

## CASE REPORT

# EUS-Guided Trucut Biopsies May Enable the Diagnosis of Lymphoepithelial Cysts of the Pancreas. Report of Two Cases

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### ABSTRACT

**Context** Lymphoepithelial cysts are rare benign lesions of the pancreas. Endoscopic ultrasound guided fine needle aspiration (FNA) has reportedly been helpful in reaching a diagnosis of lymphoepithelial cyst. In cases where it may be difficult to obtain a diagnosis using FNA a histological sample may be obtained using EUS guided Trucut biopsy. **Case reports** We describe two cases of lymphoepithelial cysts where a diagnosis of lymphoepithelial cyst was made using EUS guided Trucut biopsy. **Conclusion** In some case, lymphoepithelial cysts may present a diagnostic dilemma. EUS with FNA and Trucut biopsy may enable the clinician to make a definitive diagnosis.

### INTRODUCTION

Lymphoepithelial cysts of the pancreas are rare lesions [1]. They are considered benign with no known risk of malignant transformation [1]. Due to variable and non-specific radiologic findings [2], lymphoepithelial cysts can be confused with other pancreatic cystic lesions including neoplasms with malignant potential.

Endoscopic ultrasound with fine needle aspiration (FNA) has been reported to be helpful in obtaining a preoperative diagnosis. However, FNA may result in scant material and confusion with mucinous cystic neoplasm can be caused by contaminating gastrointestinal mucosa [3]. Thus, it may be difficult to achieve a cytologic diagnosis. We describe two cases in which EUS-guided core biopsies with a Trucut needle were helpful in obtaining a diagnosis.

### CASE 1

A 35-year-old male patient presented with periumbilical and right upper quadrant abdominal pain. His past medical history was negative for pancreatitis. Liver enzymes, white blood cell count, and serum amylase and lipase levels were within normal limits.

After an unremarkable abdominal plain film, the patient underwent a CT scan of the abdomen and pelvis, which showed two pancreatic lesions. These were hypodense with mixed solid and cystic appearance. They measured 20x15 mm and 20x20 mm in diameter, respectively. One lesion was located in the pancreatic tail and the second one in the pancreatic body, along the inferior margin of the stomach. Endoscopic ultrasound (EUS) was performed, which showed a 2.3x1.5 cm pancreatic tail lesion with slightly lobulated contour, granular echotexture and a small anechoic (cystic) component (Figure 1a). In addition, a 2.0x1.0 cm solid appearing oval mass with smooth contour and similar granular echotexture was identified adjacent to the pancreatic body (Figure 1b). EUS-FNA with a 22 G needle (Cook-Medical, Bloomington, IN, USA) was performed of both lesions in a transgastric approach. Air dried smears were stained using the Diff-Quick method and assessed immediately by an experienced cytopathologist. On-site cytology assessment showed anucleate squamous cells and crystals. EUS-guided Trucut biopsies were also performed (one pass per lesion) using a 19 G Trucut needle (Quick-Core, Cook-Medical, Bloomington, IN, USA). Prior to sending the tissue cores for histology, these were touched on glass slides, air dried and stained using the Diff-Quick method to obtain touch imprint cytology ("touch prep") as described elsewhere [4]. On-site assessment of these touch preps showed crystals and lymphoid tissue in the pancreatic tail lesion and anucleate squamous cells in the pancreatic body lesion. (Figure 1c) The final histology showed stratified squamous epithelium with prominent keratin debris, as well as a prominent lymphoid infiltrate

