Lymphoepithelial Cyst of the Pancreas Tail. 
Case Report and Review of the Literature

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ABSTRACT

Context Lymphoepithelial cyst of the pancreas is a lesion that comprises a stratified squamous epithelial lining atop dense lymphoid tissue.

Case report We report our case of a 56-year-old man presented with recurrent abdominal pain. A CT scan showed a cystic lesion between the spleen and the pancreas. A distal pancreatectomy with splenectomy was performed. All pancreatic tissue was submitted for histologic examination. The patient recovered on the ward. On postoperative day two, the patient started eating an advanced diet. He was discharged on postoperative day four. The cyst is comprised of benign stratified squamous epithelium atop dense lymphoid tissue, which was consistent with cyst.

Conclusion Good preoperative radiological anatomical mapping, good communication and good cooperation between the pathologist and the surgeon are essential to resect the lymphoepithelial cyst and exclude malignancy.

INTRODUCTION

In 1987, Truong et al. coined the name of lymphoepithelial cyst of the pancreas [1], even though it had been described earlier. About 88 cases have been previously described [2, 3]. Pathologically, the lesion is comprised of a stratified squamous epithelial lining atop dense lymphoid tissue. The present case report is that of a patient who presented to the emergency room with left upper quadrant pain. The workup showed a pancreatic tail mass which was suspicious for cancer.

CASE REPORT

A 56-year-old man presented with recurrent abdominal pain, mostly in the left upper quadrant and epigastrium. Past medical history was positive for a bout of pancreatitis two years earlier. A complete blood count, electrolytes, and liver function tests were all normal. A CT scan showed a 2.5x3.6x4cm cystic lesion between the spleen and the pancreas (Figure 1). A MRI further localized

Figure 1. CT view of the lymphoepithelial cyst tumor at the level of the pancreatic tail.
the cyst to the distal pancreas adjacent to the hilum of the spleen (Figure 2). The patient was taken to the operating room electively. After anesthesia was induced, a left subcostal incision was performed. The stomach was reflected to expose the pancreas and spleen. Upon further dissection, no definite mass was identified. The intraoperative ultrasound could not identify the cyst. During the procedure, the cyst became unapparent and could not be identified by simple palpation. Because careful cooperation between the surgeon and the pathologist occurred, the cyst was identified histologically and cancer was excluded. Because radiological examination showed the mass to have been apposed to both pancreas and spleen in close proximity to the splenic artery and vein, distal pancreatectomy and splenectomy was performed. A Jackson-Pratt drainage was left in place in the left upper quadrant. The patient recovered on the ward and the drainage was taken out postoperative day two after the patient started eating an advanced diet and discharged on postoperative day 4. The patient returned to his normal activities. At pathologic examination, no gross lesion was appreciated; the entire pancreas segment was evaluated histologically. In three distal sections, adjacent to pancreas parenchyma, lay a cyst lining comprised of benign stratified squamous epithelium atop a lymphoid tissue layer (Figures 3 and 4). The pancreas also showed focal fibrosis and inflammation. The spleen showed congestive splenomegaly. No cancer was seen, as expected in this type of cyst.

**DISCUSSION**

Lymphoepithelial cysts of the pancreas are rare lesions [2, 3, 4, 5, 6, 7]. About 88 cases have been reported. Most often, the lesions appear in middle aged men, as is true in this case. The most common symptoms are abdominal pain, nausea and vomiting, anorexia and weight-loss, general malaise and altered bowel habits, but many patients are asymptomatic, coming to the surgeons attention as incidental radiological findings. The cysts can occur at any location in the

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**Figure 2.** MRI view of the lymphoepithelial cyst tumor.

**Figure 3.** Low power view of lymphoepithelial cyst showing cyst lining on left and pancreatic parenchyma on right. (Hematoxylin and eosin. Original magnification 40x).

**Figure 4.** High power view of lymphoepithelial cyst lining showing benign stratified squamous epithelium overlaying a layer of lymphoid tissue. (Hematoxylin and eosin. Original magnification 200x).
pancreas, have been as large as 13 cm. Most such cysts are multilocular. The differential diagnosis is cancer and the final diagnosis can only be done after resection.

The etiology is unclear, but the cysts have arisen in association with Sjogren’s disease and AIDS; this patient had neither of these conditions. It has been posited that the lesion is an enlarged epithelial inclusion in a peripancreatic lymph node that has undergone squamous metaplasia; [8] the location of our patient’s lesion, immediately apposed to the pancreatic parenchyma, further supports this hypothesis. Others have identified these cysts in ectopic pancreatic tissue in peripancreatic lymph nodes [9, 10]. Although one might suggest the lesion arises from an obstructed pancreatic duct [7], the presence of a zone of lymphoid tissue cannot be explained on this basis. Others have posited these as being benign epithelial inclusions embedded in the pancreas or brachial cleft cysts fused with the pancreatic anlage during embryogenesis [1, 6].

The differential diagnosis, includes primary splenic cysts, pseudocysts, cystadenocarcinomas, left adrenal cysts, cystic aneurysms, retroperitoneal cysts, duplication cysts, and mesenteric cysts; most of these diagnoses can be excluded by MRI, as was done in this case. CT scans usually show a low-attenuation mass with a thin enhancing rim and focal wall calcification, as in our case [11]. Ultrasoundography can be used to further support the cystic nature of these lesions [12]. Most cysts are radiologically consistent with a pancreatic pseudocyst; fine needle aspiration may be of utility to exclude malignant cells [7, 13, 14]. With all of the risks related in our case, given the vicinity of the splenic hilum that was never taken into consideration. From our case, we learned that the cyst is not easily defined by gross palpation during the surgical exploration. Therefore, it is of utmost importance to have an anatomical map with CT and MRI. A good and quick pathological evaluation on the resected specimen is important to document that the cyst was resected. A good margin taken during the resection of the pancreatic segment is a key to find potential malignant lesion. The two most important lessons learned from this case are: 1) preoperative anatomical mapping is extremely important in the surgical strategy; 2) good communication and cooperation with the pathologist plays a key role in the optimization of the surgical resection, particularly if the cyst is not palpable in the operating room.

CONCLUSION

Lymphoepithelial cyst of the pancreas is a rare disease that often presents as an incidental radiological finding, but may, as in this case, cause symptoms that require an emergency room visit. Radiological procedures are of great utility in ruling out other diagnoses. Percutaneous aspiration may be of use, but for most patients surgical exploration will be required to exclude cancer. For the cysts that are close to the splenic hilum, distal pancreatectomy and splenectomy are indicated to avoid potential lesions to the spleen and complete specimen resection to exclude cancer.

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