JOP. J Pancreas (Online) 2008; 9(1):46-49.

CASE REPORT

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

Eldo E Frezza[1], Mitchell S Wachtel[2]

[1]Division of General Surgery, Department of Surgery, and [2]Department of Pathology, Texas Tech University Health Sciences Center. Lubbock, TX, USA

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

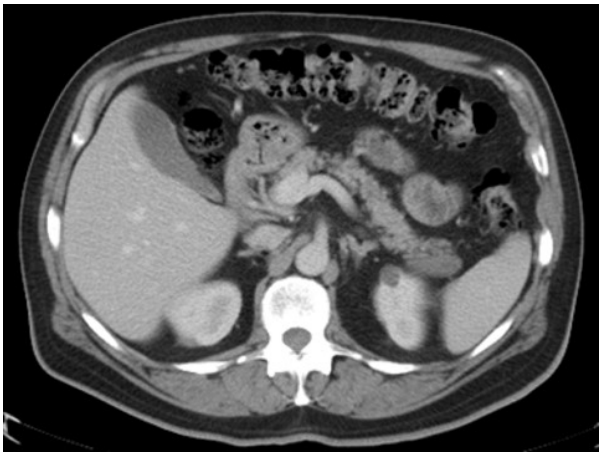

Figure 1. CT view of the lymphoepithelial cyst tumor at the level of the pancreatic tail.

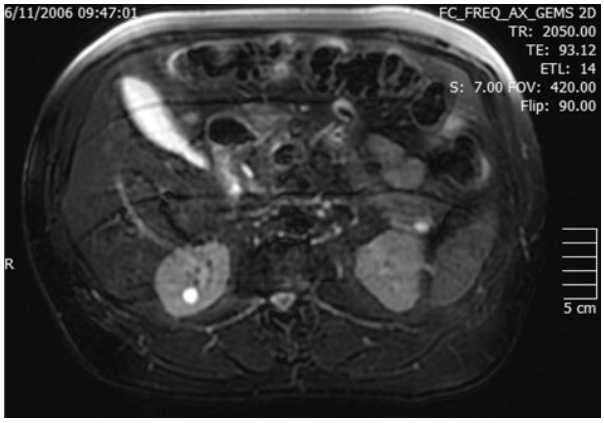

Figure 2. MRI view of the lymphoepithelial cyst tumor.

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

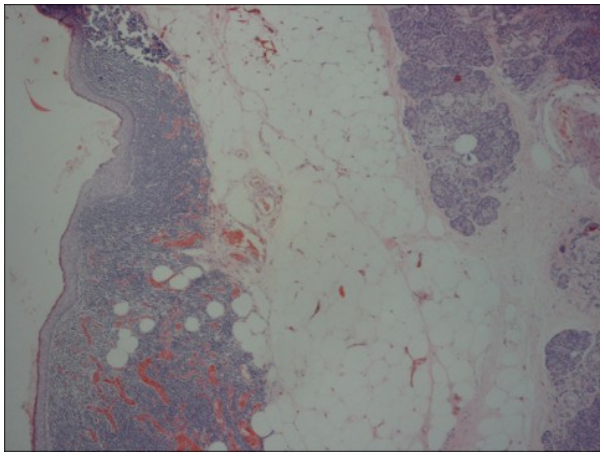

Figure 3. Low power view of lymphoepithelial cyst showing cyst lining on left and pancreatic parenchyma on right. (Hematoxylin and eosin. Original magnification 40x).

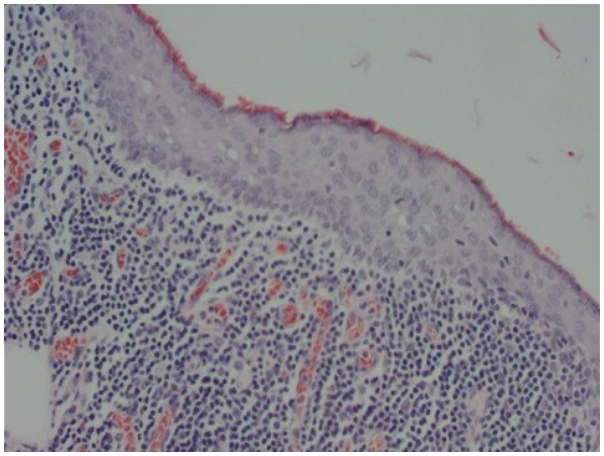

Figure 4. High power view of lymphoepithelial cyst lining showing benign stratified squamous epithelium overlaying a layer of lymphoid tissue. (Hematoxylin and eosin. Original magnification 200x).

