreatic lymphangioma); (B) EUS-FNA of cystic lesion; (C) Milky fluid obtained from EUS-FNA.

Complete surgical excision is the curative treatment, providing excellent prognosis and very low incidence of recurrence.[1–4]

It is very important to consider this entity as a differential diagnosis of cystic pancreatic lesions, in order to avoid unnecessary or aggressive treatments.

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Conflict of interest

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References

1. Karajgikar J, Deshmukh S. Pancreatic lymphangioma. J Comput Assist Tomogr. 2019;43:242–4, http://dx.doi.org/10.1097/RCT.0000000000000818.
2. Ray R, Baruah TD, Mahobia HS, Borkar A. Pancreatic lymphangioma: an unusual cause of abdominal lump. Cureus. 2021;13:e19452, http://dx.doi.org/10.7759/cureus.19452.
3. Bal M, Kathuria K, Yadav S, Shrikhande SV. Lymphangioma of pancreas masquerading as a pancreatic cystic neoplasm. Indian J Surg Oncol. 2021;12 Suppl. 1:221–3, http://dx.doi.org/10.1007/s13193-021-01295-8.
4. Pérez FA, Alcaraz ME, López MA. A rare cause of intra-abdominal cysts: pancreatic cystic lymphangiomas. Rev Esp Enferm Dig. 2021;113:682, http://dx.doi.org/10.17235/reed.2020.7671/2020.
5. Viscosi F, Fleres F, Mazzeo C, Vulcano I, Cucinotta E. Cystic lymphangioma of the pancreas: a hard diagnostic challenge between pancreatic cystic lesions-review of recent literature. Gland Surg. 2018;7:487–92, http://dx.doi.org/10.21037/gs.2018.04.02.

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