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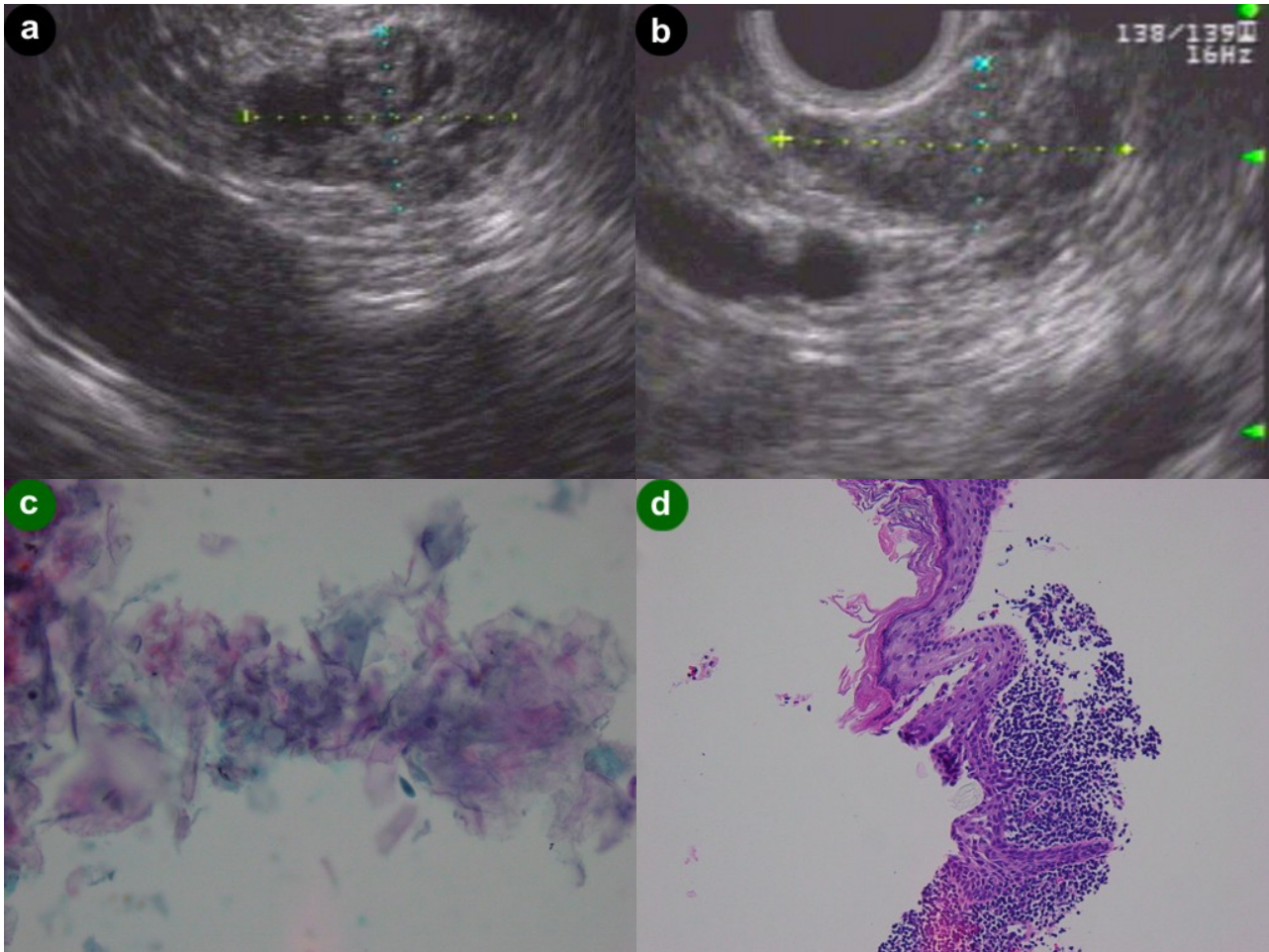
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**Figure 1. Case 1. a. EUS:** pancreatic tail lesion. 2.3x1.5 cm hypoechoic, slightly lobulated contour, granular echotexture, cystic lesion. **b. EUS:** pancreatic body lesion. Hypoechoic lesion with a granular echotexture. 2.0x1.0 cm in size. **c. On-site touch imprint cytology:** anucleate squamous cells. **d. Histology:** stratified squamous epithelium with prominent keratinaceous debris and adjacent lymphoid infiltrate without atypia.

without atypia, which was adjacent to the squamous epithelium (Figure 1d). These findings were diagnostic of a lymphoepithelial cyst of the pancreas. The initial symptoms resolved with no intervention and therefore the decision to follow the lesion with periodic CT scans was made. Follow up CT scan at 3 months demonstrated no change in size of the pancreatic tail cyst, but the lesion had developed a new peripheral solid rim. Although benign changes of lymphoepithelial cyst with proliferation of the fibrous wall were most likely, a malignant change could not be excluded. The patient remained asymptomatic, but in light of the interval change surgical intervention was recommended and the patient underwent a distal pancreatectomy and splenectomy. Both cystic lesions were removed and histology of the surgical specimens confirmed the preoperative diagnosis of lymphoepithelial cyst. On follow up one year later the patient had been doing well, with no complaints.