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]. Ultra-sonography 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.

Received September 26th, 2007 - Accepted November 7th, 2007

Keywords Lymphatic Vessel Tumors; Neoplasms, Glandular and Epithelial; Pancreatic Neoplasms; Pathology

Acknowledgements Poster presented at the Southeastern Surgical Congress Meeting; Savannah, GA, USA; February 10-13, 2007

Correspondence
Eldo Ermenegildo Frezza
Department of Surgery
Division of General Surgery
Texas Tech University Health Sciences Center
3601 4th Street, MS 8312
Lubbock, TX 79430
USA
Phone: +1-806.743.2460 Ext 263
Fax: +1-806.743.2113
E-mail: eldo.frezza@ttuhsc.edu

Document URL: http://www.joplink.net/prev/200801/09.html

References

1. Truong LD, Rangdaeng S, Jordan PH Jr. Lymphoepithelial cyst of the pancreas. Am J Surg Pathol 1987; 11:899-903. [PMID 3674287]

2. Liu J, Shin HJ, Rubenchik I, Lang E, Lahoti S, Staerkel GA. Cytologic features of lymphoepithelial cyst of the pancreas: two preoperatively diagnosed cases based on fine-needle aspiration. Diagn Cytopathol 1999; 21:346-50. [PMID 10527483]

3. Adsay NV, Hasteh F, Cheng JD, Bejarano PA, Lauwers GY, Batts KP, et al. Lymphoepithelial cysts of the pancreas: a report of 12 cases and a review of the literature. Mod Pathol 2002; 15:492-501. [PMID 12011254]

4. Adsay NV, Hasteh F, Cheng JD, Klimstra DS. Squamous-lined cysts of the pancreas: lymphoepithelial cysts, dermoid cysts (teratomas), and accessory-splenic epidermoid cysts. Sem Diagn Pathol 2000; 17:56-65. [PMID 10721807]

5. Kim YH, Auh YH, Kim KW, Lee MG, Kim KS, Park SY. Lymphoepithelial cysts of the pancreas: CT and sonographic findings. Abdom Imaging 1998; 23:185-7. [PMID 9516512]

6. Tateyama H, Tada T, Murase T, Fujitake S, Eimoto T. Lymphoepithelial cyst and epidermoid cyst of the accessory spleen in the pancreas. Mod Pathol 1998; 11:1171-7. [PMID 9872647]

7. Neyman EG, Georgiades CS, Horton KH, Lillemoe KD, Fishman EK. Lymphoepithelial cyst of the pancreas--evaluation with multidetector CT. Clin Imaging 2005; 29:345-7. [PMID 16153542]

8. Sako S, Isozaki H, Hara H, Tsutsumi A, Tanigawa N. Cystic lymphoepithelial lesions of the pancreas and peripancreatic region: report of two cases. Surg Today 1999; 29:467-71. [PMID 10333422]

9. Strapko A, Botash RJ, Murthy UK, Landas SK. Lymphoepithelial cyst of the pancreas: a case report and review of the literature. Dig Dis Sci 1998; 43:870-4. [PMID 9558046]

10. Tsuchiya Y, Suzuki S, Sakaguchi T, Kojima Y, Okamoto K, Kurachi K, et al. Lymphoepithelial cyst of the pancreas report of a case. Surg Today 2000; 30:856-60. [PMID 11039720]

11. Koga H, Takayasu K, Mukai K, Muramatsu Y, Mizuguchi Y, Furukawa H, et al. CT of lymphoepithelial cysts of the pancreas. J Comput Assist Tomogr 1995; 19:221-4. [PMID 7890845]

12. Yamamoto K, Fujimoto K, Matsushiro T, Ota K. Lymphoepithelial cyst in pancreas: a case report. Gastroenterol Jpn 1990; 25:758-61. [PMID 2279638]

13. Mandavilli SR, Port J, Ali SZ. Lymphoepithelial cyst (LEC) of the pancreas: cytomorphology and differential diagnosis on fine-needle aspiration (FNA). Diagn Cytopathol 1999; 20:371-4. [PMID 10352910]

14. Bolis GB, Farabi R, Liberati F, Macciò T. Lymphoepithelial cyst of the pancreas. Report of a case diagnosed by fine needle aspiration biopsy. Acta Cytol 1998; 42:384-6. [PMID 9568141]