

ORIGINAL ARTICLE

Lessons learned from 29 lymphoepithelial cysts of the pancreas: institutional experience and review of the literature

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Abstract

Background: Lymphoepithelial cysts (LECs) are rare pancreatic cystic lesions. Since LECs are benign, preoperative diagnosis is important to differentiate from a cystic neoplasm and avoid unnecessary surgery. The aim of this study was to identify clinical, radiographic and cytopathologic features associated with LECs.

Methods: A retrospective review was performed of patients diagnosed with LEC between 1995 and 2017 at our hospital. Clinicopathologic and radiographic imaging features were documented.

Results: Of 29 patients with pancreatic LEC, 22 underwent surgical resection. The majority were male (n = 24) with a median age of 55 years (range, 21-74). During the evaluation, all patients underwent a CT, with endoscopic ultrasound (EUS) guided fine needle aspiration (FNA) biopsy (n = 22) and/or MRI/MRCP (n = 11) performed in a smaller number of patients. A combination of exophytic tumor growth on imaging and the presence of specific cytomorphologic features on the EUS-FNA cytology biopsy led to the correct diagnosis of LEC and prevention of unnecessary surgery in 7 patients.

Discussion: Differentiating LECs from premalignant pancreatic cystic neoplasms remains difficult. Findings of an exophytic growth pattern of the lesion on abdominal imaging and the presence of specific cytomorphologic features in the EUS-FNA biopsy could help clinicians diagnose LEC preoperatively.

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Introduction

Lymphoepithelial cysts (LECs) are rare benign pancreatic cystic lesions. First described in 1985 by Luchtrath and Schriefer, the term LEC was proposed two years later by Truong and colleagues.^{1,2} They described pancreatic cysts filled with keratinous material, and lined by mature keratinizing squamous epithelium with underlying dense lymphoid tissue including

germinal centers. Since the first publication, examples of LECs are sparse, with only 185 patients reported in low-volume case series and case reports.

Pancreatic LECs account for approximately less than 0.5% of all pancreatic cysts.^{3,4} However, the increasing use of abdominal cross-sectional imaging has resulted in an increase in the detection of asymptomatic benign pancreatic cysts.^{5,6} LECs are often misdiagnosed as a pancreatic cystic neoplasm, resulting in surgical resection. Since LECs are entirely benign lesions, correct preoperative diagnosis is important in order to avoid unnecessary surgery. However, there are no comprehensive preoperative criteria to distinguish between a potential cystic neoplasm of

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the pancreas and a LEC. The aim of this study was to evaluate the diagnostic features associated with LECs and to determine which criteria would help identify LECs preoperatively by performing a review of relatively large institutional case series and a systematic review of the literature.

Methods

Patient selection

Approval from the institutional review board was obtained for this study. A retrospective review of the institutional pathology archives and the pancreatic cyst database from 1995 to 2017 was performed to identify resected and non-resected LEC.

The following clinicopathologic and radiographic data were extracted and analyzed: gender, age, race, symptoms upon initial presentation, past medical history and laboratory results including serum carbohydrate antigen (CA) 19-9 and serum carcinoembryonic antigen (CEA). Original images of the endoscopic ultrasound (EUS) procedure, computed tomography (CT), magnetic resonance imaging (MRI) and magnetic resonance cholangiopancreatography (MRCP) were reviewed and scored for radiographic imaging findings (E.K.F. and A.M.L.). Additionally, all available archived slides were reviewed by an experienced pancreatic cytopathologist (C.J.V.) and pathologist (R.H.H.) to confirm the diagnosis of LEC and to assess cytopathologic features of the FNA biopsy and pathologic characteristics of the surgical specimen.

Systematic review of the literature

A systematic search was performed in PubMed and Embase libraries for articles published up to 1 October 2017. Search terms were '[Lymphoepithelial cyst] AND [pancreas OR pancreatic]'. Titles and abstracts were screened and references of the identified manuscripts were examined for potentially relevant articles not identified by the initial search. Case reports and case series describing patient with LEC were included, while review articles that did not present new unique patients were excluded. Subsequently, clinicopathologic and radiographic data were extracted for each patient and added to the institutional series to create a combined and comprehensive cohort of patients with a LEC of the pancreas.

Statistical analysis

Continuous variables are expressed as median and range while categorical variables are presented as numbers and percentages of the group from which they are derived. Statistical analysis was performed with SPSS statistical software version 25.0 (SPSS Inc., Chicago, Illinois).

Results

A total of 29 patients were diagnosed with LEC between January 1995 and January 2017 at the Johns Hopkins Hospital. Twenty-

two patients underwent surgical resection while 7 patients had a preoperative diagnosis of LEC and were treated conservatively with interval surveillance instead of surgical resection.

Patient characteristics

Patient characteristics stratified for resected and non-resected LECs are shown in [Supplemental Table 1](#). Median age at diagnosis was 55 years (range, 21–74) and the majority of patients were male ($n = 24$). Four patients had a prior history of cancer (2 prostate, 1 uterine, and 1 hepatocellular carcinoma) and none of the patients were known to be HIV positive. Eleven of the LECs in the surgical cohort were incidentally detected, while all seven patients who underwent surveillance were incidentally identified. Some patients reported non-specific complaints such as abdominal pain ($n = 12$) and/or weight loss ($n = 4$). Serum CA 19-9 and CEA values were available for 10 and 4 patients, respectively. Three patients had elevated (≥ 37 U/mL) serum CA 19-9 (median value of 24 U/mL (14–199)). Two patients had elevated (≥ 3 ng/mL) serum CEA.

Imaging features

All 29 patients underwent a CT scan. Additional EUS ($n = 22$) and/or MRI/MRCP ($n = 11$) imaging procedures are shown in [Table 1](#). Median maximum cyst diameter on CT was 4.0 cm (range, 2.0–16.0 cm). LEC were most commonly identified in the body/tail of the gland ($n = 22$). Eleven lesions were exophytic, defined as arising from the pancreas and located on the edge of the gland, projecting outwards. Enhancement of the cyst wall and nodules were seen in 5 and 3 patients, respectively.

On CT, LECs were hypodense or cystic and occasionally heterogeneous ([Fig. 1](#)). Calcification within the cyst was uncommon, while pancreatic duct dilatation or communication between the cyst and the duct was never present. Three-dimensional mapping of CT images was performed in 13/22 resected patients and in 7/7 correctly diagnosed patients ([Fig. 1](#)). On MRI, the lesions were generally hypointense on T1-weighted MR images and hyperintense on T2-weighted images.

Endoscopic ultrasound features for all patients are shown in [Table 1](#) and more specifically for correctly diagnosed patients in [Table 2](#). The majority of the LECs (14/16) were well defined and more than half (9/16) had a mixed hypoechoic and anechoic echogenicity. A solid component was perceived in 7/11 patients in the resected group, compared to 1/7 in the patients who underwent surveillance. Additionally, the lesions appeared to have an exophytic growth pattern in 5/7 correctly diagnosed LECs.

Fine needle aspiration

Sixteen patients underwent EUS-FNA during the work-up of their pancreatic cyst. Sufficient material was obtained for cytopathologic characterization in 14 patients. The EUS-FNA sample led to an initial suspicion of LEC in 4/14 patients while in 3 other patients FNA biopsy confirmed the diagnosis of LEC after suspicion based on imaging results. In the remaining 7 patients, the

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