## CASE 2

A 54-year-old male presented with complaints of vague abdominal pain. Initial laboratory data was normal. An abdominal CT scan showed a non-enhancing, 2.2x2.0 cm cystic mass, located in the body of the pancreas.

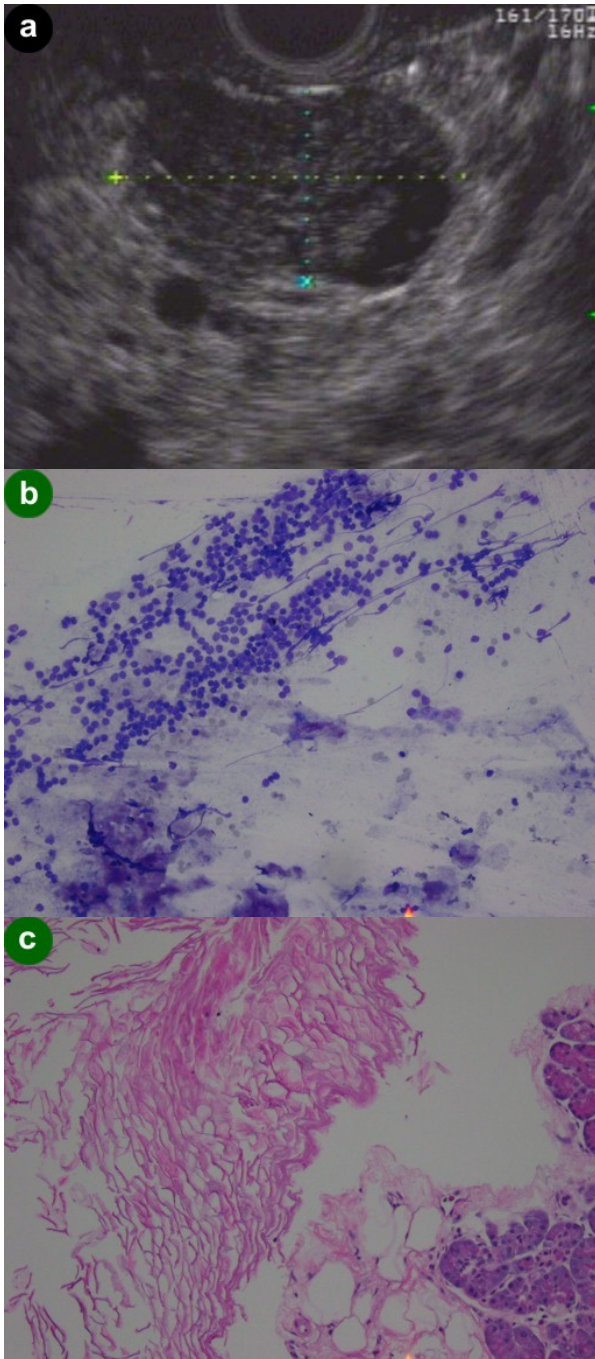
This mass was confirmed by an MRI. An EUS revealed a 2.6x1.5 cm oval, hypoechoic mass with smooth contour, granular echotexture and anechoic (cystic) component (Figure 2a). EUS-guided Trucut biopsies were obtained with a 19 gauge EUS-Trucut needle (Quick-Core, Cook-Medical, Bloomington, IN, USA). Touch preps were made for rapid assessment and cytology. The touch preps contained anucleate squamous cells with scattered viable benign squamous cells and lymphoid tissue (Figure 2b) suggestive of lymphoepithelial cyst. The tissue cores were sent for histopathology and the histologic findings of stratified squamous epithelium with prominent keratin debris (Figure 2c) along with the touch prep findings allowed a definitive diagnosis of lymphoepithelial cyst.

