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Lymphoepithelial cyst in the pancreas: A case report

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Summary: A case of lymphoepithelial cyst in the pancreas was reported. A 64-year-old man without any specific compalints was found to have a cystic lesion in the anterior portion of the pancreas, as revealed by ultrasonography of the abdomen at an annual medical examination in 1988. This was dissected easily from the pancreas. Histologically, it was diagnosed to be a benign lymphoepithelial cyst in the pancreas. Cysts of this type are rare, and their histogenesis is also not well understood. *Gastroenterol Jpn 1990;25:758–761*

Key words: *lymphoepithelial cyst; pancreas cyst*

Introduction

Lymphoepithelial cysts are occasionally observed in the lateral cervical region or parotid gland, and are called "branchial cleft cysts". However, a pancreatic involvement, such as the present case, is very rare.

Case Report

A 64-year-old man had no chief complaint and no history of alcoholism or any blunt abdominal traumas. His family history was unremarkable.

In October 1988, in an annual medical examination, an abdominal ultrasonography revealed a $3 \times 2 \times 2$ cm cystic lesion on the anterior surface of the pancreas. He was immediately admitted to our hospital for surgery. He had been healthy for the past five years.

Physical findings: Height 158 cm, weight 59 kg, well nourished. Blood pressure 142/90 mmHg, heart rate 72/min., regular. Physical examination excluded organomegaly, abdominal masses and abdominal tenderness.

Laboratory data on admission: WBC 5700 /mm^3, serum amylase 106 IU/l (normal range: 61 to 200), Ca 9.6 mg/dl, blood suger 85 mg/dl, CEA 1.3 ng/ml (less than 2.5). However, there was elevation of CA 19-9 to 70.0 U/ml (less than 37) (**Table 1**).

Abdominal ultrasonography: A slightly hypoechoic mass was located in the anterior and upper border of the pancreas. The mass was measured about 3 cm in diameter accompanied by some hyperechoic spots. Posterior echo enhancement was not clear (**Fig. 1**).

Endoscopic ultrasonography: A posterior echo enhancement and a lateral shadow echo of the mass were definite, suggesting a cystic mass, in spite of a "solid" internal echo (**Fig. 2**).

CT: An abdominal CT scan showed a superficial mass in the anterior of the body of the pancreas. Its density was identical to that of the pancreatic parenchyma. Enhancement was positive with the mass displayed in good contrast to the left renal cyst. The cyst of the pancreas was not enhanced (**Fig. 3**).

Other examinations: On MRI, the lesion was hyperintense on T_2-weighted images. ERCP showed no abnormalities in either the pancreatic

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Table 1 Laboratory data on admission

Blood chemistry		Peripheral blood	
T-Bil	0.5 mg/dl	WBC	5700 /mm^3
GOT	19 IU/l	RBC	432 × 10^4 /mm^3
GPT	11 IU/l	Hb	14.5 g/dl
Al-p	106 IU/l	Ht	44.2 %
LAP	35 IU/l	Plt	30.5 × 10^4 / mm^3
γ-GTP	21 IU/l		
ChE	0.83 ΔpH	Urinalysis	
LDH	234 IU/l	pH	5.4
TTT	1.2 KU	Prot	(−)
ZTT	3.5 KU	Glu	(−)
Na	140 mEq/l		
K	4.3 mEq/l	Stool	
Cl	107 mEq/l	Occult blood	(−)
Ca	9.6 mg/dl		
BUN	10 mg/l	Tumor marker	
Cr	0.9 mg/l	CEA	1.3 ng/ml
Total Protein	6.7 g/dl	AFP	4.2 ng/ml
Total Chol	142 mg/dl	CA19-9	70.0 U/ml
Glucose	85 mg/dl		
Amylase	106 IU/l		

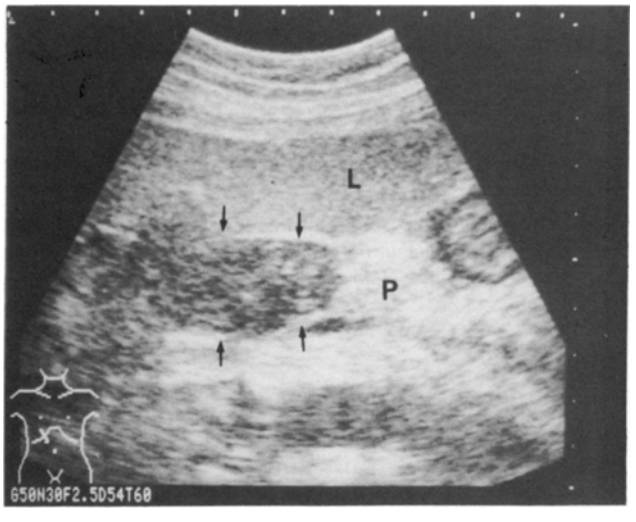

Fig. 1 Abdominal ultrasonography demonstrated a slightly hypoechoic mass with clear margin attached to the pancreas (arrows). The internal echo was solid and relatively homogenous. (L: liver, P: pancreas)

duct or extrahepatic biliary tract. An upper GI series did not show the inverted sign or ballooning of the C-loop. An abdominal angiography showed no abnormalities. Our preoperative diagnosis was thus a pancreatic cyst.

