

Lymphoepithelial cysts of the pancreas: a management dilemma

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ABSTRACT: Pancreatic lymphoepithelial cysts (LECs) are rare, benign lesions that are typically unexpected post-operative pathological findings. We aimed to review clinical, radiological and pathological features of LECs that may allow their pre-operative diagnosis. Histopathology databases of two large pancreatic units were searched to identify LECs and notes reviewed to determine patient demographic details, mode of presentation, investigations, treatment and outcome. Five male and one female patients were identified. Their median age was 60 years. Lesions were identified on computed tomography performed for abdominal pain in two patients, and were incidentally observed in four patients. Five LECs were located in the tail and one in the body of the pancreas, with a median cyst size of 5 cm. Obtaining cyst fluid was difficult and a largely acellular aspirate was yielded. The pre-operative diagnosis was mucinous cystic neoplasm in all patients. This series of patients were treated distal pancreatectomy and splenectomy. A retrospective review of radiological examinations suggested that LECs have a relatively low signal on T2 imaging and a high signal intensity on T1 weighted images. LECs appear more common in elderly males, and are typically incidental, large, unilocular cysts. Close attention to signal intensity on MRI may allow pre-operative diagnosis of these lesions.

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pancreas

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Introduction

Pancreatic lymphoepithelial cysts (LECs) are rare, benign lesions of the pancreas first described by Lühtrath and Schriefers in 1985,^[1] and are considered true cysts. Kavuturu et al^[2] reported that in the 28 years since the first report of an LEC, 109 cases have so far been documented in the literature. There have been several recent comprehensive reviews documenting the demographic features of LECs, which indicate a strong male preponderance, with lesions of variable size distributed throughout the head, body and tail of the pancreas.^[2-4] Approximately half of the patients present incidentally with the remaining patients being associated with non-specific symptoms such as nausea, vomiting, diarrhoea, abdominal pain, weight loss, and fatigue.^[2-4]

LECs are benign and do not possess malignant potential and thus accurate identification of these lesions is important to avoid unnecessary intervention. However, LECs share radiological and pathological features in common with other pancreatic cystic lesions, some of which are treated by surgical resection.

Until recently, there was no reliable means of pre-operatively diagnosing LECs. The computed tomography (CT) appearances of LECs are those of a well-defined low-attenuation lesions,^[2] often with septations or a multilocular appearance (60%), and they are frequently exophytic.^[5] As the cysts often contain a large volume of keratinized material, the pre-contrast scan may show increased density and appear solid.^[6-7] However, these features do not appear to be universal.

Endoscopic ultrasound (EUS) is of great value for the evaluation of pancreatic cystic lesions.^[8] It has been suggested that LECs have diagnostic features including the presence of squamous epithelium overlying dense lymphoid tissue, with aspirates that may be white and frothy with acellular debris and cholesterol crystals.^[2-4] Nasr et al,^[9] in the largest single series evaluating EUS,

noted that 6 of 9 patients with LECs could be managed non-operatively on the basis of EUS examination and aspiration cytology. However, others suggested that the results of EUS are not reliable, and in the absence of a squamous epithelium rich in lymphocytes it may be difficult to differentiate between LECs and other cystic neoplasms.^[10] It is generally accepted that further work is required to clarify the role of EUS.^[2,4]

Potentially, the most useful modality to establish a pre-operative diagnosis of LEC is magnetic resonance imaging (MRI), with a number of unique features having been identified.^[11-13] The thick walls of the cysts are hypointense on T1 and T2 weighted images and enhance with administration of contrast and keratinous material, where present high intensity on T1 and low intensity on T2 sequences.^[11] Nam and colleagues^[12] used a diffusion-weighted MRI protocol, and found that LECs exhibited a profound restriction of water molecule motion on diffusion-weighted imaging, with or without wall enhancement on contrast-enhanced imaging. Kudo et al^[13] who look at in/out-of-phase imaging reported a slight signal reduction during out-of-phase sequences compared with in-phase sequences, indicating the co-existence of fat and water.

The aim of this study was to review the medical histories and investigations of a cohort of patients with known LEC to identify features that may alert clinicians to the diagnosis of LEC. Given the rare nature of the lesions, two large volume dedicated pancreatic units contributed to the study.

Methods

Patients who had undergone resection of LECs were identified from the pathology department databases of two specialist pancreatic centers (Cleveland Clinic, Cleveland, Ohio, USA and St James's University Hospital, Leeds, West Yorkshire, UK).

Both departments have dedicated pancreatic multi-disciplinary teams, and each case was discussed at a multidisciplinary forum consisting of consultant surgeons, radiologists, endoscopists, pathologists and oncologists.

Data collected included demographic details and mode of presentation whether symptomatic or incidental. For cross-sectional imaging studies, data evaluated included: location; size; number; presence of septations; presence of nodules; rim calcifications; and diameter of the pancreatic duct. Data collated following endoscopic ultrasound-guided fine needle aspiration (EUS-FNA) included color, consistency, cytology, mucin and where aspirate allowed carcinoembryonic antigen (CEA) and amylase.

The indications for resection were recorded as the procedures undertaken, histopathological findings, morbidity and outcome.

Results

Six patients who were identified between February 2002 and March 2010 consisted of five males and one female. The median age was 60 years (range 48-70) at presentation. Table summarises the demographic details, investigations, management and outcome of each case.

Mode of presentation

All lesions of the 6 patients were identified on cross-sectional imaging. Four lesions were found incidentally, and two were confirmed by imaging because of upper abdominal pain of the patients. No patient had symptoms of pancreatic exocrine or endocrine insufficiency.

Investigations and findings

CT characterized the lesions initially in all patients (Fig. 1). The median size was 5.0 cm (range 3.0-6.9) with five lesions located in the pancreatic tail and one in the body. The attenuation of the cysts varied between 23 and 52 Hounsfield units (Hu). There were no features of chronic pancreatitis or other pancreatic disease; the pancreatic duct was normal in each patient.

EUS identified that the lesions were homogenous in nature though one had septations within it (Fig. 2). In half of the lesions, the cyst wall was seen to be irregular. Aspiration was attempted on all lesions; however pathologic analysis was not uniform. In each patient, aspiration was difficult or unsuccessful due to the viscous cyst content, and cyst fluid was in various colors. The level of CEA was analyzed only in one patient and found to be elevated (61 687 IU/L). Amylase analysis was completed in four patients, three of which had negative results and one had an elevated level (1600 IU/L). Five of six patients were analyzed for mucin: three showed negative results and two showed positive ones. Cytologic material was available in all patients and was non-diagnostic with normal and occasional inflammatory cells; however, the consistent feature was a largely acellular aspirate.

MRI was only performed in 3 patients in this series reflecting its historical nature and rarity of LECs. No specific diagnostic features were identified at the time of performance of the MRI scans; however, review of the imaging in the light of recent observations on the typical characteristic features of LECs confirmed that the cyst walls to be hypointense on un-enhanced T1/T2 weighted imaging (Fig. 3). A mild degree of enhancement was

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