No intervention was performed. At follow-up 6 months later the patient reported resolution of his abdominal discomfort and no new gastrointestinal symptoms.

## DISCUSSION

Lymphoepithelial cysts are rare, slow growing lesions of the pancreas that were first described by Luchtrath and Schriefers in 1985 [5, 6]. Pathogenesis and histogenesis are both unclear and no malignant transformation has been reported. Rarely these lesions

are described to undergo focal sebaceous [7] or columnar glandular differentiation [8]. Histopathologic features are distinct and include unilocular (or rarely multilocular) cysts with a lining of keratinized stratified squamous epithelium without atypia surrounded by a fibrous wall containing varying amounts of benign lymphoid tissue which may include germinal centers [7, 8].



**Figure 2. Case 2. a. EUS:** pancreatic body lesion. Hypoechoic, granular echotexture, with a cystic component measuring 2.6x1.5 cm in size. **b. On-site touch imprint cytology:** anucleated squamous cells and lymphocytes. **c. Histology:** stratified squamous epithelium with prominent keratinaceous debris and adjacent benign pancreatic tissue.

The preoperative diagnosis of lymphoepithelial cyst is difficult, since radiological and clinical criteria are not always reliable. These cysts may be mistaken for a premalignant neoplastic process preoperatively as there may be an increase in both the serum and cystic fluid tumor markers (CA 19-9 and CEA) [1, 3, 6].

Previously, CT scans provided the most accurate non-invasive diagnosis [6]. Currently, however EUS appears to be the most useful diagnostic modality because it provides superior spatial resolution and enables tissue sampling by EUS-FNA and/or EUS-Trucut biopsy.

The appearance on EUS may vary from a solid to cystic mass, or mixed solid-cystic characteristics. The EUS-characteristics in our cases were a smooth contour which may be slightly lobulated and a hypoechoic, granular echopattern with anechoic (cystic) components.

EUS guided FNA is most commonly used to reach a preoperative diagnosis. Cytology samples show squamous cells and keratinous debris. Admixed lymphocytes, histiocytes, and occasionally scattered plate-like cholesterol crystals may be seen [7, 9]. A thick milky, creamy, or frothy appearance of aspirate has also been described in a recent case series [10]. Cell blocks contain tissue fragments composed of keratinized squamous cyst lining with an adjacent stromal layer containing a lymphocytic infiltrate [9]. These findings are quite sensitive in diagnosis of lymphoepithelial cyst and may help exclude other types of cysts including those with malignant potential. However, similar findings including a mixture of squamous cells and lymphocytes have been reported in other pancreatic lesions such as dermoid cysts, splenic epidermoid cysts, adenosquamous carcinoma, and metastatic squamous cell carcinoma [9]. In addition, cytopathologic diagnosis using FNA may not always be as straightforward. Problems encountered may include contamination of the aspirated sample by interposing tissue picked up during the needle pass. This may lead to admixture of mucin and glandular type epithelium (gastric or small intestinal) [11], which may cause confusion with mucinous cystic neoplasms. Further, the cytopathologic characteristic of lymphoepithelial cyst, especially the presence of squamous epithelial cells and/or keratinized material, can be absent from the cyst fluid [12]. Elevated tumor markers in the cyst fluid, i.e. CEA and C19-9 levels, may confound the picture [13]. Thus, if a definitive diagnosis is not reached, surveillance with cross sectional imaging or surgical resection is usually recommended.

In order to avoid surgery and to reach a definitive diagnosis we propose the use of EUS-guided Trucut biopsy. This sampling method has the advantage of yielding a tissue core which allows histologic assessment. This may be more specific and easier to interpret than FNA-cytology. While it is debatable if histologic specimens are always required, EUS Trucut

biopsy could be used in cases where on-site cytology assessment of FNA smears is non-diagnostic.

## CONCLUSION

Lymphoepithelial cyst may present a diagnostic dilemma. EUS with FNA and Trucut biopsy may enable the clinician to make a definitive diagnosis.

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**Conflict of interest** None of the authors who participated in this study have commercial or other associations that might pose a conflict of interest

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