

CASE REPORT

Pancreatic Lymphoepithelial Cyst with an Intracystic Papillary Projection. Report of a Case

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ABSTRACT

Context Lymphoepithelial cysts of the pancreas show various presentations on imaging studies often making a differential diagnosis difficult. **Case report** A 58-year-old man was admitted to our hospital with epigastric discomfort and abdominal bloating. Abdominal US was carried out and demonstrated a cystic lesion 5 cm in diameter which included an intracystic projection into the pancreatic body. This intracystic projection was also seen on CT and MR images. On FDG-PET images, the projection was positively visualized. Since we could not exclude a pancreatic cystic neoplasm, a distal pancreatectomy was performed. Postoperative pathological examination demonstrated that the lesion was a lymphoepithelial cyst of the pancreas. There were no atypical cells in the intracystic projection. **Conclusion** Resection is inevitable when a true pancreatic neoplasm cannot be excluded.

INTRODUCTION

A lymphoid epithelial cyst of the pancreas is a rare lesion. It was first described in 1985 [1]. Since then, about 100 cases have been reported in the literature. The differential diagnosis is often difficult. One reason is that imaging studies of lymphoid epithelial cysts show various presentations [2] and surgery is usually indicated. We herein report the case of a lymphoid epithelial cyst with an intracystic papillary growth which was positively visualized by positron emission tomography using FDG-PET. The intracystic papillary projection of a lymphoid epithelial cyst is relatively rare in clinical practice.

CASE REPORT

A 58-year-old man was admitted to our hospital with a three-month history of abdominal discomfort and bloating. There was no body weight loss or fever, and he had no history of pancreatitis. Abdominal US was carried out and showed a cystic lesion in the pancreatic body. A small projection in the lumen was apparent (Figure 1). Laboratory evaluation demonstrated a

serum DUPAN-2 level of 193 U/mL (reference range: 0-150 U/mL); the other values were normal. Abdominal CT showed a well-circumscribed cystic lesion measuring 5 cm in the pancreatic body. The cystic content showed a slightly high intensity as compared to the pancreatic parenchyma in the unenhanced phase, and the cystic wall and intracystic projection showed contrast enhancement (Figure 2). MR images also demonstrated a well-circumscribed cystic lesion and the content showed high signal intensity on a T1-weighted image (Figure 3), and low signal intensity on a T2-weighted image. The intracystic projection appeared the same as that seen on US and CT images. ERCP demonstrated that the cystic



Figure 1. Abdominal ultrasonography image demonstrated a cystic lesion in the pancreatic body. There was a small projection in the lumen.

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Key words Cysts; Positron-Emission Tomography; Projection

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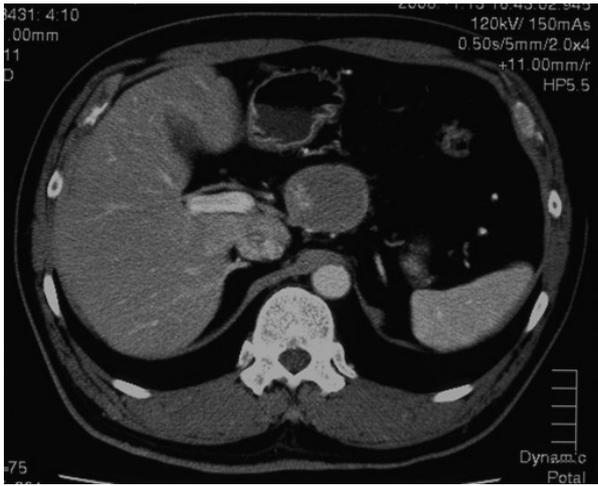


Figure 2. Abdominal axial CT image showing a well-circumscribed cystic lesion of 5 cm in the pancreatic body. The cystic wall and an intracystic projection showed contrast enhancement.