Operative findings: At laparotomy, a well-circumscribed mass was seen located over the upper

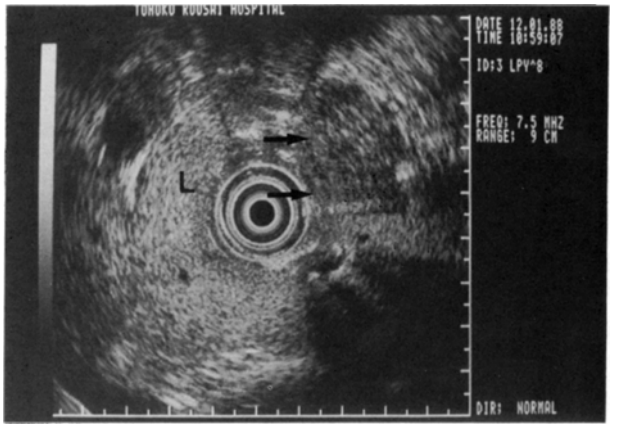

Fig. 2 Endoscopic ultrasonography. The mass accompanied with posterior echo enhancement and lateral shadow (arrows). (L: liver)

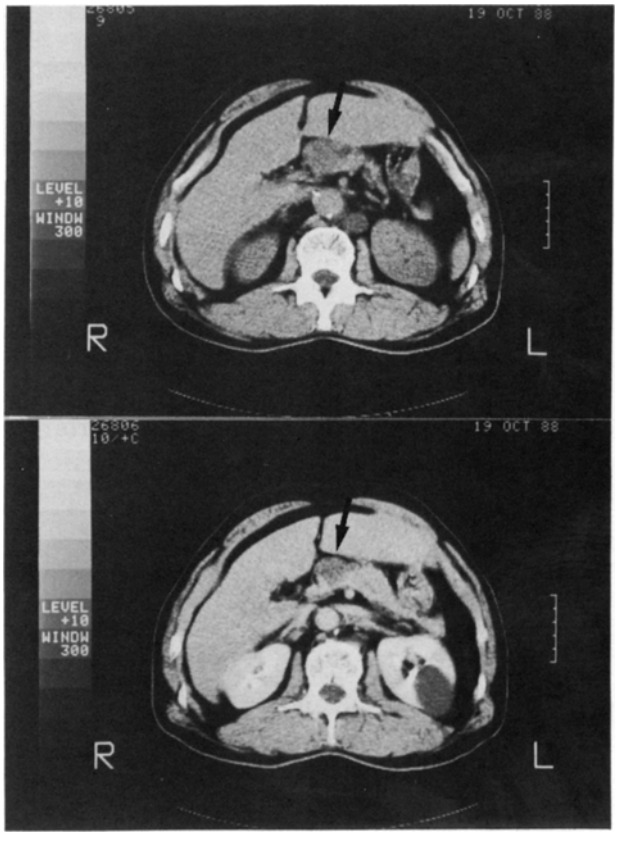

Fig. 3 a: Plain CT scan of the abdomen showed a round low density area protruding from the surface of the pancreas (arrow).
b: It was not enhanced by contrast medium.

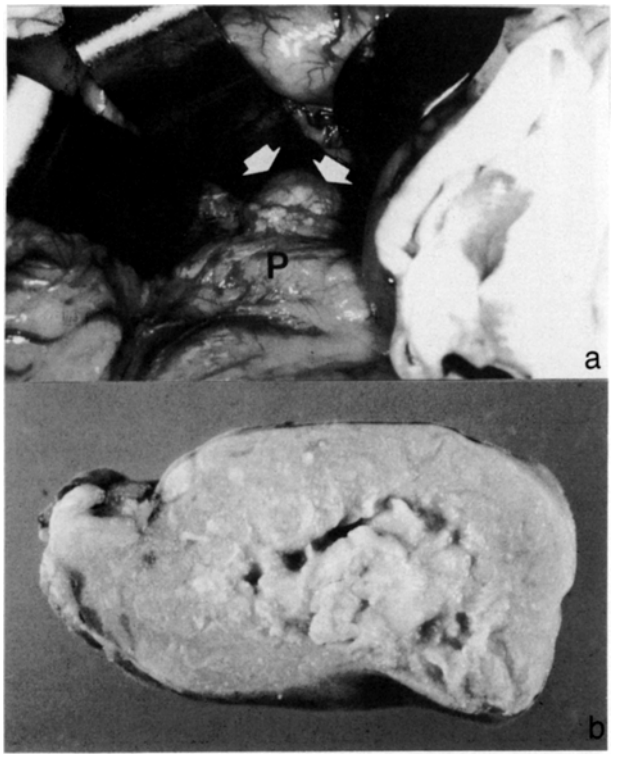

Fig. 4 a: Cyst (arrow) grew out from the anterior and upper border of the pancreas. (P: pancreas)
b: Cut surface of the cyst. Cyst contained brownish and white atheromatous substance. It looked like an epidermoid cyst.

