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CLINICAL IMAGING

Lymphoepithelial Cyst: A Rare Cystic Tumor of the Pancreas Which Mimics Carcinoma

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Pancreatic cystic lesions are usually inflammatory pseudocysts (90%) or, infrequently, neoplastic processes [1]. A lymphoepithelial cyst (LEC) of the pancreas is a rare true cyst of this organ and is lined by squamous epithelial tissue surrounded by lymphoid tissue with germinal centers [2]. This rarely seen lesion should be considered in the differential diagnosis of cystic neoplasms of the pancreas.

We present herein a 55-year old male who had non-specific urinary system complaints. Physical examination and blood tests were normal except for elevated serum CA 19-9 levels of 726 IU/mL (reference range: 0-33 IU/mL). An abdominal CT showed a well-defined low attenuating (30 Hounsfield units, HU) mass (7x8 cm) protruding anteriorly from the pancreatic tail (Image 1). The lesion was multiloculated with a cystic component. Our differential diagnosis included mucinous cystic neoplasms of the pancreas. A laparotomy with a left subcostal incision extending slightly to the left revealed a well-circumscribed encapsulated mass with a multilocular cyst protruding from the pancreatic tail (Image 2). A distal pancreatectomy involving the cystic lesion without a splenectomy was carried out. The gross pathological findings disclosed a multilocular mass, measuring 7x7x6 cm filled with a pasty material. Histologically, the cyst was lined by mature keratinizing squamous epithelium supported by a layer of lymphoid

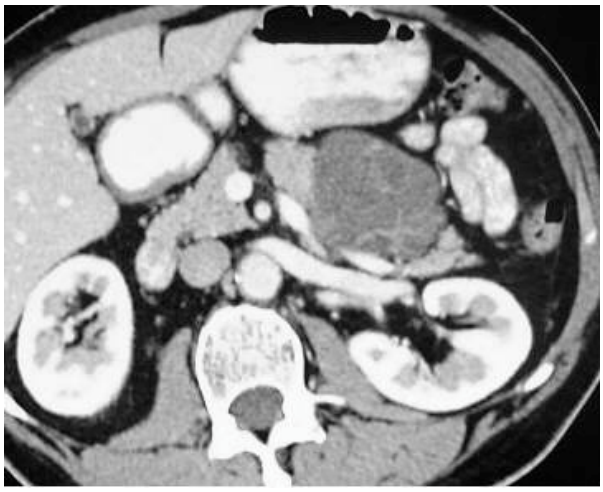

Image 1

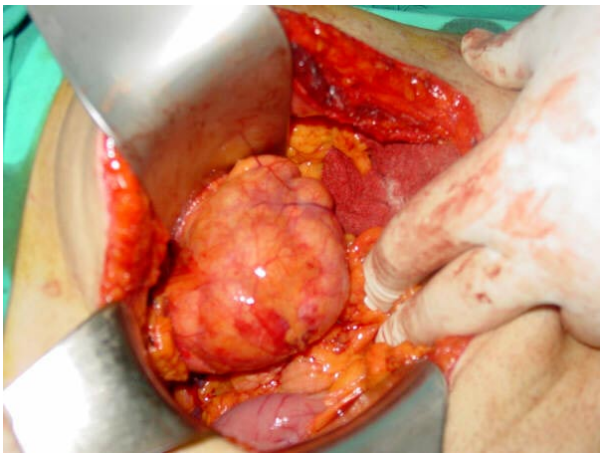

Image 2

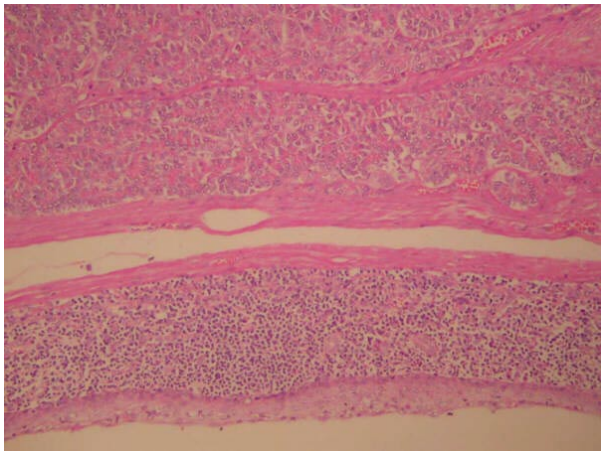

Image 3

tissue (Image 3). During the postoperative course the patient had a low volume pancreatic fistula which lasted 25 days. After resolution of the fistula, he was discharged.