lesion was not connected to the pancreatic duct. With these findings, the cystic lesion was suspected of being a cystic neoplasm of the pancreas, but we could not definitively confirm whether the lesion was malignant. The intracystic projection was positively visualized on the PET image (SUV max: 5.8) (Figure 4). A distal pancreatectomy was performed because a pancreatic neoplasm could not be excluded. On gross inspection, the cystic lesion adhered to the pancreatic parenchyma. The surface of the cyst was smooth, the wall of the cyst was not thin and the lesion was filled with yellowish pasty material. A papillary projection measuring 2 cm was noted in the lumen (Figure 5). Microscopically, the cystic wall was lined with squamous epithelium surrounded by a layer of lymphoid tissue. At the papillary projection, there were no atypical cells and the structure was same as the other areas of the cystic wall (Figure 6). The postoperative course was uneventful and the patient was discharged from hospital on the 10th postoperative day.

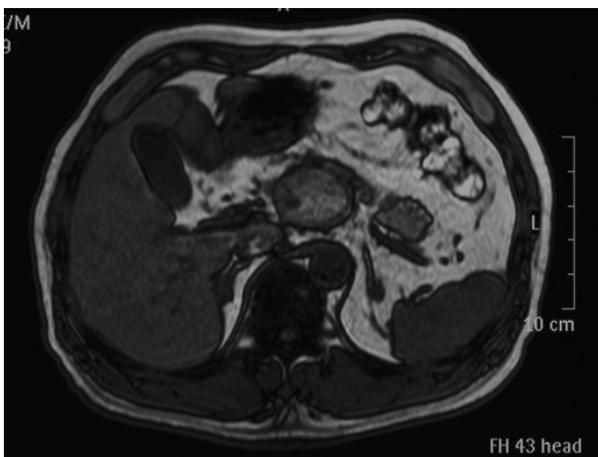


Figure 3. Axial T1-weighted magnetic resonance image of the abdomen showed a well-circumscribed cystic lesion, 5.0x4.5x4.0 cm in size. The content showed high signal intensity on T1-weighted image.

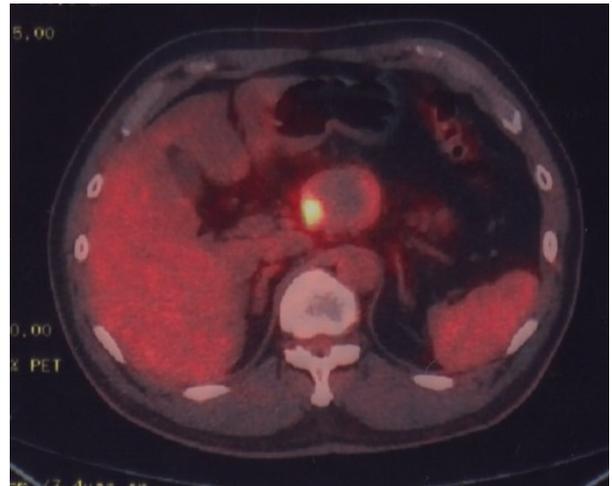


Figure 4. Positron emission tomography images using F-18 fluorodeoxyglucose: The intracystic projection was positively visualized (SUVmax: 5.8).

DISCUSSION

A lymphoid epithelial cyst is a true cyst, a benign lesion, representing approximately 0.5% of all pancreatic cystic lesions [3]. A Lymphoid epithelial cyst is typically found in middle-aged to elderly men. Almost all patients are asymptomatic and A lymphoid epithelial cyst is found incidentally during abdominal US or CT. Histologically, lymphoid epithelial cysts are lined by squamous epithelium without atypia. The wall of the cyst contains lymphocytes usually accompanied by germinal centers. Concerning the pathogenesis of lymphoid epithelial cysts, three hypotheses have been proposed. One is epithelial inclusion or ectopic pancreatic tissue in the peripancreatic lymph node, another one is squamous dysplasia caused by obstruction and dilatation of the pancreatic duct and the last is branchial cleft remnants misplaced in the pancreas during embryogenesis, but A definitive theory is still lacking.