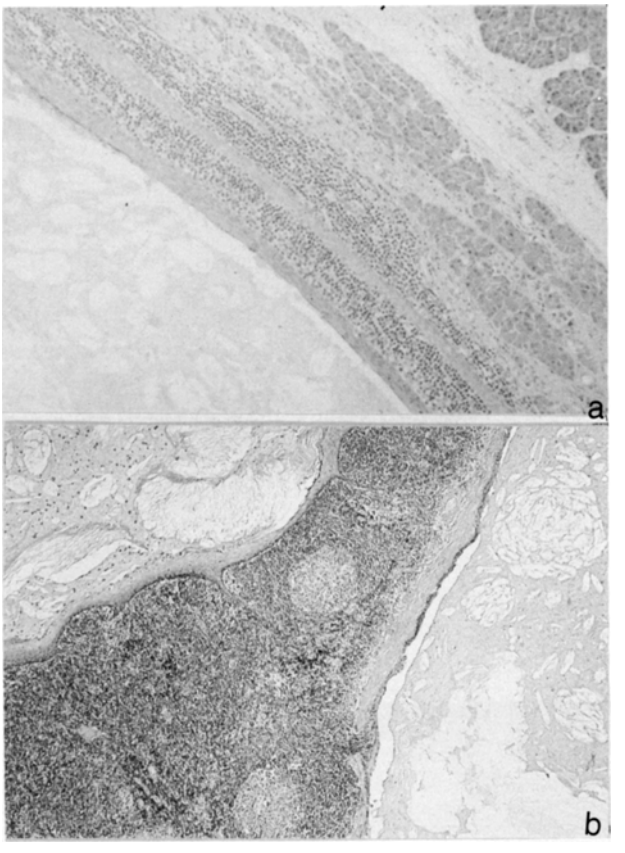

Fig. 5 a, b: Histological finding of the cyst. The cyst was lined by keratinizing squamous epithelim and filled with keratinous substance. And it was surrounded by lymphoid tissue containing germinal centers. The adjacent pancreas was atrophic due to compression by the cyst. (HE, a: ×80, b: ×32)

portion of the body of the pancreas. It protruded onto the surface of the common hepatic artery as a continuum of the organ. It was easily dissected along with small amounts of the organ. The cyst measured 4 × 3 × 3 cm and the outer surface was smooth and brownish in color. Its consistency was elastic-soft. There were no palpable lymphadenopathies (**Fig. 4a**).

Macroscopic findings: The cyst was covered by a very thin membrane and was filled with brownish and white atheromatous substance (**Fig. 4b**).

Pathological findings: Histologically, the cyst was multilocular. Its wall was lined by mature squamous epithelium which was accompanied by keratinization. It contained lymphoid tissue intermingled with foci of pancreatic tissue proper. These pancreatic foci were atrophic and fibrotic, presumably as a result of compression by the cyst. The histopathological diagnosis was a benign lymphoepithelial cyst in the pancreas (**Fig. 5**).

Postoperative course: The postoperative course was uneventful, and the patient remains well and free from recurrence as of working. CA 19-9 was still as high as 80.0 U/ml even 2 months after removal of the cyst. However, it was concluded there was no relationship between the two.

Discussion

The report describes a case of pancreatic cyst discovered incidentally during a mass health survey. In spite of the detailed classification of the pancreatic cysts by Howard and Jordan (1960)[1], the lymphoepithelial cyst, as reported here, is not included in their classification.

A search revealed only four cases of lymphoepithelial cysts associated with the pancreas previously reported. Takahashi et al. reported a case of lymphoepithelial cyst protruding onto the body of the pancreas. The wall of the cyst was composed of squamous epithelium intermingled with lymphoid follicules that had germinal centers. It contained mucinous and keratinized substance. Furthermore, they noted that there were no reports of this type of cyst arising in the abdominal cavity. Nishimura et al. also encountered a similar case. They described it as a "cystic benign lymphoepithelial tissue," according to the comments by Armed Forces Institute of Pathology. These two cases were not reported in English, however, two similar cases were reported in detail by Luchtrath et al.[2] and Truong et al.[3]. All of four reported patients were men aged from 35 to 58 years.

Lymphoepithelial cysts often arise at the lateral neck areas, and one called lateral cervical cysts. The cysts rarely involve the parotid and submaxillary glands. Their histogenesis is not fully understood. Those arising at the lateral neck regions are related to the salivary glands. The commonly accepted "Branchial Remnant Theory"[4] hypothesises that the 2nd branchial arch rapidly grows over the 5th arch, so that the 2nd branchial cleft is covered to form a cyst. According to the "Epithelial Inclusion Theory"[5] a lymphoepithelial cyst may arise from a lymph node including a misplaced epithelium. Furthermore it is suspected that this epithelium arises in the salivary gland[6]. Truong et al.[3] proposed the possibility that the same arises from a benign epithelial inclusion or ectopic pancreas in peripancreatic lymph node and subsequently encroaches upon the pancreatic tissue. Luchtrath et al.[2] suspected that lymphoepithelial cyst arises from a portion of the branchial cleft that has been misplaced and fused with the pancreas during embryogenesis.

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