A LEC of the pancreas is an extremely rare, non-neoplastic entity. Truong *et al*. first proposed the term LEC in the literature for the case based on the obvious morphologic similarity of the lesion to its more common counterpart in the parotid gland and the head and neck region [3, 4]. Middle-aged men are predominantly affected with a ratio of 4:1. The vast majority of abdominal LECs are found in the pancreatic parenchyma. Their location in the pancreas is equally distributed from head to tail. Occasionally, LECs may occur in ectopic pancreatic tissue in peripancreatic lymph nodes [5, 6].

Since these lesions are located deep in the pancreatic parenchyma and lack symptoms, they can enlarge to a maximum dimension of about 5.1 cm, as occurred in our case. If this occurs, their only symptom is vague abdominal pain.

Although the histogenesis is unknown, it is hypothesized that the lesion might have arisen from squamous metaplasia of a benign epithelial inclusion in a parapancreatic lymph node. Therefore, it is claimed that a cystic lesion formed as a result of keratinization of the squamous epithelium with invasion into the pancreas could become a LEC of the pancreas [7]. Since LECs of the parotid glands are associated with Epstein-Barr virus infection, Schwarz and Weiss [8] examined the same relationship for LECs of the pancreas by using in situ hybridization tests but they did not show any evidence for Epstein-Barr virus infection. Findings of imaging modalities are conflicting in description. Ultrasonographic findings vary from a solid mass to a cystic lesion [9]. On the other hand, abdominal CT findings in LEC are reported as more descriptive. CT reveals a low-attenuation mass (reflecting the high keratin content of the cyst) with a thin enhancing rim, and multilocular lesions and focal wall calcification [10]. On CT, these cystic lesions appear as a unilocular or multilocular mass, or a cyst with or without calcification. MR imaging does not provide additional data to those of the CT findings except for the relationship between the surrounding tissue and the vascular structures. In our case, we applied all of these modalities, and CT was diagnostic with its multilocular, low-attenuated cystic mass view. However, since these lesions are very rare and can easily mimic cystic neoplasms of the pancreas, we performed surgical excision.

Mandavilli *et al*. mentioned the importance of fine needle aspiration biopsy (FNAB) of the lesion in the preoperative diagnosis [4]. Bolis *et al*. also stated that FNAB was a rapid and reliable method which could be used as the first diagnostic step in the case of cystic masses of the pancreas [11]. However, we did not perform FNAB as an extra invasive procedure, since the local surgical excision was chosen as the final diagnostic and curative method for these lesions. The differential diagnosis included all cystic lesions and cystic teratoma. The characteristic histologic features of the lesion confirm the diagnosis of a LEC. The absence of skin appendages or mature mesenchymal elements exclude cystic teratoma. A LEC of the pancreas is a clinically benign lesion, frequently cured by local excision (enucleation) of the cyst with a rim of normal pancreatic tissue. Sometimes, as in our case, due to the location of the mass, distal pancreatectomy with or without splenectomy can be performed. After local resection, the prognosis is excellent with no incidence of

recurrence [4]. An accurate cytopathological examination may be necessary before more radical surgical approaches are carried out and may help in better preoperative planning. Unfortunately, cytopathological diagnosis is not as straightforward as histopathologic examination of the lesion.

This rare lesion of the pancreas has a uniform and typical clinico-pathological profile leading the surgeon to an easy and accurate diagnosis. These lesions with their benign, curable nature should be considered in the differential diagnosis of cystic lesions of the pancreas.

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Keywords Cysts; Pancreas; Therapeutics

Abbreviations HU: Hounsfield units; LEC: lymphoepithelial cyst

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