Distinguishing lymphoid epithelial cysts from other pancreatic cystic neoplasms is often difficult because

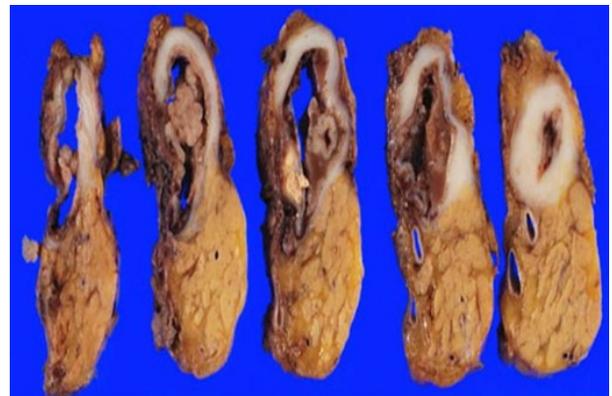


Figure 5. Surgical specimen of the pancreas. The cystic lesion adhered to the pancreatic parenchyma. The surface of the cyst was smooth and a papillary projection measuring 2 cm was noted in the lumen.

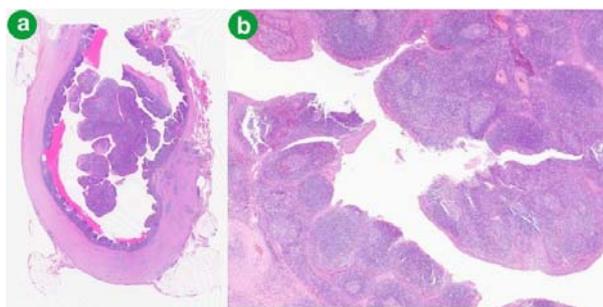


Figure 6. Histopathological finding of the cyst. **a.** Squamous epithelium surrounded by a layer of lymphoid tissue was seen in the cystic wall. **b.** The same structure was seen in the papillary projection (H&E stain x40).

the clinical incidence of lymphoid epithelial cysts is rare and radiological findings of lymphoid epithelial cysts are similar to other cystic pancreatic tumors. Neyman *et al.* suggested that the CT findings of lymphoid epithelial cysts described in the literature are quite varied [2]. A lymphoid epithelial cyst is usually multicystic and is less commonly unilocular; the appearance of an intracystic solid component of a lymphoid epithelial cyst as in our case is relatively rare [4, 5, 6]. In our case, the FDG-PET study positively visualized the papillary projection. We performed surgery based on this finding, but we could not determine why only the papillary projection was positively visualized. There were no atypical cells detected histopathologically in the papillary projection. Recently, FNA studies for the diagnosis of lymphoid epithelial cysts have been reported. Some reports have suggested FNA to be useful for diagnosis and helpful in avoiding unnecessary surgery [7, 8, 9]. Jian *et al.* suggested that FNA studies may help in rendering a correct diagnosis and reducing unnecessary surgery [10]. On the contrary, Zielinska-Pajak *et al.* [11] have suggested that FNA may be useful for the differential diagnosis but the cytologic picture may not be specific for lymphoid epithelial cysts. Histopathological examination is probably the only method of establishing a reliable diagnosis [11]. Some reports have suggested that, if the preoperative examinations suggest that a pancreatic cyst is a neoplastic lesion, FNA should be considered in order to avoid the risk of tumor dissemination [12, 13, 14, 15]. Kobayashi *et al.* have reported a case of a lymphoid epithelial cyst diagnosed with FNA. They performed FNA under laparoscopy and then performed enucleation [16]. In the future, if a lymphoid epithelial cyst is suspected, minimal resection should be considered with intraoperative pathological confirmation.

In conclusion, a lymphoid epithelial cyst is a rare pancreatic cystic lesion; the differential diagnosis was difficult in our case because the lesion included an intracystic papillary projection and the projection stained positively on FDG-PET. However, resection

was necessary because the lesion was symptomatic in our case.

Conflict of interest The authors have no potential conflicts of interest

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